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Titre :

« PARTENARIAT PATIENT DANS UNE DEMARCHE D'AMELIORATION DE LA QUALITE DES SOINS : L'EXPERIENCE DU PROGRAMME QUALITE EN MUCOVISCIDOSE »

VOLUME 1

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« Ce n'est pas parce que les choses sont difficiles que nous n'osons pas, c'est parce que nous n'osons pas qu'elles sont difficiles ».

Sénèque, 4 av. J.-C. – 65

« L'essentiel n'est pas ce qu'on a fait à l'homme, mais ce qu'il a fait de ce qu'on a fait de lui ».

Sartre, 1979

« L'histoire de mon enfance m'avait orienté vers le choix de la psychiatrie, ou plutôt vers l'idée que je me faisais de cette discipline. Je crois qu'il en est de même pour tout choix théorique. Les abstractions ne sont pas coupées du réel, elles donnent une forme verbale à notre goût du monde. La cohérence théorique nous rassure en nous donnant une vision claire et une conduite à tenir. Mais une autre histoire de vie aurait donné cohérence à une autre théorie ».

Cyrulnik, Les âmes blessées, 2014

Partenariat patient dans une démarche d'amélioration de la qualité des soins : l'expérience du programme qualité en mucoviscidose

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Article II Pougheon Bertrand D, Rault G. Ethique du programme qualité de la coopération des parents et des soignants : parent et coordinatrice du programme PHARE-M d'amélioration des soins dans la mucoviscidose. Archives de Pédiatrie 2015;22(HS2):91-2

Article III Pougheon Bertrand D, David V, Lombrail P, Rault G. Introduction of a Collaborative Quality Improvement Program in the French Cystic Fibrosis Network: the PHARE-M initiative. OJRD 2017; à paraître.

Article IV Pougheon Bertrand D, Nowak E, Dehillotte C, Lemmonier L, Rault G. Quality of care in cystic fibrosis: assessment protocol of the French QIP PHARE-M. OJRD 2017; à paraître.

Article V D Pougheon Bertrand, G Minguet, R Gagnayre, P Lombrail. Lessons from patient and parent involvement in the Quality Improvement Program in Cystic Fibrosis care in France. OJRD 2017; à paraître.

PUBLICATIONS COMPLEMENTAIRES

Article 6 Rault G, Pougheon Bertrand D. Programme d'accompagnement des patients et des familles par des pairs. Revue des maladies respiratoires. Actualités 2013 ;5(8) :713-4

Article 7 Stern M, Pougheon Bertrand D, Bignamini E, Corey M, Dembski B, Goss CH, Pressler T, Rault G, Viviani L, Elborn JE, Castellani C. European Cystic Fibrosis Society Standards of Care : Quality Management. J Cyst Fibros 2014;13 Suppl 1:S43-59

Article 8 Rault G, Pougheon D, Moisdon JC, Pepin F, Bellon G et al. Analyse de la prise en charge hospitalière ambulatoire de la mucoviscidose. Santé Publique 2015 ;27(3):363-72

COMMUNICATIONS ORALES LORS DES CONFERENCES

2015, Tours : Société Française de Pédiatrie – 28 Mai 2015 – TR 18-3 : Ethique de la collaboration Parents-Soignants - Communication orale : « Des soins à l'amélioration de la qualité des soins »

2016, Lille : Journées Francophones de la Mucoviscidose – Mars 2016 – Session4 : « PHARE-M, un programme collaboratif, pluridisciplinaire, partenaire, pour améliorer la qualité de la prise en charge des patients atteints de mucoviscidose »

2016, Genève : Congrès de la Société Européenne d'Education Thérapeutique (SETE;1079) – Communication orale : « Programme spécifique d'éducation thérapeutique pour les adolescents atteints de mucoviscidose : le lien avec le programme d'amélioration de la qualité et les indicateurs de santé »

2016, Bobigny : Journée annuelle du LEPS – Animation de l'Atelier : « Quelles collaborations des Patients dans les démarches qualité des soins »

COMMUNICATIONS AFFICHEES

Octobre 2012, Orlando, FL : North American Cystic Fibrosis Conference – Poster : « CF Quality improvement Program: a pilot phase to experiment the US QIP approach in France »

Mai 2014, Bobigny : Congrès de la Société Européenne d'Education Thérapeutique (SETE) – Poster : « Education thérapeutique et démarche qualité : Quelle synergie ? »

Avril 2015, Londres : BMJ Quality & Safety in Healthcare – Poster : « PHARE-M Performance Protocol »

Octobre 2015, Phoenix, AZ : North American Cystic Fibrosis Conference – Poster : « A study to assess effectiveness of the French QIP PHARE-M »

Octobre 2017, Indianapolis, Indiana : North American Cystic Fibrosis Conference – Poster : « Quality of care in cystic fibrosis: performance of the French QIP on patient health indicators »

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Toutes les équipes des centres de ressources et de compétences de la mucoviscidose qui se sont engagées dans le programme PHARE-M depuis son lancement en 2011 pour leur enthousiasme, les actions d'amélioration menées malgré les difficultés rencontrées et pour les résultats obtenus au bénéfice des patients.

Les parents et les patients qui ont accepté de s'impliquer dans la démarche qualité aux côtés de leur équipe soignante, en prenant le risque de ce changement de point de vue, pour améliorer les soins de tous les patients suivis dans leur CRCM.

Antoine et Thomas pour m'avoir encouragée et accompagnée tout au long de ce cheminement.

LEXIQUE & ABBREVIATIONS

ANAP
Agence Nationale pour l'Amélioration de la Performance

ANSM
Agence Nationale de Sécurité du Médicament

AQ
Amélioration de la qualité

ACQ
Amélioration continue de la qualité

ETP
Education thérapeutique du patient

CFF
Cystic Fibrosis Foundation (USA)

CRCM
Centre de ressources et de compétences pour la mucoviscidose

CRMR-M
Centre de référence maladie rare mucoviscidose

HAS
Haute Autorité de Santé

HCSP
Haut Conseil de Santé Publique

IHI
Institute for Healthcare Improvement (Harvard)

IOM
Institut de Médecine américain (Etats-Unis d'Amérique)

PHARE-M
Programme Hospitalier d'Amélioration des Résultats et de l'Expertise

SQUIRE
Standards for QUality Improvement Reporting Excellence

UTET
Unité Transversale d'Education Thérapeutique

GLOSSAIRE

La définition des mots ou expressions permettent d'éclairer le sens qui leur est donné dans la suite du document. Les références citées dans la définition permettent de situer leur origine et leur source.

Soins

Le mot « *soins* » est utilisé comme synonyme de l'expression « *prise en charge* », c'est-à-dire qu'il inclut l'ensemble des soins et services, y compris l'accompagnement psychologique ou social, réalisés par une équipe pluridisciplinaire auprès d'un patient, incluant des professionnels des centres hospitaliers ou des professionnels de santé intervenant au domicile des patients ou en ville auprès du patient.

Meilleures Pratiques

Perleth, Jakubowski et Busse proposent une définition du concept de meilleures pratiques dans le système de santé (best practice) : les activités, les disciplines et les méthodes pour identifier, mettre en œuvre et piloter les recommandations fondées sur la preuve dans le système de santé sont appelées « meilleures pratiques » ([1]). Il en résulte que ce concept est de nature organisationnelle et a été transposé de l'industrie, de façon comparable au concept de « benchmarking ». Il s'applique aussi bien aux activités liées aux soins qu'aux autres activités administratives, financières et d'organisation du système de soins. L'information nécessaire pour établir la preuve de meilleures pratiques relève aussi bien de la sécurité, l'efficacité, l'effectivité, le rapport coût-efficacité, la pertinence, le respect des valeurs sociales et éthiques. Un cadre d'évaluation a été proposé par les auteurs qui fait appel à 1) l'évaluation des technologies de santé, 2) la médecine fondée sur la preuve, 3) les recommandations de pratique clinique.

Démarche qualité collaborative

Caractéristique d'une démarche qualité qui associe plusieurs centres de soins ou plusieurs équipes soignantes dans un même cursus de formation-action ; ce type de démarche est aussi dénommé « *Learning and Leadership Collaborative* » par le Harvard Institute qui l'a développée dans le domaine de la formation d'équipes pluridisciplinaires à la méthode et aux outils d'une démarche qualité en santé.

Formation collaborative

Une formation est qualifiée de collaborative lorsqu'elle est conçue et délivrée à un public pluridisciplinaire, dont la vocation est de travailler en équipe. Ceci constitue la mise en œuvre du message clé de l'OMS « Apprendre ensemble pour travailler ensemble » dans son rapport de 1988. *Apprendre ensemble pour œuvrer ensemble au service de la santé. Rapport d'un groupe d'étude de l'OMS sur la formation pluri-professionnelle du personnel de santé : la formation en équipe*. Les enseignants d'une telle formation sont eux-mêmes de disciplines diverses et peuvent inclure des patients ou proches de patients.

Pratique collaborative (en Soins)

Cette expression désigne des pratiques professionnelles pluridisciplinaires, pouvant associer des professionnels d'établissements différents ou travaillant dans une relation ville-hôpital, et impliquant le patient dans une relation de partenaire pour ses propres soins, notamment à travers un processus de prise de décision partagé. Le *Guide d'implantation du partenariat de soins et de services – Vers une pratique collaborative optimale entre intervenants* ***et*** *avec le patient* publié par l'Université de Montréal (2014) en donne une définition précise et indique les compétences requises pour les professionnels et les conditions de la mise en œuvre de ce partenariat. La HAS a publié en 2014 sur son Webzine un numéro intitulé *Initiatives et Développement de pratiques collaboratives*, en soulignant que « *les pratiques collaboratives se développent si la formation est interprofessionnelle dès le départ* ».

https://www.has-sante.fr/portail/jcms/c_1721119/fr/les-pratiques-collaboratives-se-developpent-naturellement-si-la-formation-est-interprofessionnelle-des-le-depart

Soins centrés sur le patient

Cette expression est la traduction française de l'expression anglaise « *Patient Centred Care – PCC* » (2). La définition peut être résumée en *l'obligation professionnelle de prendre en charge et de répondre aux besoins du patient*. Le soin centré sur le patient est donc *défini par le patient mais délivré par le professionnel de santé*. Il inclut (3) : le respect du patient en tant qu'individu **;** l'écoute et la compréhension de ce que veut le patient, ses valeurs, ses objectifs vis-à-vis des traitements, le contexte et l'environnement dans lequel il vit ; la délivrance d'une information non biaisée et adaptée au contexte du patient et de la maladie ; l'empouvoirement du patient pour lui permettre de participer activement au processus de décision concernant sa santé ; l'accompagnement des patients pour leur permettre de fixer des objectifs atteignables et des stratégies leur permettant d'améliorer leur état de santé ; la continuité des soins incluant la revue régulière de l'avancement du patient vers ses objectifs de santé.

Activation du patient

L'activation du patient désigne « *les connaissances, les compétences et la confiance en soi d'une personne aux fins de la prise en charge de sa santé et de ses soins de santé.* » Dans le rapport publié en mai 2014 par le King's Fund, intitulé *Supporting people to manage their health: An introduction to patient activation,* Judith Hibbard et sa coauteure Helen Gilburt ont étudié les utilisations possibles de la mesure d'activation du patient (PAM), notamment dans des programmes visant à accroître l'activation du patient et dans l'affectation ciblée des ressources.

Activation du patient dans la démarche qualité

Nous avons utilisé cette expression, par extension, pour désigner la posture active du patient par rapport à la démarche qualité PHARE-M : sa compréhension de l'objectif de la démarche, de sa contribution personnelle dans l'amélioration de la qualité des soins, et sa volonté de prendre part à la démarche. Nous n'avons pas développé de mesure pour caractériser cette posture active. Nous avons caractérisé le processus par lequel le patient a pu être activé pour la démarche qualité et évalué sa mise en œuvre : l'information donnée périodiquement sur la maladie – la recherche, les traitements, l'information spécifique donnée sur PHARE-M, la procédure de recrutement explicitant les conditions et motifs de sa participation, l'information donnée pour le consentement à cette participation. Un document d'accompagnement de la démarche intitulé *Registre, Outil de la Qualité (ROQ*) a été conçu dans le cadre du PHARE-M pour activer les patients/parents. Dans notre exposé, l'activation du patient dans la démarche précède son empouvoirement (cf ci-dessous).

Empouvoirement du patient

L'OMS définit l'empouvoirement du patient comme « *un processus au cours duquel le patient acquiert une plus grande maîtrise sur les décisions et les actions qui concernent sa santé* » (4). Quatre éléments fondamentaux sont nécessaires pour mettre en œuvre le processus d'empouvoirement : *la compréhension par le patient de son rôle ; l'acquisition par le patient de connaissances suffisantes pour lui permettre de s'engager avec son médecin ; les compétences du patient ; un environnement facilitant.* L'empouvoirement du patient peut être développé par l'éducation thérapeutique du patient (Thèse I. Aujoulat, Université de Louvain, 2007), la co-construction d'un projet thérapeutique qui prend en compte ses objectifs, ses attentes et ses craintes, ou par les innovations technologiques de la e-santé auquel ce concept est fréquemment associé.

Empouvoirement du patient dans la démarche qualité

Nous avons utilisé cette expression dans le cadre de la démarche qualité PHARE-M pour caractériser « *un processus au cours duquel le patient acquiert une plus grande maîtrise de la démarche qualité pour lui permettre de participer, avec l'équipe pluridisciplinaire, au travail sur l'amélioration des processus de soin dans son centre* ». Quatre composantes ont été mises en œuvre et analysées pour permettre l'empouvoirement du patient dans la démarche qualité : *la compréhension par le patient (et par l'équipe) de son rôle dans le cadre de la démarche qualité ; la mise en place d'un environnement facilitant incluant le dédommagement de ses frais de participation à la démarche; sa formation à la démarche qualité ; l'organisation de sa participation aux réunions de l'équipe locale en toute transparence des données utiles à la qualité (sous réserve du respect de la confidentialité des données personnelles de santé des autres patients)*. Ainsi le « pouvoir d'agir » du patient provient à la fois de l'identification de son rôle de patient ressource pour l'amélioration de la qualité, dans le cadre d'une démarche qualité structurée, de l'acquisition de connaissances adaptées, et par la reconnaissance, de la part de l'organisation, des attributs associés à son rôle social (environnement facilitant).

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I- INTRODUCTION : L'IMPLICATION DES MALADES CHRONIQUES DANS L'AMELIORATION DE LA QUALITE DES SOINS

I-1. Le patient-usager au cœur de l'amélioration de la qualité des soins ?

Dans un document de travail publié en 2008, l'IRDES écrivait en introduction : « *Définir et évaluer la qualité des soins est une démarche fondamentale si l'on veut améliorer le système de santé. (…) Il n'existe pas à l'heure actuelle de système d'information permanent sur la qualité et la sécurité des soins, les données demeurent partielles, parfois contradictoires et difficilement accessibles.* »

L'Institut soulignait d'emblée le lien entre évaluation et information des patients : « *malgré une confiance très forte des Français en leur système de soins (86 % ont le sentiment qu'il est meilleur en France que dans d'autres pays), deux tiers d'entre eux pensent que la qualité des soins en France se détériore. Mais de quelles informations les patients disposent-ils en réalité pour juger de la qualité des soins qui leur sont administrés ?* ».

La définition de la qualité des soins la plus largement employée vient de l'Institute Of Medecine des Etats-Unis (IOM, 2001) qui précise que la qualité est « *la capacité des services de santé destinés aux individus et aux populations d'augmenter la probabilité d'atteindre les résultats de santé souhaités, en conformité avec les connaissances professionnelles du moment* » (5).

Cette définition apparemment simple a fait l'objet d'une caractérisation en six dimensions (IOM, 2001) : **l'efficacité clinique, la sécurité des soins, la délivrance de soins centrés sur le patient, dispensés au moment opportun, efficients et équitables.** Cette définition est centrée sur les ***résultats*** du système de soins. Toutefois les critères d'évaluation présentés ci-après rendent souvent compte de résultats caractérisant plusieurs dimensions à la fois.

L'**efficacité clinique** correspond à l'obtention des résultats des soins souhaitables, dispensés à ceux qui en ont besoin (PubMed Health). Il convient pour l'évaluer de disposer d'informations sur l'ensemble des ***résultats***, aussi bien les effets bénéfiques que les effets néfastes des soins, couramment appelés « *patient-relevant outcomes* ».

On suit le plus souvent les indicateurs de morbidité ou de mortalité par pathologie, par cause ou groupe de patients pour lesquels des soins efficaces peuvent améliorer le taux de survie, ainsi que des indicateurs relatifs aux résultats de la politique de prévention tels que le taux de dépistage, le taux d'immunisation et le taux d'incidence de maladies ou événements évitables.

Une autre série de données concerne l'adéquation de la ***structure*** des ressources et des équipements nécessaires aux différents aspects de la prise en charge. Elles sont collectées dans le cadre de la certification des établissements ou d'enquêtes spécifiques réalisées par groupes de pathologies ou par territoires de santé.

Une dernière série de mesures s'attache à l'évaluation de la ***qualité technique*** des soins fournis par les professionnels de santé, notamment leur *opportunité* (encore appelée *pertinence*) par rapport à des critères cliniques et des recommandations scientifiques. Ces indicateurs d'opportunité participent aussi de l'analyse de l'***efficience*** de la prise en charge, au regard de l'application des recommandations cliniques et des coûts induits. Des indicateurs de délai entre les étapes d'un processus peuvent être aussi déterminants pour ***l'accessibilité*** (et les résultats de

santé) et, déclinés par type de population, permettre d'obtenir une mesure de ***l'équité*** d'accès aux soins. Soulignons que pour les personnes éloignées du système, les indicateurs développés au sein du système de soin sont insuffisants pour rendre compte de l'accessibilité ou de l'équité, puisqu'ils concernent des personnes entrées dans ce système, et doivent être complétés par des mesures épidémiologiques de mortalité ou de morbidité en population générale, ainsi que par une analyse de la densité de l'offre de soin sur le territoire et du délai de prise de rendez-vous (Rapport de la DREES[*]).

Améliorer l'efficacité clinique est souvent considéré comme « trop technique » pour permettre des contributions des patients et l'utilité des enquêtes auprès des patients dans l'évaluation de la *qualité médicale* des soins reste discutée (6). La publication des indicateurs et leur comparaison entre diverses structures de soins n'est pas généralisée et peu ou pas accessible aux patients – hormis les classements publiés annuellement par certains médias grand public. Dans le cadre du Plan Cancer, le rapport « Les cancers en France », édité à partir de données robustes et partagées et permettant l'appropriation par le plus grand nombre des informations collectées, est une exception notable (7).

Concernant la **sécurité des soins** dispensés dans un établissement de santé, les informations sur les erreurs médicales ou les effets indésirables sont recueillies par le biais d'entretiens avec des patients ou d'enquêtes, et analysées pour identifier les problèmes à chaque étape du processus de soins (c'est le cas par exemple des enquêtes confidentielles sur la mortalité maternelle ou les accidents anesthésiques dans notre pays). Des systèmes de déclaration des événements indésirables sont mis en place, pour les professionnels et pour les patients (pharmacovigilance ANSM par exemple). Des études épidémiologiques complétées de questionnaires auprès des professionnels et des patients ont été menées en France entre 2004 et 2013 et en 2014 en Europe pour identifier les risques associés aux soins en établissement de santé et en soins primaires et évaluer l'efficacité de différentes stratégies d'amélioration de la qualité et de la sécurité des soins dans les établissements de santé (8). Ainsi, dans le domaine de la sécurité des soins, les patients sont reconnus comme étant capables de contribuer de manière substantielle en aidant à identifier les problèmes et les circonstances dans lesquelles ils se sont produits, dans le but de mettre en place des actions correctrices afin d'éviter qu'ils ne se reproduisent (9 ;10 ;11).

L'appréciation de **soins centrés sur le patient** revêt un caractère plus qualitatif. Ils ont été caractérisés (IOM, 2001) par : le respect des préférences du patient, l'information, l'explication du traitement, la coordination des soins, le support émotionnel, le confort physique, la participation de la famille, la continuité des soins incluant la gestion des transitions, et l'accessibilité aux soins. Une revue récente de la littérature a montré que les patients priorisent 10 critères de qualité des soins : la communication avec les soignants, l'accès aux soins, la prise de décision partagée, les compétences et les connaissances du soignant, l'environnement de soins, l'éducation du patient, le dossier électronique du patient, le contrôle de la douleur, l'organisation du processus de sortie, et les services de prévention (12).

[*] Rapport de la DREES, mai 2017. Déserts médicaux : comment les définir, comment les mesurer ?

La mesure usuelle de « soins centrés sur le patient » est la ***satisfaction*** du patient, relevée à l'issue d'une prise en charge ou d'un séjour hospitalier, et utilisée pour évaluer des interventions axées sur l'amélioration de l'une des dimensions des soins. Cette mesure de la satisfaction fait néanmoins l'objet de critiques quant à sa pertinence (13) et aux dérives qui pourraient découler de sa primauté sur les critères d'efficacité clinique ou d'optimisation des coûts de santé (14). Une approche par ***l'expérience patient*** le long du parcours de soins tend à se démarquer des enquêtes de satisfaction (15). Capter l'expérience patient implique d'enquêter sur ce qui est réellement advenu au patient au cours de son parcours de soin au regard de ce qui aurait « dû » se passer, et à quelle fréquence l'événement est advenu. Elle vise à évaluer à la fois l'efficience des soins (conformité aux recommandations cliniques) ET la réalité de « soins centrés sur le patient ». Toutefois, la mesure de l'expérience patient n'échappe pas non plus aux critiques (16).

La définition des différentes mesures de résultats utilisées et la technicité concernant le mode de collecte des données, leur analyse, leur interprétation contextualisée – c'est-à-dire l'indication des biais qui leur sont inhérents, et les règles de comparaison utilisées (entre établissements, zones géographiques ou groupes de pathologies), rendent difficile la compréhension et l'interprétation des résultats pour les soignants et pour les patients. La HAS, qui a pour mission de coordonner l'élaboration et d'assurer la diffusion d'une information adaptée sur la qualité des soins dans les établissements de santé à destination des usagers et de leurs représentants, mentionne dans le Guide méthodologique de diffusion publique des indicateurs de qualité des soins (17) : « *l'information en santé est d'un maniement complexe. Les indicateurs de qualité des soins portent en eux-mêmes cette complexité et peuvent engendrer des difficultés, voire des erreurs d'interprétation. L'usager ou le professionnel a besoin de connaître le sens d'un indicateur et comment l'utiliser ».* Le besoin de formation à l'interprétation de ces indicateurs s'impose aussi bien pour les professionnels que pour les patients impliqués dans la réorganisation de l'offre de soins.

Face à l'étendue du champ de la qualité des soins et à la technicité des critères d'évaluation, la question de l'information des usagers concernant la qualité des soins offerts par le système de santé peut sembler un défi et l'objectif de *placer les usagers au cœur de la réorganisation de l'offre de soins* « **une fiction utile** » (18).

I-2. Un cadre d'évaluation de la qualité des soins adapté à la prise en charge des maladies chroniques

Les enjeux de la qualité des soins des personnes atteintes de maladies chroniques ont été formalisés en 2010 par le HCSP (19) : un parcours de soins personnalisé et coordonné, la pluridisciplinarité ou inter-professionnalité des intervenants, l'éducation thérapeutique du patient (ETP), la continuité des soins, la prévention des complications, le support des services sociaux et des financements adaptés à cette prise en charge.

La question de l'évaluation de la qualité des soins s'est ainsi posée spécifiquement pour la prise en charge des patients atteints de maladies chroniques au regard de ces enjeux qui reflètent la complexité de la prise en charge, liée 1) à la durée

(associée à l'évolution et aux complications), 2) au champ des spécialités mobilisées et 3) à la répartition territoriale de l'offre de soins et de support.

Les composantes nécessaires à une prise en charge de qualité des malades chroniques ont été modélisées par E. Wagner en 2001 avec le ***Chronic Care Model*** (CCM) (20) selon 6 dimensions : l'existence d'objectifs de progrès continus de l'organisation de soins ; la pluridisciplinarité de l'équipe soignante ; le soutien à l'autogestion de ses soins par le patient (dont l'éducation du patient) ; un système d'information performant (dossier électronique du patient) ; l'utilisation des recommandations scientifiques (guidelines) et l'organisation de ressources dans la communauté de vie du patient. *Ce modèle conceptuel permet d'adopter une vision globale et fournit des lignes directrices cohérentes pour transformer le système et améliorer la prise en charge des maladies chroniques (25).*

Pour mesurer l'adéquation d'un process de soin au CCM, un cadre conceptuel a été développé caractérisant les activités, les sources des données et les protocoles afférents à ces activités (21). Des données provenant aussi des patients sont utilisées pour l'évaluation et l'amélioration des soins et services. L'utilisation de cette méthodologie en pre- et post-intervention d'amélioration permet ainsi d'évaluer les variations de l'implémentation du CCM dans les organisations concernées.

Coleman et Wagner ont réalisé une revue de la littérature en 2009 des interventions d'amélioration des soins s'appuyant sur une ou plusieurs caractéristiques du CCM (22). Les conclusions rapportent que le CCM est un modèle intégré qui peut guider efficacement le re-design des pratiques de l'organisation, conduisant à l'amélioration de la qualité des soins et de certains indicateurs de santé des patients. Le modèle doit toutefois évoluer pour mieux prendre en compte le critère de coût-efficacité de la prise en charge.

La déclinaison de ce modèle pour caractériser les particularités de la prise en charge de diverses maladies chroniques a été utilisée pour évaluer la qualité des soins ainsi que l'impact d'interventions visant à l'améliorer. Une revue systématique d'interventions visant l'implémentation du CCM dans des organisations de soins pour le diabète de type 2, a montré des résultats significatifs concernant l'amélioration des soins de première ligne et de certains indicateurs de santé des patients, le maximum d'impact étant observé lorsque les 6 composantes du CCM sont mobilisées (23).

Le CCM offre donc un cadre général de formalisation et une méthodologie pour l'évaluation de la qualité des prises en charge des pathologies chroniques et des interventions visant à les améliorer. Ce cadre, adapté aux enjeux de ces prises en charge, permet une évaluation du process et une mesure de son amélioration suite à une démarche d'AQ. Diverses publications ont montré que les résultats de l'évaluation par le CCM sont corrélés aux résultats sur certains indicateurs de santé des patients associant ainsi qualité du process et efficacité clinique.

I-3. Les malades chroniques, des usagers empouvoirés pour l'amélioration de la qualité des soins

D'après l'OMS, une maladie chronique est ***une maladie nécessitant des soins à long terme, pendant une période d'au moins plusieurs mois***. En France, il est admis qu'environ 15 millions de personnes seraient touchées par une ou plusieurs maladies chroniques (24). Ces chiffres se fondent notamment, mais pas exclusivement, sur le nombre de personnes inscrites au titre d'une Affection de Longue Durée (ALD). La classification par pathologie ne prend pas en compte les conséquences de ces maladies en termes d'incapacité et de difficultés personnelles qui ont un retentissement sur la qualité de vie des malades. L'importance des coûts de santé et des coûts sociaux liés à ces pathologies chroniques est soulignée de façon récurrente, au niveau international, et en France par le rapport charges et produits de l'assurance maladie.

La fréquentation régulière du système de soins, les échanges avec d'autres patients ou sur les réseaux sociaux, et diverses stratégies mises en œuvre pour développer leur autonomie pour leurs soins et la gestion de la maladie dans leur vie quotidienne, ont pour résultat des patients progressivement *empouvoirés*. Le CCM (ou modèle de soins de longue durée au Canada) vise, comme résultat « intermédiaire », la mise en place ***d'interactions productives*** entre une équipe soignante formée et proactive et un patient informé, actif et motivé pour améliorer les résultats cliniques et fonctionnels (25).

Les stratégies développées et évaluées pour empouvoirer le patient visent à l'informer, l'éduquer et l'impliquer dans sa propre prise en charge dans le but d'améliorer la qualité des soins auto-dispensés, l'observance des traitements, et la qualité de vie avec la maladie (26). L'OMS souligne que « *l'éducation thérapeutique du patient fait partie intégrante et de façon permanente de la prise en charge du patient* ». La loi HPST de 2011 a précisé les trois modalités opérationnelles de l'ETP : les programmes médicalisés qui font l'objet d'un programme personnalisé ; les programmes d'apprentissage ayant pour objet l'acquisition des gestes techniques et les actions d'accompagnement qui ont pour objet d'apporter un soutien et une assistance aux malades dans la prise en charge de leur maladie (27) et qui peuvent être dispensées par divers promoteurs y compris les associations agréées de patients. Les « actions d'accompagnement » proposent ainsi des possibilités très diversifiées, utilisant aussi les nouvelles technologies de communication.

Un nouveau ***modèle de soins partenaire*** s'est progressivement mis en place entre les patients et les soignants qui reconnaît le savoir expérientiel et les compétences du patient pour faire des choix éclairés pour ses soins (28 ; 29). Des processus formalisés tels que la ***prise de décision partagée*** (*shared decision making*) ont été développés pour soutenir l'engagement des patients dans le choix de leurs options de soins (30). Ce processus met l'accent sur l'information du patient – sur sa pathologie, les options de traitement possibles, leurs effets scientifiquement démontrés – et le respect de ses préférences.

Une conséquence majeure de ces évolutions n'a pas été appréhendée, semble-t-il

jusqu'ici : la capacité de participation d'un patient empouvoiré à l'amélioration de l'organisation des soins, au-delà de l'amélioration de la qualité pour ses propres soins.

La connaissance qu'ont les patients du système de soin est bien, de plus en plus, perçue comme importante par les instances qui déploient des dispositifs institutionnels pour associer les patients à l'amélioration du système de santé aux niveaux du mésosytème (Commission des Usagers dans les établissements) et du macrosystème (Institut pour la Démocratie en Santé, 2015 ; Projet « Participations des usagers et démocratie en santé », 2016). Dans les services de soins, divers moyens sont mis en œuvre par les professionnels de santé pour recueillir l'avis des patients : des enquêtes ou des interviews ou une participation organisée à des groupes de réflexion thématique leur permettent de recueillir leurs préférences et leurs attentes afin de réaliser une synthèse pour l'intégrer au projet développé par l'équipe[†]. La satisfaction ou les plaintes des patients sont régulièrement recueillies et traitées par les professionnels pour améliorer les soins. Des témoignages individuels (« story telling ») sont utilisés occasionnellement pour relater des défaillances du système et développer des points de sécurité.

Mais rares sont les expériences de **participation directe des patients à des projets d'AQ comme partenaires des soignants**, apportant leur connaissance expérientielle de l'organisation réelle de la prise en charge le long de leur parcours de soin en complément de la connaissance des professionnels. Le rôle de patients impliqués dans des instances ou des commissions dans l'établissement de soin ou au-delà, pour important qu'il puisse être dans une vision transversale de la qualité et de défense des droits des patients, ne peut traiter des questions liées à l'organisation détaillée d'une prise en charge par une équipe de première ligne. Or cette connaissance basée sur l'expérience patient du système de soin qu'il pratique durablement et régulièrement est unique et incontestablement riche pour l'amélioration de l'organisation collective des soins.

Le modèle de Montréal représente le continuum de l'engagement du patient dans ses propres soins (soins directs) et, en parallèle, l'engagement du patient dans l'amélioration des soins (organisation des services et gouvernance) (29). Dans ce schéma, le partenariat est caractérisé par la co-construction de services, de programmes et de démarches d'amélioration continue de la qualité des soins.

Toutefois, à notre connaissance, la ***participation de patients à une démarche d'AQ*** n'a pas été documentée et aucune publication en France ou à l'étranger ne rapporte une évaluation d'une telle expérience de participation aux côtés des équipes soignantes.

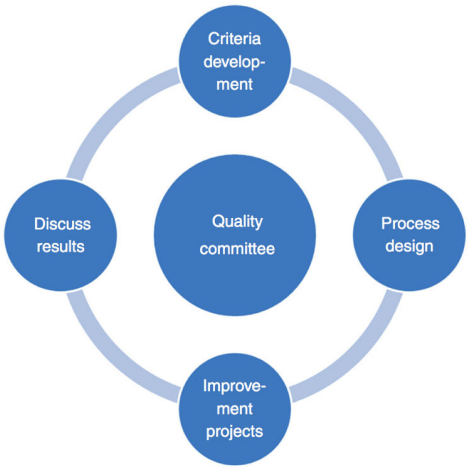
Ce mémoire propose ainsi une contribution à l'analyse de la contribution de patients à l'amélioration des soins, en présente les conditions et les effets, et identifie des facteurs de succès pour la participation d'un « patient partenaire dans une démarche d'amélioration de la qualité des soins ».

[†] Université d'été de l'Agence Nationale pour l'Amélioration de la Performance (ANAP), septembre 2017, « Comment innover au mieux pour adapter nos organisations aux enjeux de santé de demain ? ». Expérience de la réorganisation de l'hôpital de jour du Pôle Viscéral du CHU de Rouen, rapportée par l'équipe du Pr Pierre Michel (non publiée).

I-4. Co-design, action et évaluation… les trois mots clés pour l'implication des patients à tous les niveaux de l'amélioration de la prise en charge

Pour tenter de caractériser les diverses formes de participation des patients à la gestion de la qualité des soins, Groene et Sunol ont proposé le cadre conceptuel représenté sur la figure 1 (31).

Figure 1 : Cadre conceptuel de la participation des patients à la gestion de la qualité



L'examen de la littérature et une enquête transversale dans les hôpitaux dans le cadre du projet européen DUQuE leur ont en effet permis d'identifier des expériences de participation des patients à ces différentes ***activités de gestion de la qualité des soins*** (32) :

1) ***criteria development*** : le développement des recommandations scientifiques (guidelines), pour répondre aux besoins générés par les malades chroniques y compris sur des aspects de continuité des soins et de transition ;

2) ***process design*** : l'expression des préférences des patients dans la conception des processus de soins, grâce à des enquêtes ou des groupes de discussion, des observations directes ;

3) ***quality committee*** : l'évaluation de la qualité et la sécurité des soins lors de réunions formelles régulières au sein des instances d'établissement ;

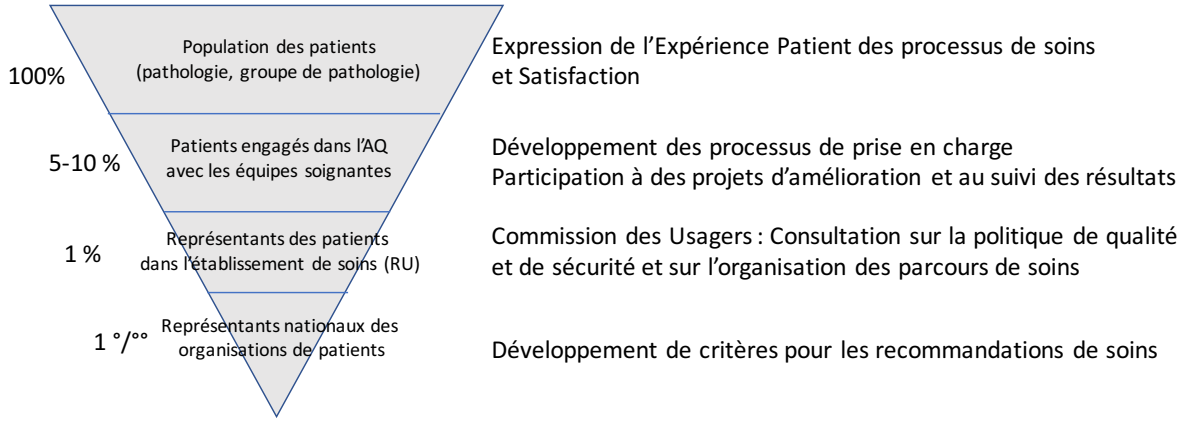
4) ***improvement projects*** : la planification et la mise en œuvre d'interventions d'amélioration de la qualité à travers une série de cycles plan-do-study-act (PDSA), dans le cadre d'un partenariat avec l'équipe chargée de la qualité ;

5) ***discuss results*** : plus rarement, l'évaluation des actions d'amélioration, en prenant part à la discussion des résultats.

Nous proposons d'articuler ce modèle avec les niveaux de représentation des acteurs, notamment les patients et leurs représentants, pour examiner les conditions de son opérationnalisation.

La figure 2 représente sur une pyramide inversée les niveaux d'intervention auxquels les patients pourraient exercer les activités répertoriées par Groene et Sunol pour contribuer à l'amélioration de la qualité des soins. L'objectif est d'articuler la nature de l'intervention, son champ d'application et la représentation des patients dans le système de soins.

Fig.2 : Niveaux d'intervention des patients pour l'amélioration de la qualité des soins



Nous faisons l'hypothèse que les différentes activités par lesquelles les patients peuvent contribuer à l'amélioration du système de soins devraient s'organiser de façon cohérente entre les différents niveaux du système (micro, méso, et macro), leur participation à des projets d'AQ se situant précisément au niveau du microsystème clinique, entre d'une part la représentation des usagers dans l'établissement de santé et d'autre part la collecte la plus large possible de l'expérience ou des préférences des patients dans la population concernée. L'expérience de la filière mucoviscidose rapportée dans la suite du document nous permettra d'illustrer cette hypothèse dans ce contexte particulier.

Plus précisément :

Le ***développement de critères*** pour les recommandations de soins (guidelines) étant à portée nationale (ou internationale) elle nécessite des connaissances sur les publications scientifiques et une expertise de la pathologie et de ses traitements. La contribution des patients est réalisée via leurs organisations nationales agréées, en les personnes de leurs délégués aux affaires médicales ou cliniques (quelques individus au plan national) et les sociétés savantes pour la pathologie. Leur contribution peut être majeure pour les recommandations de structure, d'organisation de la prise en charge, les conditions d'accueil des patients dans le système de soins, l'accessibilité aux soins et l'équité des soins.

A l'opposé, la réalité des processus de soin, pour la pathologie ou le groupe de pathologies, doit être basée sur la plus large ***expérience patient*** de façon à refléter le plus grand éventail de situations possibles et leur fréquence d'occurrence. Les enquêtes auprès de l'ensemble des patients atteints de la pathologie au plan national doivent être organisées en visant le plus grand nombre de répondants (100%). La définition de l'expérience patient du parcours de soin sera évoquée plus loin dans ce document, comme un moyen de nourrir la démarche qualité dans le microsystème clinique et une mesure d'évaluation de la qualité des soins ou des interventions visant à l'améliorer.

Au niveau du ***développement des processus de prise en charge***, la participation des patients à des projets d'amélioration des soins, requiert un « collectif » de patients volontaires qui s'impliquent sur la durée du projet, avec un équilibre entre une continuité de l'engagement et une possible rotation des personnes dans le temps : de 5 à 10% des patients suivis, impliqués pendant des durées allant de quelques semaines à plusieurs mois. C'est à ce niveau que les démarches d'AQ

peuvent associer les équipes de première ligne et les patients bénéficiaires de la prise en charge pour concevoir ensemble des améliorations et accompagner leur mise en œuvre.

Enfin, la participation de patients à ***des instances d'établissement*** telles que la Commission Des Usagers est prévue pour permettre une « *consultation sur la politique de qualité et de sécurité des soins et sur l'organisation des parcours de soins* » : elle répond à des règles de représentation par des patients proposés par les associations agréées (décret n°2016-726 du 1er juin 2016). Elle concerne quelques représentants de patients suivis dans l'établissement.

La participation des patients pourrait se concrétiser à ces quatre niveaux permettant de structurer leur contribution à l'amélioration de la qualité des soins. Cette participation serait donc articulée au sein des établissements hospitaliers et en lien avec le niveau national, selon les objectifs et les activités présentées dans le *modèle de Groene et Sunol*.

La thèse illustre la participation de patients au niveau du développement des processus de prise en charge d'une pathologie, la mucoviscidose, au sein de services spécialisés (centres de ressources et de compétences de la mucoviscidose – CRCM) et en lien avec les services référents et les intervenants au domicile du patient, conformément au parcours de soin de cette pathologie.

II- PROBLEMATIQUE : L'IMPLICATION DES MALADES CHRONIQUES DANS UNE DEMARCHE QUALITE COLLABORATIVE CENTREE SUR LE MICROSYSTEME CLINIQUE COMME SOURCE D'AMELIORATION DES SOINS

II-1. La nécessité d'une culture qualité pour les équipes soignantes

De nombreux articles ou communications récentes soulignent que les équipes de première ligne (Front Line) sont le lieu privilégié pour des actions conjointes d'amélioration de la sécurité et de la qualité des soins (33 ;34). Dans leur ouvrage de référence, Quality by Design (35), Nelson, Batalden et Godfrey rappellent que « ***chaque soignant a deux métiers : le premier est de faire son travail, le second est de l'améliorer*** ». Malheureusement, si le soignant est formé à « son travail de soignant », au cours de son cursus initial et en formation continue, rares sont les opportunités de formation à « l'amélioration de son travail ».

De nombreux freins culturels et organisationnels existent à l'amélioration de la qualité des soins par les équipes de première ligne. Tucker le montre dans l'article intitulé « Pourquoi l'hôpital n'apprend pas de ses erreurs » qui analyse le quotidien dans 9 hôpitaux et auprès de 26 infirmières dans des services d'hospitalisation variés (36). Elle a observé que des erreurs manifestes ou des problèmes dans l'application des procédures ne sont pas rares mais au contraire, font partie intégrante du travail des équipes de première ligne dans la délivrance des soins. Comme Mintzberg l'analyse, le fonctionnement des organisations hospitalières relève de la « bureaucratie professionnelle » dans laquelle la qualité est assurée par la standardisation des qualifications dans le respect de normes essentiellement externes à l'organisation, produites par les Sociétés Savantes ou les associations professionnelles, mais internalisées par les acteurs (37). Sur ce modèle, les hôpitaux ont misé sur des professionnels hautement qualifiés dans leur discipline pour pallier les faiblesses de l'organisation : « *Great doctors and nurses, not great organization or management* ». Les problèmes relevés concernent des transmissions d'informations incorrectes, des équipements manquants ou cassés, des ressources indisponibles, des fournitures manquantes ou inadaptées, des demandes simultanées contradictoires. Quant aux erreurs, elles concernent des actions incorrectes exécutées par les infirmières ou exécutées par d'autres personnels, et des tâches inutiles exécutées du fait de mauvais processus. L'analyse des causes a permis d'identifier des ***causes systémiques*** et non des causes liées à des compétences ou des individus en particulier (38). L'intérêt est ici de comprendre comment sont traités ces problèmes et pourquoi ils se reproduisent.

Toute réponse à un problème de non qualité dans le cours d'un soin doit s'organiser à deux niveaux : le premier niveau consiste à ***trouver un remède à court-terme*** qui permet de poursuivre le processus de soin pour le patient avec un minimum d'inconvénient pour lui, et le second niveau consiste à ***rechercher les causes du problème et à mettre en place des solutions à plus long terme*** pour éviter qu'il ne se répète.

L'analyse de Tucker met en évidence qu'un remède immédiat est généralement trouvé (par l'infirmière) pour résoudre le problème, quoiqu'il puisse générer des difficultés en cascade ailleurs dans l'organisation (rupture de fourniture répercutée dans un autre service suite à un dépannage « sauvage »). Mais paradoxalement, cette résolution immédiate masque le problème qui n'est que rarement exposé dans une réunion de service pour être traité au second niveau. Seuls remonteront des

problèmes qui ont entraîné de réels inconvénients pour le patient ou des disfonctionnements graves dans l'organisation.

Ceci témoigne d'une méconnaissance profonde de la culture de la qualité qui consiste à résoudre les difficultés dès qu'elles apparaissent et au moment où elles n'ont pas encore causé de désordre important dans l'organisation ou pour le patient.

Un autre bénéfice indirect plus insidieux de la résolution immédiate identifié par Tucker est la valorisation des personnes qui résolvent les problèmes sans en faire état et s'attirent ainsi la bienveillance des autres personnels et des louanges sur leurs compétences et leur autonomie. Ceci relève de la culture du héros si contraire à la ***culture de la qualité*** qui prône la constance et la cohérence : « *Do it always the same way rather than the way YOU think is the best* ». Ainsi, note A. Tucker, l'organisation semble fonctionner normalement, mais au prix d'un stress important des personnels de première ligne, contribuant au burn-out et à un turn-over majoré observé dans les établissements de soins, jusqu'à causer une pénurie d'infirmières aux Etats-Unis. Dans la culture de la qualité, le profil de l'employé « idéal » est au contraire le professionnel qui prête attention aux disfonctionnements avant qu'ils n'occasionnent de conséquences importantes, et les porte à la connaissance de l'équipe et du management pour trouver des solutions et veiller à leur application.

Les recherches ont permis d'identifier plusieurs leviers pour améliorer la qualité des soins délivrés par les équipes de première ligne et permettre à l'hôpital d'apprendre des erreurs et des problèmes.

Ces leviers consistent à développer une ***culture de la qualité*** dans les équipes de première ligne grâce à plusieurs actions concomitantes :

1) développer le ***leadership du/des manager(s)*** afin qu'il(s) organise(nt) les conditions d'une réflexion sur les pratiques de l'organisation en accordant l'importance nécessaire à la résolution de second niveau des problèmes et en encourageant les personnels à faire état des problèmes sans craindre d'être dévalorisés ou culpabilisés ;

2) mettre en œuvre au sein de l'équipe de soin pluridisciplinaire les ***méthodes et les outils de la qualité*** pour analyser les causes de problèmes, mobiliser les services transversaux lorsque la solution préconisée a un impact au-delà de l'équipe, mettre en place les changements selon une approche Plan-Do-Study-Act (PDSA) ;

3) mettre en place une ***démarche qualité continue***, basée sur des alertes produites en routine et un système de surveillance incluant les patients.

II-2. L'enjeu d'une démarche qualité centrée sur les microsystèmes cliniques

Diverses publications font état depuis la fin des années 90 de constats assez largement partagés qui limitent la délivrance de soins de qualité :

- Un fossé existe entre les connaissances scientifiques (guidelines ou recommandations scientifiques) et leur application dans les pratiques de soin ;
- Des variations importantes sont omniprésentes dans les pratiques de soin, qui ne semblent pas majoritairement liées au patient (qui les constate à l'occasion d'un changement d'établissement ou même de médecin dans le même établissement) et dont l'origine n'est pas clairement établie : sont-elles dues à une absence de consensus scientifique, ou à un manque de consensus entre

les cliniciens ou encore à un manque d'adhésion des cliniciens à ces consensus ?
- Des exemples de « meilleures pratiques » (best practice) existent dans de nombreux domaines des soins, mais ne sont ni décrits ni diffusés dans la communauté soignante ;
- Le cloisonnement entre les disciplines et l'animosité fréquente entre les cliniciens et l'administration hospitalière est source d'incompréhension et d'une contribution minorée de chacun à l'optimisation du système ;

Ces constats ne pointent pas la responsabilité d'un individu mais d'un système d'organisation. Est-il besoin de rappeler que « *aucun soignant n'est à lui seul responsable des résultats de santé du patient* » (35) ? ***Le système et les processus produisent les résultats, non les individus***. Pour améliorer les résultats, il est donc incontournable de reconfigurer le système de soins.

Parallèlement, une ***science de la conduite du changement*** dans les organisations, élaborée initialement dans le secteur industriel, a été transposée au domaine de la santé (39). Cette science se fonde sur trois piliers : l'analyse statistique des événements survenus (défauts et variations), les techniques de mise en place des changements avec des cycles de test/évaluation/ajustement (PDSA) avant standardisation, et les approches motivationnelles et la dynamique de groupe pour obtenir l'adhésion des individus au changement.

Dès 1998, Kilo décrivait les objectifs et la démarche des ***Breakthrough Series*** (BTS), cycles de formation-action collaboratifs destinés aux équipes soignantes et à leurs leaders, animés par les experts du Harvard Institute for Healthcare Improvement (40). Le premier objectif des BTS est d'obtenir des ***améliorations rapides***, dès le cycle de formation-action de 9 à 12 mois, mesurables et soutenables dans le temps, afin de construire la capacité de l'organisation à accomplir des changements dans ses pratiques. Le deuxième objectif est de rester ***centré sur un objectif clinique***, et de considérer l'apprentissage de la méthodologie de la qualité comme un outil au service de l'objectif à atteindre. Ainsi, l'équipe acquiert la conviction que la démarche d'amélioration fait partie intégrante de son travail et n'est pas une démarche « à côté » ou « en plus » de son travail de soignant. Enfin, le troisième objectif du IHI lors de l'animation des sessions collaboratives est de proposer des ***listes d'idées de changement*** qui apportent un effet levier maximum pour l'amélioration des soins. En effet, des centaines d'idées de changement peuvent être testées, mais celles qui permettent de mettre en pratique les connaissances issues des ***recommandations ou des meilleures pratiques de soin*** sont plus à même d'apporter des résultats sur la qualité des soins rapidement.

L'aspect ***collaboratif*** de cette démarche, au sens de la définition présentée dans les préliminaires de ce mémoire, est justifié par la volonté d'accélérer l'amélioration de la qualité des soins en faisant participer simultanément de multiples organisations (20 à 40 par cycle), et de créer une ***communauté apprenante*** au cours du cycle d'un an et au-delà, favorisant les échanges de bonnes pratiques et stimulant les efforts, pour maintenir une amélioration continue de la qualité. La conception, l'organisation et la mise en œuvre de la démarche BTS reposent aussi sur ***l'expertise et la mobilisation du IHI*** (et d'autres instituts universitaires aux Etats-Unis et en Europe) auprès des établissements de santé.

L'évaluation et la recherche sont d'emblée associées au déploiement de ces démarches qualité collaboratives, en vue d'établir leur impact dans le système de

santé et en analysant les barrières et les facteurs de succès à leur introduction (41, 42, 43). La recherche, d'abord essentiellement quantitative, cherchant à évaluer l'impact sur les résultats de santé des patients ou sur des indicateurs de performance de la prise en charge (délais, réadmissions après une sortie d'hospitalisation, mise en place de plans de soins pour les patients…) a progressivement développée une méthodologie d'***évaluation réaliste*** de ces ***interventions complexes*** afin d'identifier les ***mécanismes*** en œuvre dans différents ***contextes*** d'établissement ou d'organisation qui sont associés à une variation de leur impact (44,45,46,47). L'approche réaliste, plus largement, vise à comprendre ce qui fonctionne, pour qui et selon quelles circonstances (contexte) (48). Un débat méthodologique est engagé concernant la nécessaire distinction entre l'évaluation du ***process d'implémentation*** d'une intervention complexe et l'évaluation de ***l'impact de cette intervention complexe*** (telle qu'une démarche qualité) (49,50,51,52).

L'enjeu d'une démarche qualité dans le microsystème clinique est donc un enjeu ***organisationnel*** visant l'amélioration d'un ***objectif clinique,*** grâce notamment à l'application des recommandations de soin (guidelines) et la diffusion des meilleures pratiques (au sens de la définition présentée dans les préliminaires) et la réduction des variations non souhaitées (non dépendantes de la situation du patient).

Cet enjeu est porté par l'équipe soignante emmenée par ses ***leaders***, selon les sujets traités. La mise en œuvre de changements dans les pratiques est conçue comme faisant partie intégrante du rôle de l'équipe – et non pas procédant de décisions imposées de l'extérieur – en vue de maintenir une ***démarche qualité continue*** intégrée dans le fonctionnement permanent. L'accélération de la diffusion de la culture qualité repose sur le caractère ***collaboratif*** de la démarche.

La ***recherche internationale*** sur l'implémentation et l'impact de ces démarches qualité (interventions complexes), les freins et les facteurs de succès ainsi que les mécanismes en jeu dans différents contextes d'organisation est consubstantielle du développement de ce type de démarche qualité.

II-3. Les malades chroniques partenaires de leur équipe soignante dans le microsystème clinique

L'OMS indique en préambule de son rapport sur l'état de la santé du monde (2008) : « *parce qu'elle repose sur un riche partenariat et une vision d'ensemble du système, la pensée systémique offre la possibilité, encore inexploitée, de mettre au point et d'évaluer des interventions destinées à renforcer les systèmes »* (53).

La représentation systémique du système de soin permet d'inscrire le patient chronique et son entourage au cœur du microsystème clinique. Le niveau du méso-système inclut les services de l'hôpital support du microsystème clinique auprès desquels des marges de manœuvre et des changements devront être négociés tout en maintenant la cohérence du fonctionnement général, le macro-système représentant le niveau où sont définies les règles régissant le système de soin mais aussi les recommandations scientifiques qui président à la prise en charge des malades (35).

De même que le développement des programmes et outils d'ETP ne peut aujourd'hui se concevoir sans la participation des patients – pour l'expression des besoins, la conception des référentiels et des outils – le développement de programmes d'amélioration de la qualité dans le microsystème clinique ne saurait ainsi se priver

de l'apport des patients, notamment des patients atteints de maladie chronique, aux différentes étapes de la démarche qualité.

Citons pour l'exemple, les processus de soin – le circuit de la consultation pluridisciplinaire, l'articulation entre les examens et les interventions des professionnels des différentes disciplines, l'élaboration des projets de soins suite aux bilans annuels, la relation entre le service ambulatoire et l'hospitalisation conventionnelle ou l'arrivée aux Urgences, l'organisation des soins à domicile, la coordination des demandes entre deux venues à l'hôpital – qui sont autant de composantes dont le patient chronique a acquis l'expérience et sur lesquels il peut apporter une vision constructive. Le partage de cette expérience avec les soignants dans des groupes de travail pluri-professionnels aurait pour objectif l'identification des problèmes selon la vision du patient et obligerait à la recherche de solutions avec les différentes parties concernées. L'objectif d'amélioration devrait répondre au besoin du patient malgré les contraintes éventuelles des services, qui ne peuvent justifier une inefficacité de la prise en charge. En cas de blocage, des arbitrages par le méso-système deviennent incontournables. A l'échelon du mésosytème, les commissions d'établissement pourraient être saisies par le RU de ces arbitrages afin que chaque niveau de responsabilité puisse contribuer à la recherche de solutions.

Comme le propose la pyramide inversée de participation des patients, il est possible d'intégrer la vision du patient dans la démarche d'AQ du microsystème clinique sous deux angles complémentaires :

- La capture de l'expérience patient du parcours de soins permet ***d'apprendre des patients*** grâce à des enquêtes qui permettent simultanément une mesure la fréquence des observations ;
- La participation régulière de patients à des groupes de travail avec l'équipe soignante permet ***d'apprendre avec les patients***, d'intégrer de façon dynamique leur vision aux étapes de la démarche qualité ; diverses techniques peuvent être utilisées au cours de la démarche telles que le « *shadowing*» (*suivre le patient comme son ombre*) au cours de certains processus, ou le « *design thinking* » (co-conception de solutions à des problèmes récurrents vécus par les patients)

Toutefois, le travail en groupe interprofessionnel incluant le(s) patient(s) n'est pas inné : le Canadian Interprofessional Competency Framework décrit les compétences nécessaires à tout professionnel impliqué dans un travail collaboratif avec des patients et des familles (54) dans les différents domaines suivants :

- Le fonctionnement de l'équipe : communication responsable, écoute mutuelle, respect des compétences et de l'expérience de chacun, réflexivité sur le fonctionnement, éthique de confidentialité, gestion de la dynamique du groupe, environnement sécurisant pour tous ;
- La clarification des rôles et responsabilités de chacun : responsabilité clinique ou soignante, compétences et expérience du patient, rôle des autres professions dans l'hôpital ;
- Le leadership collaboratif : encouragement à l'expression de chacun et sa participation à la prise de décision, répartition du leadership sur différents membres du groupe en fonction des sujets, développement des solidarités dans la mise en œuvre ;
- La gestion des conflits éventuels au sein du groupe – générés par les différences de points de vue, l'ambigüité des rôles ou des difficultés antérieures non dépassées – et développement de points de consensus.

Il est aisé d'observer que ces compétences pourraient aussi s'appliquer aux patients engagés dans un travail collaboratif avec les soignants, car elles ne sont pas spécifiques d'une discipline médicale ou soignante.

La participation des patients au sein de groupes de travail interprofessionnels dans le microsystème clinique leur confère une place de ***partenaires*** des soignants permettant ***d'apprendre*** ensemble de leurs expériences et de concevoir des actions d'amélioration pour une prise en charge centrée sur leurs besoins. Un cadre de ***compétences*** a été décrit (au Canada), centré sur l'acquisition des compétences de fonctionnement en équipe, pour engager les professionnels soignants, hautement qualifiés et répondant aux normes de leur profession (selon l'acception de Mintzberg) dans un travail pluridisciplinaire et en collaboration avec les patients.

L'article I présenté dans les pages suivantes synthétise ces réflexions présentées et débattues dans le cadre d'un Atelier organisé lors des Journées annuelles du Laboratoire (LEPS) sur le thème « Quelles collaborations des Patients dans les démarches qualité des soins ? ».

organisation

Vers une participation active des usagers dans les démarches qualité

Depuis les années 1990, la démarche qualité se développe dans les hôpitaux selon une logique d'évaluation, sur la base de référentiels, ou selon une logique d'amélioration continue de la qualité, plus propice à la participation des acteurs. L'augmentation des maladies chroniques invite à la participation des patients, porteurs d'une expertise sur la maladie et les soins associés. Les patients peuvent également s'inscrire dans une intervention à visée collective, favorable à l'amélioration de l'offre de soins.

Mots clés – collaboration patient-soignant ; démarche qualité ; expertise patient ; maladie chronique

Towards the active participation of users in quality approaches. Since the 1990s, the quality approach has been developing in hospitals in accordance with an assessment strategy, based on reference guidelines, or with a continuous quality improvement strategy, more favourable to the participation of players. The increase in chronic diseases encourages the participation of patients, with their own particular expertise on the disease and the associated care. Patients can also be involved in a collective mission, favourable to the improvement of the care provision.

Keywords – chronic disease; patient-caregiver collaboration; patient expertise; quality approach

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Selon les fondements du management de la qualité posés par William Edwards Deming, statisticien, au début des années 1940[1], les produits défectueux sont le plus souvent liés à des défauts d'organisation et non à la faute de l'ouvrier. Les normes internationales ISO 9000[2], parues à partir de 1987, établissent les principes du management de la qualité (PMQ) : orientation client, *leadership*, implication du personnel, approche processus, amélioration continue, prise de décision fondée sur des preuves, management des relations avec les parties intéressées. Toutefois, le client n'est pas directement associé à la démarche qualité menée au sein de l'entreprise, en dehors de la prise en compte de ses exigences et attentes.

L'introduction des démarches qualité dans le service public français a connu plusieurs étapes, depuis les cercles qualité (1987) jusqu'à des approches plus globales et cohérentes, telles que les engagements de services, mesurables par la qualité du service rendu aux usagers, les chartes pour l'accueil du public ou la gestion publique par programmes budgétaires assortis d'indicateurs de qualité. Différentes critiques ont été formulées quant à la compatibilité entre évaluation de l'action publique et mesure d'indicateurs, parfois réducteurs et pouvant orienter le jeu des acteurs [1]. Depuis 1996, les établissements de santé ont l'obligation de mener une procédure d'accréditation où la qualité des soins est appréciée par un organisme externe, sur la base d'un référentiel. Par ailleurs, les agences régionales de l'hospitalisation (ARH) ont pris en compte des objectifs de sécurité et de qualité dans les contrats pluriannuels conclus avec ces établissements. De nombreuses initiatives de démarches qualité internes ont ainsi été mises en œuvre dans le but de modifier le fonctionnement de l'organisation, dans l'intérêt des patients, et en s'adaptant aux progrès médicaux et technologiques. Le guide de l'Agence nationale d'accréditation et d'évaluation en santé (Anaes, aujourd'hui Haute Autorité de santé [HAS]), publié en 2002, préconise l'attention aux "clients externes" (patients et professionnels ayant adressé des patients à l'établissement) comme l'un des principes essentiels des démarches qualité en établissement de

Notes

[1] Dans les années 1940, Edwards Deming, ingénieur statisticien américain, a mis en évidence par observation de pratiques industrielles que, contrairement à ce que l'on pensait, les produits défectueux avaient majoritairement pour origine des défauts d'organisation et non la faute de l'ouvrier. Il en déduit que l'engagement des dirigeants, aptes à modifier une organisation, engage à réfléchir un système d'amélioration de la qualité qui comprend notamment la formation des directeurs et une réflexion sur toute la chaîne de décision.

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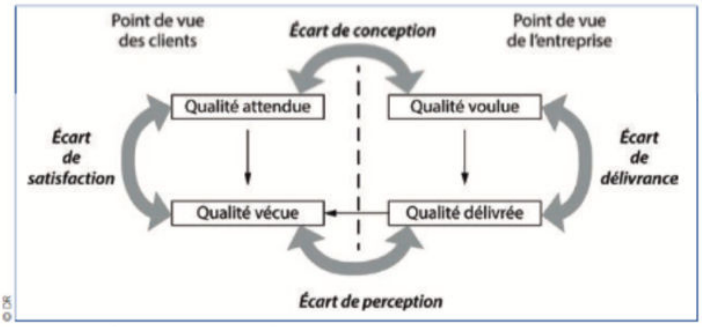

Figure 1. Schématisation de la qualité dans le domaine des services et adaptée à la santé (d'après [3]).

santé [2]. Il distingue notamment les points de vue des clients et de l'établissement, l'écoute des clients devant réduire les écarts de conception, de perception et de satisfaction *(figure 1)* [3].

Le patient traceur

La procédure de certification V 2014[1] introduit la méthode du patient traceur comme outil permettant aux experts visiteurs de la Haute Autorité de santé (HAS) et aux équipes de soins, la prise en compte de l'expérience des patients hospitalisés. Elle est également promue pour évaluer, analyser le dossier du patient et mettre en œuvre des actions d'amélioration rapide de la qualité et de la sécurité des soins. La méthode prend en compte l'expérience de patients identifiés selon des critères définis au préalable par l'équipe, par un entretien mené la plupart du temps par des soignants.

Dans ce contexte, il est apparu important pour l'hôpital d'instruction des armées Bégin (DCSSA, Saint-Mandé [94]) de mener une réflexion sur les bénéfices de la réalisation de ces entretiens par un binôme représentant d'usagers (RU)/soignant. À la suite de la formation de trois binômes à la démarche et à la conduite conjointe de l'entretien, neuf entretiens avec des patients traceurs ont été réalisés. L'observation, non participante, de la formation et des entretiens, ainsi que des entretiens semi-directifs avec les binômes et les patients, ont permis d'évaluer le ressenti des patients et le type d'informations transmises.

Les résultats ont permis de constater que la présence du RU améliore la prise de parole du patient et permet la prise en compte globale de la parole du patient. Le RU s'investit personnellement dans l'entretien, l'enrichit et permet au patient d'être plus libre dans son expression. Il permet également de s'assurer de la prise en compte réelle de la parole des patients en la covalidant avec le soignant et en la portant auprès de la direction lors de la Commission des usagers.

Pour être réussie, la collaboration RU/soignant doit passer par une interprofessionnalité effective, favorisée par la formation action, la mise en place d'une confiance mutuelle et l'accompagnement du binôme.

[1] *Haute Autorité de santé (HAS). La V 2014. www.has-sante.fr/portail/jcms/r_1495044/fr/la-v2014*

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Des instances et des outils ont été créés afin d'améliorer la participation des usagers et de promouvoir leurs droits individuels et collectifs dans le système de santé (commission des relations avec les usagers et de la qualité de la prise en charge [CRUQPC], aujourd'hui commission des usagers [CDU], enquêtes de satisfaction sous forme de questionnaires de sortie). Toutefois, la participation des usagers dans les démarches qualité internes n'a guère été envisagée au-delà du recueil de leurs attentes et de leur satisfaction vis-à-vis des soins reçus.

Démarches qualité ou culture de la qualité

Dès 2001, deux types de démarches qualité se sont distinguées dans les établissements de soins, l'une s'appuyant sur une référence pour créer une dynamique d'amélioration, tandis que l'autre crée une dynamique d'amélioration pour ensuite intégrer des références [4].

Le premier type est caractérisé par une évaluation initiale selon un référentiel qui dépend de l'organisme initiateur de la démarche qualité (référentiel normatif de la série ISO 9000 ou référentiels professionnels de type accréditation/certification). La démarche, menée par un organisme externe ou par les pairs, évalue des écarts entre les pratiques et le référentiel, et se traduit par un plan d'action d'amélioration à relativement court terme.

Le second type débute par une mobilisation des professionnels autour de l'amélioration de la qualité, puis l'intégration de référentiels pour améliorer le fonctionnement de l'organisation. Il s'agit ici de commencer par faire évoluer la culture de l'organisation pour, *in fine*, changer les résultats produits. Ces approches participatives s'appuient sur le *leadership* pour conduire progressivement à des changements portant à la fois sur le fonctionnement interne du service et sur l'évolution de son positionnement dans l'établissement. Elles s'inscrivent dans une dynamique d'amélioration continue de la qualité. Ces démarches de type 2, plus rares en France, sont fréquentes en Amérique du nord, développées notamment par le Harvard Institute for Healthcare Improvement[5] (IHI) et The Dartmouth Institute Microsystem Academy[6] (TDIMA). Elles sont centrées sur le microsystème clinique, c'est-à-dire *« le lieu qui regroupe les soignants et les patients/familles pour une prise en charge régulière du patient, là où la prise en charge réelle traduit la réalité de la pratique »* [5]. C'est en effet au niveau du

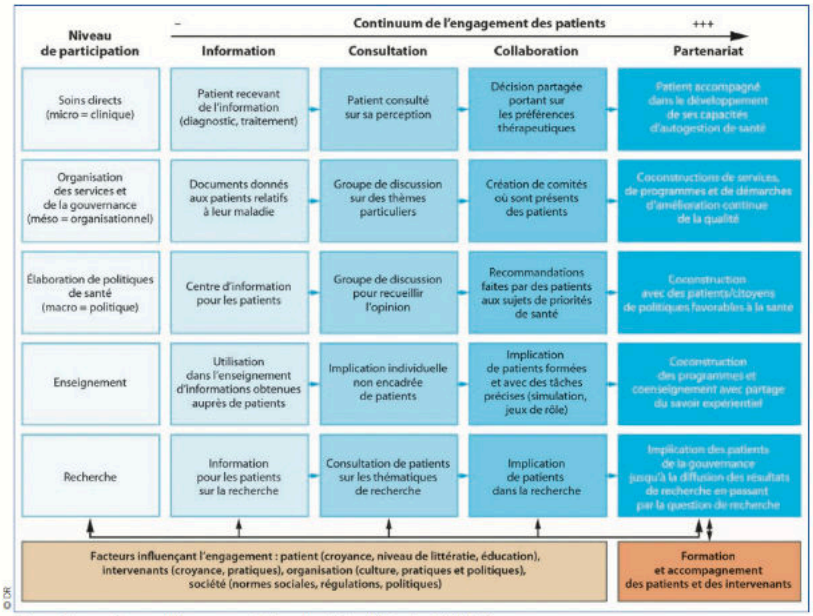

Figure 2. Le continuum de l'engagement des patients (traduit et adapté de [11]).

microsystème clinique que sont produits la qualité et la sécurité des soins, les coûts et la performance, la satisfaction des patients et des professionnels. Pour assurer une reconnaissance par l'administration hospitalière des équipes engagées, ce type de démarche qualité doit converger vers la certification de l'établissement à travers la déclaration des actions en évaluation des pratiques professionnelles (EPP), le suivi d'indicateurs intégrés au tableau de bord de l'établissement et la valorisation des professionnels impliqués (développement professionnel continu [DPC]).

Le défi de la prise en charge des maladies chroniques

Le nombre croissant de personnes atteintes de maladies chroniques constitue un défi majeur pour le système de santé, en termes de morbidité, de mortalité, d'impact financier, mais également en termes de qualité et de sécurité des soins. Or, les démarches qualité sont configurées pour une organisation hospitalière centrée sur des prises en charge de court séjour plus que sur celles de longue durée articulant plusieurs spécialités à l'hôpital, les professionnels de ville, ainsi que le patient lui-même et/ou son entourage. Des référentiels de prise en charge de certaines pathologies chroniques sont définis sous la forme de parcours de soins d'un patient, intégrant l'évolution de la pathologie dans la durée [6]. Leur déclinaison sous la forme d'un chemin clinique [7] tout au long du parcours selon l'analyse des processus n'est pas généralisée [8]. Les instruments d'évaluation ciblés sur une venue du patient dans l'établissement, pour utiles qu'ils puissent être, ne s'appliquent qu'à un fragment de ce parcours de soins, indépendamment de la continuité de celui-ci dans la durée. La question du modèle et des critères d'évaluation des prises en charge des pathologies chroniques est donc posée.

À ces difficultés inhérentes aux particularités de la prise en charge des maladies chroniques dans la durée, s'ajoutent les caractéristiques des patients eux-mêmes. En effet, ces patients sont des usagers permanents du système de santé. Ils connaissent les étapes du parcours de soins et les complications, sont devenus les experts de l'expression de la maladie et des effets des traitements pour eux-mêmes et peuvent souvent bénéficier de l'expérience ou du soutien d'autres patients, ou des associations. Le *Chronic Care Model* propose de définir un modèle générique d'évaluation de la prise en charge de ces

Notes

[2] Les normes ISO 9000 rassemblent une série de démarches qui s'intéressent aux différents aspects du management de la qualité. Elles proposent des lignes directrices et des outils aux systèmes de production dans le but d'améliorer constamment la qualité de leurs actions et de leurs produits, en adéquation avec les besoins et les attentes des publics destinataires. Ces démarches se traduisent par des normes qui établissent les exigences relatives à un système de management de la qualité (ISO 9001:2015), qui couvrent les notions fondamentales et la terminologie (ISO 9000:2015), qui montrent comment augmenter l'efficience et l'efficacité d'un système de management de la qualité (ISO 9004:2009) ou qui établissent des lignes directrices pour les audits internes et externes des systèmes de management de la qualité (ISO 19011:2011). Initialement développées dans le domaine industriel, les normes ISO ont été transposées dans les établissements de santé.

[3] Institute for Healthcare Improvement. www.ihi.org/engage/fellowships/Pages/default.aspx

[4] The Dartmouth Institute Microsystem Academy. http://tdi.dartmouth.edu/education/professional-education/coaching-in-quality-improvement

"partenariat de soins et de services", entre les soignants et le patient, ont été présentés selon sept domaines :

- **l'éducation thérapeutique du patient** à sa maladie et aux traitements ;
- **l'éthique clinique** ;
- **la clarification des rôles et des responsabilités** ;
- **la prévention et la résolution des conflits** ;
- **la communication** efficace et responsable ;
- **le *leadership*** collaboratif ;
- **la capacité de travail en équipe**.

Ces compétences concernent les soignants et les patients.

Des moyens pratiques complémentaires sont nécessaires *a minima* pour permettre d'intégrer de(s) patient(s) volontaire(s) dans une démarche qualité :

- **leur disponibilité** sur la durée (1 an ou plus), à adapter selon la démarche qualité ;
- **la possibilité d'associer plusieurs patients** de profils différents pour tenir compte de divers points de vue et de prises en charge différenciées ;
- **l'utilisation d'Internet** pour travailler avec l'équipe à distance ;
- **le dédommagement des frais de déplacement** pour les réunions physiques avec l'équipe et la prise en charge des frais de participation à la formation, dans les mêmes conditions que les professionnels.

Dans le cadre de la démarche qualité PHARE-M (Programme national d'amélioration de la qualité, *encadré 1*), le patient (ou le parent, en pédiatrie) qui s'est impliqué aux côtés de l'équipe a utilisé des compétences professionnelles antérieures (informatique et systèmes d'information, qualité dans l'industrie ou les services, communication et relations publiques, compétences associatives) qui lui ont conféré une certaine assurance personnelle et une valeur ajoutée immédiate auprès de l'équipe soignante. Une procédure de recrutement a été mise en œuvre au sein de la file active afin de préciser les conditions de participation du patient/parent et de vérifier les critères évoqués plus haut. Un correspondant est désigné parmi l'équipe professionnelle pour être l'interlocuteur du patient tout au long de sa participation à la démarche. Il a la charge de l'informer régulièrement, de lui communiquer les notes des réunions, de l'inviter aux différentes réunions et de veiller au respect des engagements réciproques tout au long de la démarche. Enfin, une évaluation régulière de l'impact de la participation du patient sur son état psychologique ou sa santé, et de sa volonté de continuer à participer (notamment en cas d'évolution de sa santé ou d'autres facteurs personnels), est réalisée.

Les points à retenir

- **Le développement de la culture qualité dans les hôpitaux** suppose des démarches d'amélioration rapide de la qualité, centrées sur le microsystème clinique, et d'amélioration continue de celle-ci, fondées sur des référentiels professionnels.
- **L'engagement des patients** passe par des enquêtes de consultation sur leur expérience des soins, leur inclusion dans des commissions de travail et leur participation active aux démarches qualité.
- **La participation active des patients aux démarches qualité** repose sur des compétences adaptées, des processus de recrutement, des dispositifs pratiques et un dialogue permanent entre les acteurs.

Conclusion et perspectives

Le partenariat patient-soignants dans l'amélioration de la qualité se construit progressivement et suit l'évolution du modèle relationnel pour les soins eux-mêmes. Les équipes les plus "ouvertes" sont celles qui ont déjà intégré la pratique de l'éducation thérapeutique des patients dans leur prise en charge.

Une telle évolution culturelle est forcément longue, mais ne peut s'opérer que s'il existe une volonté forte des instances de santé, à tous les niveaux du système. Que vaudrait, en effet, une volonté de participation des patients au niveau du microsystème, qui se heurterait à des freins au niveau du mésosystème et/ou du macrosystème ? Que pourrait obtenir une volonté manifestée au niveau du macrosystème qui ne serait pas relayée aux niveaux méso- ou micro- ?

La révolution technologique du numérique en santé ouvre la voie à la collecte et à l'échange de données massives concernant les patients, et permet la construction d'une nouvelle connaissance partagée entre patients, soignants et chercheurs. Cette révolution, qui concerne au premier chef les patients atteints de maladie chronique, peut conduire à des tensions fortes entre des patients "informés et activés" et des équipes qui ne seraient pas "préparées et proactives". ■

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Déclaration de liens d'intérêts
Les auteurs déclarent ne pas avoir de liens d'intérêts.

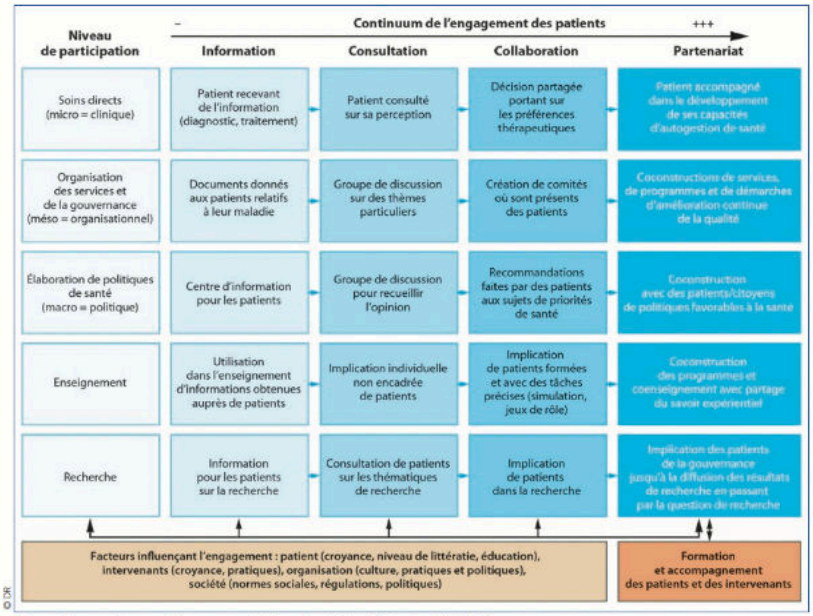

Figure 2. Le continuum de l'engagement des patients (traduit et adapté de [11]).

microsystème clinique que sont produits la qualité et la sécurité des soins, les coûts et la performance, la satisfaction des patients et des professionnels. Pour assurer une reconnaissance par l'administration hospitalière des équipes engagées, ce type de démarche qualité doit converger vers la certification de l'établissement à travers la déclaration des actions en évaluation des pratiques professionnelles (EPP), le suivi d'indicateurs intégrés au tableau de bord de l'établissement et la valorisation des professionnels impliqués (développement professionnel continu [DPC]).

Le défi de la prise en charge des maladies chroniques

Le nombre croissant de personnes atteintes de maladies chroniques constitue un défi majeur pour le système de santé, en termes de morbidité, de mortalité, d'impact financier, mais également en termes de qualité et de sécurité des soins. Or, les démarches qualité sont configurées pour une organisation hospitalière centrée sur des prises en charge de court séjour plus que sur celles de longue durée articulant plusieurs spécialités à l'hôpital, les professionnels de ville, ainsi que le patient lui-même et/ou son entourage. Des référentiels de prise en charge de certaines pathologies chroniques sont définis sous la forme de parcours de soins d'un patient, intégrant l'évolution de la pathologie dans la durée [6]. Leur déclinaison sous la forme d'un chemin clinique [7] tout au long du parcours selon l'analyse des processus n'est pas généralisée [8]. Les instruments d'évaluation ciblés sur une venue du patient dans l'établissement, pour utiles qu'ils puissent être, ne s'appliquent qu'à un fragment de ce parcours de soins, indépendamment de la continuité de celui-ci dans la durée. La question du modèle et des critères d'évaluation des prises en charge des pathologies chroniques est donc posée.

À ces difficultés inhérentes aux particularités de la prise en charge des maladies chroniques dans la durée, s'ajoutent les caractéristiques des patients eux-mêmes. En effet, ces patients sont des usagers permanents du système de santé. Ils connaissent les étapes du parcours de soins et les complications, sont devenus les experts de l'expression de la maladie et des effets des traitements pour eux-mêmes et peuvent souvent bénéficier de l'expérience ou du soutien d'autres patients, ou des associations. Le *Chronic Care Model* propose de définir un modèle générique d'évaluation de la prise en charge de ces

Notes

[2] Les normes ISO 9000 rassemblent une série de démarches qui s'intéressent aux différents aspects du management de la qualité. Elles proposent des lignes directrices et des outils aux systèmes de production dans le but d'améliorer constamment la qualité de leurs actions et de leurs produits, en adéquation avec les besoins et les attentes des publics destinataires. Ces démarches se traduisent par des normes qui établissent les exigences relatives à un système de management de la qualité (ISO 9001:2015), qui couvrent les notions fondamentales et la terminologie (ISO 9000:2015), qui montrent comment augmenter l'efficience et l'efficacité d'un système de management de la qualité (ISO 9004:2009) ou qui établissent des lignes directrices pour les audits internes et externes des systèmes de management de la qualité (ISO 19011:2011). Initialement développées dans le domaine industriel, les normes ISO ont été transposées dans les établissements de santé.

[3] Institute for Healthcare Improvement. www.ihi.org/engage/fellowships/Pages/default.aspx

[4] The Dartmouth Institute Microsystem Academy. http://tdi.dartmouth.edu/education/professional-education/coaching-in-quality-improvement

III- CADRE DU TRAVAIL, STRATEGIE D'IMPLEMENTATION ET DESIGN DE L'INTERVENTION PHARE-M

L'exposé précédent conduit au constat que « toutes les briques existent » pour envisager la participation de patients atteints de maladie chronique à une démarche qualité collaborative dans le format des *Breakthrough Series,* en tant que partenaires de leurs équipes soignantes.

Cette démarche qualité a été initiée en France à partir de 2011 dans la filière de soin mucoviscidose, adaptée du programme qualité mis en place par la Cystic Fibrosis Foundation (US CFF) aux Etats-Unis depuis 2002. Elle constitue une innovation en France, dans son format collaboratif de programme de formation-action annuel (modèle des BTS) et dans la participation de patients (ou parents d'enfants en pédiatrie) dans l'équipe de pilotage organisée dans chaque centre de ressources et de compétences (CRCM) engagé dans ce programme dénommé PHARE-M[‡].

Nous présentons ci-après un survol de l'histoire de la prise en charge de la mucoviscidose et des caractéristiques qui en ont fait une maladie « ***modélisante*** » pour les innovations organisationnelles dans le système de santé français concernant les maladies rares, et un exemple de collaboration de longue date entre les équipes soignantes et les parents d'enfants malades.

Nous indiquons ensuite les conditions qui ont accompagné l'introduction de cette démarche qualité dans le contexte national de la mucoviscidose et le positionnement des différents acteurs de la filière face à l'introduction de cette innovation dans la conduite de l'amélioration de la qualité (« *implementation strategy »).*

Nous décrivons enfin les caractéristiques du programme PHARE-M et les adaptations principales apportées lors de l'adoption en France de cette démarche qualité développée pour la mucoviscidose dans le contexte américain, notamment le pré-requis de l'intégration de patients et de parents dans les équipes de pilotage de la qualité constituées dans les CRCM.

III-1. Historique et caractéristiques de la mucoviscidose

L'histoire de la mucoviscidose, maladie génétique, rare, chronique et évolutive, illustre ***l'évolution conjointe des connaissances scientifiques internationales et de l'organisation de la prise en charge***, qui en quelques décennies ont considérablement amélioré la survie et les conditions de vie des patients, dans les pays dans lesquels le système de santé a organisé une prise en charge adaptée de ces enfants et de leurs parents.

Depuis le milieu des années 60, cette maladie a été identifiée comme une entité pathologique à part entière et le diagnostic a été mis au point avec le « test de la sueur » (Gibson et Cooke, 1959). Grâce au développement des antibiotiques, le traitement agressif des infections respiratoires a été recommandé dès 1969 (Lawson), avec la mise en place de cures IV régulières et la prévention des contaminations entre patients et par l'environnement ; ces traitements antibiotiques intraveineux d'abord réalisés lors de séjours de 2 à 3 semaines à l'hôpital ont évolué vers une administration à domicile puis vers des nébulisations antibiotiques à partir de 1981 (Margaret Hodson). Les progrès au niveau des soins chirurgicaux et médicaux néonataux et l'évolution de l'attitude nutritionnelle – de la préconisation de

[‡] Programme Hospitalier d'Amélioration des Résultats et de l'Expertise en Mucoviscidose

l'« Allan Diet » à une prise en charge nutritionnelle combinant l'apport des enzymes pancréatiques et un régime hypercalorique (1978), ont été associés à un meilleur pronostic au niveau de la fonction respiratoire et de la survie. La réussite de la première transplantation cœur-poumons en 1984 (à Chapel Hill, USA) a permis d'envisager ce traitement de l'ultime chance pour les patients atteints de mucoviscidose ; elle a rapidement évolué vers la transplantation bi-pulmonaire seule pour ces patients qui sont jeunes et ne présentent pas d'altération de leur fonction cardiaque. La découverte du gène en 1989 (Lap-Chi Tsui) a ouvert la voie à un effort de recherche sans précédent sur les causes de la maladie et abouti très récemment (2015) à la mise sur le marché des premiers médicaments correcteurs et potentiateurs de la protéine du gène *CFTR* ciblant certaines mutations génétiques (2012, Kalideco ; 2015, Orkambi).

Parallèlement à la mise au point et à la mise sur le marché de ces traitements, un consensus s'est progressivement installé (Peter Phelan, 1984) pour une prise en charge pédiatrique spécialisée en équipe pluridisciplinaire, intégrant les aspects nutritionnel, respiratoire, psychologique, social, l'éducation des parents et des enfants et la coordination des soins à domicile, l'orientation vers des soins de spécialités lors de l'apparition des complications à l'adolescence. En quelques années, un allongement spectaculaire de la durée de vie des patients a été observé : l'espérance de vie au milieu des années 60 était inférieure à 7 ans tandis que les décès à l'âge pédiatrique sont devenus exceptionnels au tournant des années 2000. Des centres spécialisés dans la prise en charge des adultes apparaissent avec l'arrivée à l'âge adulte des enfants et des recommandations pour leur organisation sont édictées par l'US CFF dès les années 90. A partir des années 2010, la moitié des patients recensés dans les Registres dans les pays ayant développé une prise en charge pédiatrique spécialisée sont des adultes (Registre canadien, 2010 ; Registre français de la mucoviscidose, 2013). L'évolution de la maladie vers les états respiratoires sévères est aujourd'hui majoritairement prise en charge dans ces centres adultes. L'articulation des centres adultes spécialisés avec les centres de transplantation pulmonaire s'établit de façon plus ou moins fluide dans les différents pays selon leurs schémas d'organisation et de financement du système de santé.

En France, la mucoviscidose a été de longue date une maladie « ***modélisante*** » pour les maladies rares, à travers la ***structuration pionnière de son organisation de soins en centres spécialisés*** et la diffusion rapide des innovations thérapeutiques dans cette filière, soutenue par une ***collaboration étroite et continue entre les cliniciens, les chercheurs et les parents*** d'enfants malades réunis dans l'association de lutte contre la mucoviscidose (AFLM), rebaptisée Vaincre la Mucoviscidose.

La mise en place expérimentale en Bretagne, région de forte prévalence de la maladie, du dépistage néonatal systématique (DNS) dès 1989 a été promue par un trio regroupant clinicien – généticien – parent dans la foulée de la découverte du gène (55). Malgré l'absence de preuve scientifiquement irréfutable de son efficacité, l'Assurance Maladie a pris, en 2001, la décision de financer le DNS de la mucoviscidose, motivée par les perspectives crédibles d'efficacité de l'organisation proposée pour la prise en charge des enfants et des familles dans des centres de ressources et de compétences de la mucoviscidose - CRCM (56). Cette organisation préfigurait la création de la filière maladie rare Muco-CFTR publiée dans le cadre du 2ème plan national maladies rares en 2014 (INSTRUCTION N°DGOS/PF2/2014/126 du 18 avril 2014).

L'Observatoire national de la mucoviscidose a été créé dès 1992, sur le modèle du Registre américain. Il est géré par l'association Vaincre la Mucoviscidose et a été qualifié par le Comité national des Registres en 2007 sur des critères d'exhaustivité et de qualité. En 2015, le Registre recense 6585 patients suivis dans les CRCM, dont 53% d'adultes.

Dès 2005, l'éducation thérapeutique du patient (ETP) s'est développée, anticipant son inscription dans la loi comme un élément intégré au parcours de soin (loi HPST 2009), avec la création d'un groupe national animé par le médecin responsable du CRCM pédiatrique de Nantes et la directrice médicale de l'association Vaincre la Mucoviscidose, pour élaborer un référentiel de compétences et des outils éducatifs à l'attention des parents ainsi que de leurs enfants et des adolescents atteints de mucoviscidose.

Dans le cadre du 1er plan national maladies rares, deux centres de référence maladie rare (CRMR) mucoviscidose ont été labellisés en 2006 sur appel d'offres parmi les 49 CRCM – les CRCM du CHU Nantes et des Hospices Civils de Lyon, chargés de mettre en œuvre des missions nationales transversales (notamment l'ETP) et le second plan national maladies rares a officialisé la Filière Muco-CFTR.

Les travaux en ETP se prolongent vers la population de patients adultes, aux étapes critiques de leur prise en charge : diagnostic éducatif adultes (2010) ; codification de la transition pédiatrique-adulte (2014) ; transition vers les états d'insuffisance respiratoire sévère et la transplantation pulmonaire (2016-2017).

Le développement de la transplantation pulmonaire a été soutenu par la création de l'association Gregory Lemarchal – suite au décès sur liste d'attente du patient atteint de mucoviscidose et chanteur de 24 ans, Grégory Lemarchal, en mai 2007. La liste de Super Urgence a été mise en place en 2007 et a permis d'éviter des décès sur liste par une gestion des priorités d'attribution des greffons. Le soutien fort des deux associations rassemblées sur cette cause et les campagnes successives de dons d'organes ont permis d'améliorer l'accès à cette thérapie en augmentant l'offre de greffons. L'arrivée de la technologie de réhabilitation des greffons (Hôpital Foch à Suresnes) a permis d'accroître cette offre avec des greffons à critères élargis, refusés sans cette technique. Cette évolution a permis de proposer de façon plus systématique cette intervention aux patients atteints de mucoviscidose en phase d'insuffisance respiratoire sévère et d'infection chronique résistante aux antibiotiques. La transplantation pulmonaire pour mucoviscidose représentait, en 2013, ***30% des indications de transplantation en France***. En 2015, 700 patients adultes vivaient avec un greffon pulmonaire, soit ***20% de la population adulte atteinte de mucoviscidose***. De nouveaux défis se posent pour cette population qui cumule deux « pathologies rares » – la mucoviscidose avec des complications (diabète) et la transplantation pulmonaire, qui expose à de nouvelles complications (rejet chronique du greffon, insuffisance rénale, cancers) et dont la prise en charge est éclatée entre deux services spécialisés, le CRCM et le centre de transplantation.

La ***collaboration entre les associations de parents, les soignants et les chercheurs*** en mucoviscidose a opéré historiquement au niveau du macro-système de santé, sur les questions liées à la structuration des CRCM, au soutien financier au recrutement de professionnels dans les équipes pluridisciplinaires et au soutien à la recherche via le financement d'appels à projets nationaux. L'association Gregory Lemarchal développe quant à elle des actions structurantes de réaménagement des

locaux de soins ou d'accueil des patients et familles dans les hôpitaux hébergeant des CRCM ou des centres de transplantation.

Si l'extension de cette collaboration au domaine de l'amélioration de la qualité des soins apparaît comme le prolongement logique de l'implication associative dans la structuration des ressources (et la nécessité d'évaluer l'efficacité des financements accordés sur des fonds associatifs), elle constitue en réalité le « franchissement » d'un niveau du système de soin, associé à des difficultés nouvelles liées à : 1) ***la diversité des contextes locaux des 45 CRCM***[§], qui se traduit par une application variable du cahier des charges des CRCM et du PNDS mucoviscidose (57) 2) ***des ressources associatives limitées localement*** et plus orientées sur la collecte de fond (Virades de l'espoir) que sur la relation avec les hôpitaux et 3) ***une relation individuelle entre parent et soignants,*** entre dépendance (et vulnérabilité lors des aggravations de santé de l'enfant) et vigilance, voire agressivité lors des incidents vécus lors de la prise en charge d'un épisode difficile. Pour exemple, l'incitation de l'association à la mise en place de collectifs de parents dans chaque CRCM, dès la création de ceux-ci, avait été suivie de peu de réalisations, faute de parents volontaires pour s'investir et de confiance accordée à cette initiative de la part d'une majorité de médecins.

Une nouvelle alliance entre parents et soignants au niveau du microsystème clinique devait donc se construire pour améliorer la qualité des soins dans le CRCM. La démarche qualité collaborative présentée au § III-3 pouvait en être le vecteur, comme l'éducation thérapeutique est le vecteur d'une alliance thérapeutique entre les parents et les soignants pour les soins individuels apportés à l'enfant.

III-2. Un parcours personnel à l'origine de l'engagement dans l'amélioration des soins avec les soignants

Suite au diagnostic tardif de mucoviscidose de mon fils aîné (avant la généralisation du DNS et la création des CRCM) j'ai été brutalement confrontée, comme de nombreux parents, au rôle de « soignant à domicile » et « coordinateur des soins » de l'enfant. Une formation d'ingénieur, et une longue expérience de consultante dans l'industrie dans l'amélioration de la qualité et de l'organisation des processus industriels, m'ont très vite fait porter attention à l'organisation des processus de soin, la coordination des intervenants et la transmission des informations, la réactivité de mise en route des traitements. Sans culture clinique, et sans l'éducation thérapeutique dispensée aujourd'hui par les équipes des CRCM, cette « culture de la qualité » m'a permis de partager avec les soignants des observations sur la « non qualité » de certains processus, par exemple : 1) des délais trop longs d'obtention des résultats des examens cytobactériologiques des crachats (ECBC) pour la mise en route des traitements antibiotiques, pourtant déterminants de la restauration de la fonction respiratoire du patient après une exacerbation respiratoire, 2) l'idée de tester la faisabilité d'un prélèvement des crachats une semaine avant la consultation de l'enfant pour disposer des résultats au cours de celle-ci et débuter le traitement, si besoin, sans délai, 3) l'importance de développer les compétences psycho-sociales de l'enfant pour lui permettre d'exposer ses besoins spécifiques à l'école, et en conséquence, 4) l'importance d'intégrer l'éducation thérapeutique dans le suivi courant et le temps des consultations….

[§] Quarante cinq des 49 CRCM ont été requalifiés en 2015.

Mon engagement dans l'association Vaincre la Mucoviscidose, puis la fonction d'administratrice, m'a permis de participer à divers groupes de travail nationaux, associant Vaincre la Mucoviscidose et les centres de référence maladies rares mucoviscidose (CRMR-M) et à des congrès internationaux de la mucoviscidose (North American CF Conference, European CF Conference).

La question des ressources professionnelles nécessaires pour assurer la prise en charge des patients conformément aux recommandations nationales (PNDS) et aux publications internationales a conduit à lancer en 2009 une analyse d'activité des ressources disponibles pour la prise en charge ambulatoire des patients en France, dans un échantillon de 7 CRCM pédiatriques et adultes : cette étude a été suivie d'une publication en vue des négociations de la revalorisation de la *MIG*[**] *Mucoviscidose* auprès du Ministère (58).

Les échanges internationaux ont ainsi ouvert sur des collaborations avec divers groupes, notamment ceux la Cystic Fibrosis Foundation (US-CFF) et du Dartmouth Institute. La participation à la formation qualité dispensée par le Dartmouth Institute paraissait donc être la suite logique de cet engagement pour préparer la transposition et le lancement du programme qualité dans la mucoviscidose en France.

A l'occasion de rencontres avec de nombreux parents et patients adultes, en France, aux USA et en Europe, j'ai pu constater la richesse de leur ***expérience de l'organisation des soins***, leur motivation à améliorer les soins pour tous et l'enjeu de relations ouvertes et constructives avec les équipes soignantes pour dépasser les difficultés inéluctablement rencontrées au cours d'un suivi de longue durée dans le système de soin. La collaboration soignants-parents/patients portée par l'éducation thérapeutique ouvrait la voie à une collaboration pour améliorer l'organisation des soins. Une ***démarche qualité collaborative*** ancrée dans le fonctionnement de l'équipe pluridisciplinaire, reposant sur une méthode, des outils et une animation du travail en équipe pouvait créer les conditions d'une mobilisation des professionnels et des patients/parents sur des objectifs d'amélioration partagés dans les centres. La démarche qualité mise en œuvre aux USA par la Fondation américaine et l'institut de Dartmouth depuis 2002 répondait à cet objectif et semblait pouvoir s'appliquer dans les CRCM en France. Aux différentes étapes de l'implémentation du programme depuis 2011 mon rôle a alterné entre celui de « parent coordinateur » et/ou de « parent formateur » du programme qualité en France.

L'article présenté dans les pages suivantes fait suite à une communication orale plaidant pour la collaboration entre parents et soignants pour améliorer les soins, dans le cadre d'une table ronde sur l'éthique de la collaboration en pédiatrie lors du congrès de la société française de pédiatrie en mai 2015 (59).

[**] MIG : Mission d'Intérêt Général

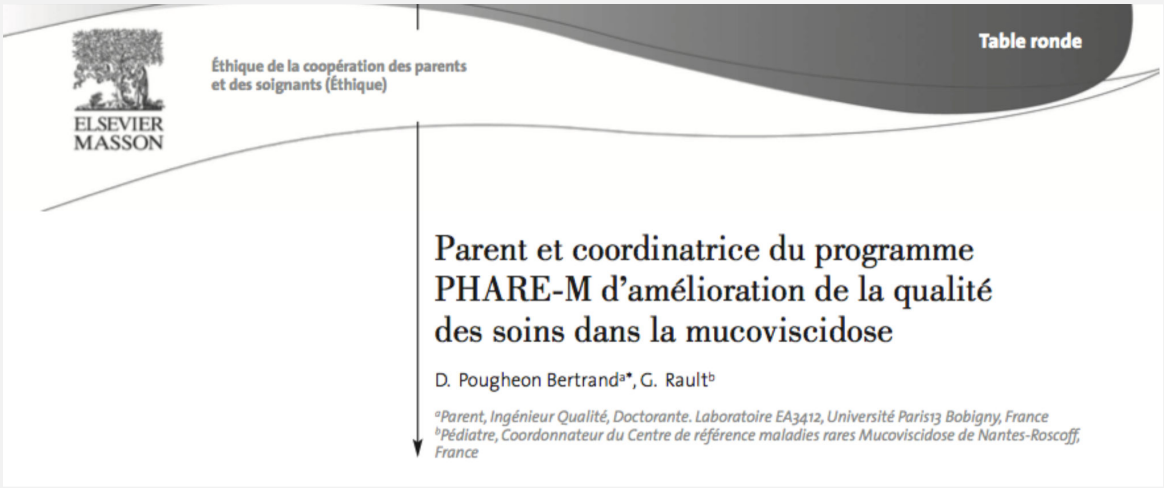

Table ronde

Éthique de la coopération des parents et des soignants (Éthique)

ELSEVIER MASSON

Parent et coordinatrice du programme PHARE-M d'amélioration de la qualité des soins dans la mucoviscidose

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1. Des soins à l'amélioration de la qualité des soins

La relation de coopération entre parents et soignants a pour enjeu les soins donnés à l'enfant. Elle évolue avec la maladie chronique et les projets de vie de la famille puis de l'adolescent. Pour une maladie génétique rare comme la mucoviscidose, le chemin à parcourir ensemble commence avec le diagnostic, moment crucial destructeur et refondateur qui conduira à une alliance avec l'équipe soignante. Chaque annonce difficile qui ravive les émotions, les incompréhensions inévitables et l'évolution des connaissances de part et d'autre mettront à l'épreuve cette relation singulière de coopération. L'objectif de bien-faire peut alors se transformer en celui de continuellement mieux faire en considérant les points de vue de chaque personne impliquée dans le microsystème de soins.

2. L'engagement du parent dans la coopération avec les soignants pour les soins de l'enfant

L'annonce de la maladie est un traumatisme qui s'apparente à une sortie de route, un pare-brise qui vole en éclats masquant la perspective devant soi et stoppant net le mouvement de la vie. Aussi essentielles que soient les conditions mesurées et humaines de l'annonce, pour longtemps gravées dans la mémoire, la brutalité de la situation la rend irréelle : les cauchemars en témoignent qui promettent, à l'instant du réveil, le miracle du retour à la vie passée et dans les instants suivants, une prise de conscience réitérée de l'irréversible réalité. Dans cet arrêt sur image qui peut durer des semaines, monte la souffrance qui se nourrit de toutes les informations et de toutes les angoisses et alimente des visions effrayantes de mort. « Il y a dans la souffrance une absence de tout refuge. Elle est faite de l'impossibilité de fuir et de reculer » [1].

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Dans le même temps, le parent doit « faire avec » et trouver l'énergie pour intégrer la maladie dans le nouvel ordre des choses : apprivoiser l'environnement lié à la maladie, donner des soins et des médicaments, organiser les passages des soignants à domicile, et accepter de s'installer durablement dans ces occupations. Et en s'accrochant aux fondations de la vie d'avant comme à une planche de salut, revoir l'organisation de son travail, établir une coopération au sein du couple, trouver de l'aide, évaluer les conséquences économiques, répondre à l'entourage, prendre soin le mieux possible des autres enfants.

Des ressources individuelles de chaque parent, du réconfort trouvé dans l'entourage, de la qualité de l'information et de l'encouragement donnés par l'équipe pluridisciplinaire dépendent le retour à un équilibre qui lui permet de devenir l'acteur principal dans les soins de l'enfant. L'éducation thérapeutique permet d'installer dans la durée l'alliance avec l'équipe en vue de développer l'autonomie pour les soins et les compétences d'adaptation aux situations de la vie quotidienne des parents, puis de l'enfant, indispensables pour retrouver une liberté d'exister, développer des projets de vie et préparer l'adolescent à devenir un adulte capable de se prendre en charge. La posture éducative de l'équipe, partant des connaissances et des représentations des parents, permet de les ajuster régulièrement en fonction des besoins de l'enfant et de travailler à la levée des freins pour la mise en place des traitements. Elle permet aussi à l'équipe d'obtenir une vision réaliste de la capacité des parents à assumer certaines prises en charge lourdes à domicile et de rechercher des solutions alternatives sur des aidants et de proposer des répits lorsque c'est nécessaire.

3. L'engagement dans un programme d'amélioration de la qualité des soins une nouvelle forme de coopération parents-soignants

L'histoire de la mucoviscidose en France témoigne du fait que l'engagement associatif des parents aux côtés des représentants des soignants pour améliorer la prise en charge a été la clé pour obtenir des avancées telles que la généralisation du dépistage néonatal et la création des centres spécialisés (CRCM). Cette

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dynamique se concrétise aujourd'hui au sein de la Filière maladie rare mucoviscidose.

Dans son CRCM, chaque parent acquiert une connaissance pratique du système de soins à travers l'expérience vécue avec son enfant et devient un expert de la vie quotidienne avec la maladie en s'appuyant sur toutes les ressources qu'il sait mobiliser : ses propres compétences, l'équipe pluridisciplinaire, les soignants en ville, l'entourage, les ressources associatives. Il évalue sa propre capacité d'analyse, d'adaptation et d'action : ai-je bien fait ? Suis-je intervenu(e) à temps auprès des bonnes personnes ? Avec en filigrane la question : suis-je capable de prendre soin de mon enfant ? Il approfondit progressivement ses connaissances sur la maladie de son enfant, évalue l'efficacité de certains soins, les risques et les effets secondaires. Cette expertise construite sur l'expérience peut nourrir des échanges réguliers avec les soignants sur la perception de l'efficacité du dispositif, permettre une réflexion croisant les points de vue et soutenir la relation de coopération autour des besoins de l'enfant. Le désir du parent d'améliorer la qualité des soins pour son enfant est « naturel » et constitue un moteur : faire mieux ensemble ouvre une perspective de collaboration positive respectueuse des capacités et des ressources de chacun.

Mais toute amélioration n'est pas qu'une affaire de « cas particulier ». Dans le cadre d'une démarche structurée de réflexion-action, nous constatons que des parents, parmi ceux qui ont pu prendre du recul par rapport à leur parcours personnel et pensent pouvoir contribuer utilement, sont prêts à s'engager comme partenaire des soignants pour améliorer la qualité de la prise en charge dans leur centre. Le programme qualité dans la mucoviscidose leur propose une telle coopération, qui vise à analyser ensemble les processus pour améliorer les résultats du centre en termes de santé des patients, de satisfaction des patients/familles et des soignants, et d'efficience. Ce programme, déployé à partir de 2004 par la *Cystic Fibrosis Foundation* dans les centres mucoviscidose américains [2], propose une perspective nouvelle de coopération entre parents/patients et soignants pour l'amélioration de la prise en charge comprise comme la production du « microsystème » de soins : « tout système est parfaitement organisé pour produire les résultats qu'il produit » [3]. Elle s'inscrit dans une éthique de non jugement, d'écoute des différents points de vue, de reconnaissance des efforts, de respect des responsabilités de chacun, mais sans renoncer à la levée des contraintes d'organisation ou de fonctionnement qui pénalisent la qualité des soins délivrés. Elle implique de la part des soignants la transparence des indicateurs du CRCM, dans le respect de la confidentialité des données personnelles de santé. La participation des parents à la démarche qualité devient alors très vite « une évidence et un atout ».

L'amélioration des pratiques vise notamment à trouver des modalités concrètes pour :

- Produire de l'interdisciplinarité au-delà de la juxtaposition de ressources pluridisciplinaires et favoriser une forme de leadership qui en permet l'exercice.
- Assurer les transmissions d'informations pour permettre de prendre des décisions éclairées ;
- Organiser les consultations pour réaliser les évaluations, actes de soins et séances d'éducation nécessaires dans les temps impartis.
- Développer une communication responsable entre soignants et parents/patients qui permette d'anticiper les décisions de traitement afin de prévenir le déclin ;
- Mobiliser et convaincre au-delà du microsystème pour adapter les ressources aux besoins du centre.

Au-delà de l'amélioration des processus de prise en charge, la démarche qualité construit une dynamique de coopération entre « des parents/patients informés et activés et des soignants préparés et proactifs » pour un système de coproduction des soins [4].

Remerciements

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Références

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L'arrivée des adultes en nombre croissant dans les CRCM adultes des Services de Pneumologie et leur investissement progressif dans l'association Vaincre la Mucoviscidose – à travers un conseil des patients adultes, la publication d'une *Lettre aux Adultes,* une participation accrue à des groupes de travail et une rencontre annuelle réservée aux patients adultes – modifie la sociologie de la communauté mucoviscidose, à la fois soignante et associative.

Les équipes des CRCM adultes sont organisées au sein de la médecine pour adultes, structurée par une haute spécialisation, alors que cette pathologie est caractérisée par un besoin de multi-spécialités et de coordination inhérente à une prise en charge morcelée. Les équipes doivent intégrer de nouveaux besoins relatifs d'une part à l'évolution de l'état de santé des patients, avec l'apparition de complications majeures, d'autre part à l'entrée dans la vie sociale de ces adultes jeunes, leur intégration dans le monde du travail, leur souhait de fonder une famille…

Rapidement, les adultes s'émancipent de leurs parents et s'autonomisent économiquement. Ils revendiquent une place de ***pilotes de leurs soins*** et de ***partenaires de l'équipe soignante*** dans les décisions qui les concernent, une ***voix*** dans le système de santé, le ***respect*** de leurs droits, notamment le droit d'accès à l'information, et ***l'accès*** à une vie sociale « normale ». Les adultes sont actifs sur les ***réseaux sociaux***, à travers des échanges et un soutien dans tous les aspects de leur vie et de la maladie. Des jeunes patients adultes se découvrent une motivation activiste, à l'instar de leurs aînés d'autres pathologies, dans tous les domaines de la formation des soignants et des patients, l'amélioration des soins et la recherche.

La démarche qualité collaborative en mucoviscidose propose aux patients adultes comme aux parents d'enfants malades de s'investir au sein de leur CRCM aux côtés de leur équipe soignante **pour améliorer la qualité des soins pour tous.**

III-3. La stratégie d'implémentation de l'intervention PHARE-M dans la filière mucoviscidose

Les stratégies d'amélioration de la qualité des soins héritées des expériences industrielles émergent au tournant des années 2000, adaptées au monde de la santé par le Harvard Institute of Healthcare Improvement à travers les Breakthrough Series (BTS), et se diffusent rapidement à la suite du constat de l'IOM publié sous le titre *Crossing the Quality Chasm, a new health system for the new century* (2001). De nombreuses publications ont fait état d'expériences d'application de ces démarches aux USA, et la recherche a construit un large champ de connaissances basé sur ces expériences (60).

Une telle démarche collaborative a été développée et adaptée par l'US CFF et le Dartmouth Institute pour les centres spécialisés de la mucoviscidose (61). Des « CF Collaboratives » sont déployés depuis 2002 par les experts de Dartmouth Institute sous la direction de l'US CFF dans les centres mucoviscidose américains (et à Toronto). Les objectifs en sont de diffuser et implémenter les recommandations de traitement basées sur les preuves (guidelines), d'identifier les pratiques de soins conduisant aux meilleurs résultats de santé des patients à partir de la comparaison des indicateurs de santé dans le Registre US et de visites de benchmarking des centres ayant les meilleurs résultats, dans le but de réduire les écarts de résultats entre les centres. Les constats établis aux Etats-Unis sur la variabilité de la prise en charge dans les centres et la disparité des résultats des indicateurs de santé des patients, étaient également observés en France, en plus des différences observées

sur les ressources disponibles dans les équipes pluridisciplinaires, notamment entre les CRCM pédiatriques et les CRCM adultes (57).

L'introduction de la démarche qualité américaine en France répondait donc à un besoin identique et s'inscrivait dans le prolongement de la structuration de la filière et la culture de collaboration entre les professionnels et l'association Vaincre la Mucoviscidose. La stratégie d'implémentation en France a ainsi mobilisé les acteurs « institutionnels » qu'étaient le CRMR-M de Nantes-Roscoff et l'association Vaincre la Mucoviscidose, avec une répartition des rôles et des ressources entre les deux présentant des similitudes avec le modèle d'implémentation de la démarche aux USA qui repose sur la fondation américaine et l'institut de Dartmouth. Toutefois, l'introduction de cette innovation en France a été source de tensions inattendues au cours de son implémentation, liées **au repositionnement des deux acteurs institutionnels** et à **la représentation directe de patients et parents** au niveau du microsystème clinique. Ces tensions sont apparues dès l'année pilote de lancement opérationnel de la démarche et ont conduit à la mise en œuvre d'un process d'implémentation évolutif en fonction des points d'appui trouvés au cours de sa progression.

L'enjeu de la place du parent coordinatrice du programme PHARE-M, entre bénévolat associatif et professionnalisation, a focalisé ces tensions.

a) Phase préparatoire de l'intervention PHARE-M (2006-2011)

Au cours de cette phase, le ***cadre du 1er Plan National Maladies Rares*** a permis de mobiliser les acteurs autour des missions transversales portées par les CRMR-M. A l'occasion de sa labellisation en 2006, le CRMR Mucoviscidose (CRMR-M) de Nantes-Roscoff a élaboré son plan d'action, affichant des orientations nationales, à développer dans les CRCM avec le soutien de l'équipe nationale du CRMR-M : le développement de l'ETP, notamment pour les adultes et les patients transplantés, la mise en place d'un programme d'amélioration de la qualité de la prise en charge, le développement des systèmes d'information de la filière, notamment l'évolution du Registre Français de la Mucoviscidose, le développement de la recherche en transplantation pulmonaire et en sciences humaines et sociales.

Ce plan d'action a été présenté et co-signé par les instances de la Filière – la Société Française de la Mucoviscidose, le 2ème CRMR-M de Lyon et l'association Vaincre la Mucoviscidose. Dans les suites de son officialisation, le médecin coordonnateur du CRMR-M de Nantes-Roscoff a réalisé un séjour de 6 mois aux USA pour se former à la démarche qualité au Dartmouth Institute, nouer des relations avec l'US-CFF et visiter des centres spécialisés mucoviscidose aux USA. A son retour en France et sous son impulsion, un groupe de travail national a été constitué en 2010, incluant des représentants de l'association, de santé publique et du CRMR-M de Nantes-Roscoff pour prendre connaissance du programme développé par l'US-CFF, examiner sa faisabilité en France et définir les conditions de sa transposition (formation à la méthode, relations avec l'US-CFF et le Dartmouth Institute, soutien financier de l'association à la formation, réalisation de la traduction/adaptation des supports).

Au cours de cette phase préparatoire, deux personnes se sont portées volontaires pour s'engager dans la transposition du programme en France, sous la direction du médecin coordonnateur du CRMR-M de Nantes-Roscoff :

- Une kinésithérapeute, membre à mi-temps du comité de pilotage du CRMR-M sur le site de Roscoff, déjà impliquée dans des formations à la kinésithérapie respiratoire auprès des soignants et des parents d'enfants et engagée auprès de l'association,
- Un parent d'adolescent malade, ingénieur de formation et ayant l'expérience des démarches qualité dans l'industrie, administratrice de l'association « détachée » sur cette démarche qualité ;

Les deux personnes ont à leur tour séjourné aux USA, pour suivre la formation de deux mois au Dartmouth Institute et visiter des centres spécialisés ayant mis en œuvre la démarche et obtenu des résultats remarquables. Au retour elles ont réalisé la traduction des outils de la démarche et préparé le lancement de la première session « pilote » du programme PHARE-M pour la rentrée de septembre 2011, sous la direction du médecin coordonnateur du CRMR-M.

L'introduction de l'intervention à travers la session pilote a posé la question de la place du parent-ingénieur entre sa position associative et la nouvelle position de coordinatrice du programme PHARE-M qui lui était proposée en raison de son expérience professionnelle antérieure et des compétences acquises. La vision associative requerrait un strict bénévolat (donc l'impossibilité d'un financement associatif pour financer cette position de coordinatrice) et aucun poste n'était ouvert dans le cadre du CRMR-M pour la coordination du programme. En conséquence, au moment du lancement de la phase pilote, la mission de coordination du programme a été confiée au parent sans financement associé et avec une exclusion des instances associatives (notamment du conseil d'administration) pour cause de « conflit d'intérêt ». Par comparaison, la coordination de la démarche aux USA est assurée par un parent dont la position est reconnue au sein de l'institution hospitalière du Dartmouth Hitchcock Medical Centre et financée par une subvention annuelle de la CFF depuis 2002.

Dès la préparation de l'intervention pour la session pilote, la participation d'un CRCM au PHARE-M était ***conditionnée*** par le ***recrutement*** d'un patient adulte ou d'un parent d'enfant et son ***engagement bénévole*** comme membre à part entière de l'équipe de pilotage de la qualité du CRCM, incluant sa ***formation*** à l'ensemble de la démarche qualité et sa participation aux réunions ***nationales*** et ***locales***. Par comparaison, le programme américain prévoyait la participation de patients ou parents dans les réunions locales des équipes de pilotage mais pas leur formation à l'ensemble de la démarche au même titre que les soignants. Leur contribution était néanmoins significative au cours des réunions locales, par les témoignages (story telling) et la formulation de leurs attentes. Certains étaient invités occasionnellement aux réunions collaboratives dans l'optique d'apporter leur témoignage sur des expériences vécues dans leur centre. Le dispositif de participation du patient ou parent à la démarche a été le seul écart apporté au design de l'intervention par rapport au modèle américain, l'objectif étant de rester fidèle à ce modèle pour maintenir sa cohérence interne et se donner toutes les chances d'observer, en France, les mêmes résultats positifs que ceux observés aux USA.

L'***équipe de pilotage*** constituée pour mener le programme PHARE-M dans le CRCM inclut ainsi 4 à 5 membres issus des différentes disciplines soignantes (infirmière, kinésithérapeute, diététicienne, psychologue, secrétaire…) autour d'un médecin leader, et d'un parent (pédiatrie) ou d'un patient (adulte). Des ***subventions associatives ont été attribuées aux CRCM*** engagés dans le programme, à hauteur d'un financement évalué à 0,20 ETP d'une infirmière coordinatrice par CRCM

pendant l'année de formation et des frais de déplacement des membres des équipes de pilotage (professionnels et parents ou patients) pour assister aux journées de formation à Paris. Cette mesure (sans aucune condition préalable d'adhésion des parents ou patients) a permis la participation des patients et parents à toutes les étapes du programme avec l'équipe de leur CRCM.

Cette étape préparatoire de l'intervention, dans le cadre institutionnel proposé par le plan maladies rares et avec des financements complémentaires associatifs, a abouti à la mise au point d'une version « test » de l'intervention fidèle au modèle de l'US CFF à l'exception d'une intégration systématique d'un patient ou parent dans l'équipe de pilotage aux côtés des soignants du CRCM. La stratégie d'implémentation a révélé des tensions entre les acteurs autour de la coordination du programme par le parent « professionnalisé », sans position ni dans l'association ni dans le CRMR-M.

b) Phase pilote (2011-2013)

La phase pilote a permis une collaboration forte entre l'équipe nationale du CRMR-M et les équipes des 7 CRCM volontaires pour tester et adapter le programme aux particularités du modèle de soin de la mucoviscidose en France et à la culture des soignants. L'éducation thérapeutique a été très rapidement intégrée comme un point fort du modèle français, et le développement de l'ETP au cours des venues des patients un enjeu de la réorganisation des processus. La coordination entre le kinésithérapeute du CRCM et les kinésithérapeutes qui suivent les patients en ville a été l'objet d'un travail systématique, incluant les patients adultes. A l'inverse, une opposition marquée des soignants aux parallèles entre les démarches qualité dans l'industrie et celles initiées dans les soins aux USA ont conduit à alléger la présentation de l'origine de la démarche des BTS construite par le IHI et à reformuler les principes d'optimisation des processus sans référence au contexte industriel.

Au niveau des « acteurs institutionnels », deux évaluations externes ont été mandatées en 2012 sur la session pilote :

- l'une, par le médecin coordonnateur du CRMR-M auprès d'un chercheur sociologue pour analyser l'appropriation par les équipes de la démarche qualité, les outils à améliorer, les ajustements à apporter en vue d'un déploiement national, les réactions à la participation des patients et parents dans les équipes ;
- l'autre, par l'association auprès d'un cabinet de consultants pour évaluer l'efficacité du programme et ses impacts au bout d'un an et décider de la reconduction ou non de son financement aux CRCM pendant l'année de formation.

Les deux évaluations indépendantes ont été très positives pour le programme et l'organisation nationale, malgré des recommandations d'ajustements entre certaines étapes de formation et d'intensification d'un « coaching » des équipes sur site.

Parallèlement à ces évaluations préliminaires, et dans le but de produire des connaissances scientifiques sur l'introduction et l'impact de ce programme en France, un projet de recherche portant sur la « performance de l'intervention à échéance de 3 ans (2015) » a été soumis par le CRMR-M à l'appel à projets du PRePS 2012 : il a été retenu et financé par le ministère (décision décembre 2012).

La décision de poursuite du financement associatif pour les équipes des CRCM qui s'engagent dans l'année de formation PHARE-M a été motivée par la sélection au PRePS 2012 du programme de recherche et attribuée pour la seule année 2012-

2013, afin de compléter le périmètre des centres inclus dans la recherche. La recommandation du consultant, reprise par le conseil d'administration de l'association, était de se séparer du parent et de confier la coordination du programme au cabinet sur une mission financée par l'association. Cette recommandation, reprise par le conseil d'administration de l'association, n'a pas été suivie par le responsable du CRMR-M qui a maintenu le parent dans sa fonction de coordination et obtenu une subvention auprès d'un autre organisme pour l'année suivante du programme. Un déploiement inter-régional (Rhône Alpes Auvergne et Bretagne Pays de la Loire) a été organisé (Sept. 2012 – Juin 2013) afin de valider la version ajustée du programme auprès de 7 CRCM supplémentaires et de constituer ainsi le périmètre des 14 CRCM inclus dans la recherche.

Cette phase d'introduction de la démarche (2011-2013) a abouti à un programme PHARE-M ajusté et consolidé en vue d'un déploiement national, un financement associatif de l'intervention insécure et un programme de recherche financé par le ministère pour établir la performance à 3 ans de l'intervention engagée dans les 14 CRCM. La position professionnalisée du parent coordonnateur du programme a été confirmée, en grande tension avec la vision associative de l'engagement des parents dans la communauté mucoviscidose. La participation des patients et parents aux équipes de pilotage a été permise par le remboursement de leurs frais par l'association, sans pour autant qu'elle soit en lien avec la représentation de l'association dans les instances locales (délégations territoriales) et sans reconnaissance de cette participation au sein de l'association.

L'introduction de la démarche a donc pris appui sur des forces d'opposition (notamment associatives) et de mobilisation (notamment des centres volontaires), induisant un clivage entre les pro et les anti programme PHARE-M – qui a conduit à la démission de la directrice médicale de l'association, soutien tout à la fois de l'ETP et du programme qualité. La sélection et le financement du programme de recherche par le ministère a permis la survie du programme PHARE-M.

c) Normalisation de l'intervention en un programme de développement professionnel continu (2014)

Le troisième temps est le temps de la normalisation du programme PHARE-M et de la structuration du programme de recherche. Au vu de la fragilité du financement de l'intervention, et suivant les orientations de la HAS publiées fin 2012, la décision a été prise par le CRMR-M de proposer le programme PHARE-M comme programme de développement professionnel continu (DPC) répondant aux directives de la HAS.

Extrait du site HAS :

https://www.has-sante.fr/portail/jcms/c_1348527/fr/developpement-professionnel-continu-des-professionnels-de-sante-la-has-presente-la-liste-des-methodes-et-des-modalites

A partir du 1er janvier 2013, les professionnels de santé devront satisfaire, tous les ans, à une obligation de Développement Professionnel Continu (DPC). Pour répondre à cette obligation, ils devront s'inscrire dans un programme annuel ou pluriannuel de DPC. La HAS vient de valider la liste des méthodes et des modalités de DPC. Cette liste précise les exigences méthodologiques portant sur les programmes, les supports utilisés, les intervenants et la traçabilité de l'engagement des professionnels.

Ce programme, dans son format définitif, a été accepté dès la phase transitoire du DPC et la session suivante (2015) s'est tenue dans le cadre de la formation continue hospitalière. Les agréments des deux commissions scientifiques indépendantes médicale et paramédicale ont été obtenus respectivement en 2016 et 2015, pour une période allant jusqu'en 2021.

Les soignants s'inscrivent pour l'année de formation sur le site de l'OGDPC, leurs frais de déplacement sont pris en charge et leurs absences du service sont justifiées. Le parent intervient dès lors au titre de formatrice-coordinatrice du programme PHARE-M auprès de l'institut de formation porteur du DPC. L'association est sollicitée pour les frais des patients et parents, et par exception, pour ceux des soignants qui n'auraient pas obtenu l'autorisation de s'inscrire au DPC.

Le déploiement national du programme se poursuit sur la base du volontariat des centres, et fin 2017 24 CRCM auront été formés à la démarche. Des ajustements sont intégrés d'une session à l'autre en fonction des profils de centres, afin de « *customiser* » les supports de cours aux priorités des soins en pédiatrie et en médecine adulte. Ces personnalisations sont prévues dans la démarche des BTS, à travers l'apport des recommandations et des bonnes pratiques de soins, adaptées à la pédiatrie ou à la médecine d'adulte.

La normalisation du programme PHARE-M dans le cadre de la formation continue hospitalière a permis d'assurer sa pérennité et sa diffusion auprès des établissements, en facilitant la participation des soignants et en valorisant leur participation. Elle a également donné un cadre institutionnel à l'équipe de formateurs et à la coordination du programme. Elle a permis de se dégager en grande partie de l'instabilité des prises de position associatives, qui semblent évoluer positivement au stade actuel de la formation de plus de la moitié des centres.

III-4. Design de l'intervention PHARE-M

L'intervention PHARE-M qui résulte de ce process d'implémentation consiste à installer, former et accompagner une équipe de pilotage composée de 4 à 5 membres de l'équipe pluridisciplinaire du CRCM, et un patient ou parent de la file active. Les membres de l'équipe de pilotage sont formés aux outils de la démarche qualité et à la conduite du changement dans l'organisation du CRCM.

La démarche PHARE-M repose sur les étapes suivantes :

- Constituer une équipe de pilotage de la qualité dans le CRCM, reflet de l'équipe pluridisciplinaire intégrant un patient ou parent (microsystème clinique), installer les conditions de travail de l'équipe pour la démarche qualité et communiquer avec les patients/parents et les autres services de l'hôpital en lien avec le CRCM sur le lancement de la démarche qualité PHARE-M,
- Comparer les indicateurs de santé entre les CRCM et par rapport à la moyenne nationale pour permettre à l'équipe d'identifier un objectif d'amélioration pour une population de patients et une échéance de quelques mois à quelques années,
- Analyser les causes de la situation actuelle et les leviers d'amélioration permettant d'atteindre cet objectif, sur les dimensions des patients, des professionnels, des processus, et des particularités du contexte,
- Intégrer des « idées de changement » issues des recommandations scientifiques et des bonnes pratiques repérées grâce à des analyses de benchmarking,

- Planifier des cycles de changement (PDSA) pour mettre en œuvre des actions d'amélioration et en mesurer l'efficacité sur les résultats cliniques ainsi que sur des résultats organisationnels,
- Mettre en œuvre les cycles de changement successifs jusqu'à l'échéance de l'objectif ;

PHARE-M se déroule en quatre grandes phases qui regroupent ces étapes au cours d'une session annuelle selon le curriculum ci-après (Tableau I).

Tableau I : Curriculum d'une session annuelle PHARE-M

Phase	Activité
Phase 1 : Structuration des équipes de pilotage	**Réunion d'information sur le programme PHARE-M**
	Structuration des équipes de pilotage des CRCM et inscription à la formation continue
	WebC : point d'avancement de la phase préparatoire
Phase 2 : Analyse du microsystème clinique	**EPE1 : Présentation de la méthodologie et des outils d'analyse (5P) & initialisation des analyses en pratique**
	Analyse du microsystème clinique par l'équipe de pilotage du CRCM
	WebC : point d'avancement des analyses dans les CRCM
Phase 3 : Planification des Actions d'amélioration dans le microsystème clinique	**EPE2 : Présentation des résultats des analyses, choix des thèmes d'amélioration et des objectifs chiffrés, examen des idées de changement et préfiguration des actions d'améliorations (cycles PDSA)**
	Structuration des Actions et Préparation de la communication
	WebC : point d'avancement de la définition des cycles PDSA
Phase 4 : Mise en œuvre des Actions d'amélioration selon les cycles PDSA et mesure des résultats	**EPE3 : Visite de Benchmarking, intégration des bonnes pratiques dans les Actions d'amélioration et revue des plannings de mise en œuvre des cycles PDSA**
	Mise en œuvre des premiers cycles PDSA et des indicateurs de mesure opérationnels
	WebC : point d'avancement de la mise en œuvre des cycles PDSA
	EPE4 : Présentation des Posters des équipes et des communications

Les principales adaptations apportées, au cours du processus d'implémentation, à la version initiale du programme et aux supports de formation (à l'origine fidèlement traduits du programme américain) sont présentées ci-après.

Elles correspondent à 1) une description des expériences françaises plutôt qu'américaines, 2) un renforcement de ***l'accompagnement sur site*** des équipes dans un contexte plus économe de ressources qu'aux USA (où l'accompagnement sur site est confié à des coachs formés au Dartmouth Institute et rémunérés par la fondation américaine à hauteur de 2 à 4 jours par mois pendant un an), et 3) un « ***alignement*** » avec la politique qualité portée par les départements qualité hospitaliers.

1 Une nouvelle version du « Guide d'Action pour accélérer l'amélioration de la qualité des soins en mucoviscidose » *a permis de substituer aux exemples de réalisations d'équipes américaines des exemples de réalisations des équipes françaises engagées dans la phase pilote ;*

2 *Les réunions de formation nationales ont intégré davantage de travail par équipe pour* ***mettre en application les notions théoriques*** *et le temps consacré à l'exposé de la théorie a été allégé ;*

3 *Une plus* ***large diffusion de l'outil intitulé "Registre, Outil de la Qualité (ROQ)"*** *a été organisée auprès des soignants et des patients/parents de tous les CRCM dans la perspective de préparer la communauté à s'engager dans les sessions futures du programme : ce document présente une vulgarisation des données du registre, explique leur utilisation pour la démarche qualité avec des recommandations d'interprétation des indicateurs, et illustre la contribution possible de chacun, professionnels et patients/parents, à l'amélioration de la qualité des soins ;*

4 Un rôle de "référent PHARE-M*" a été défini pour consolider la démarche sur site en impliquant plus fortement un soignant non médecin de l'équipe du CRCM, et lui confier des missions ciblées sur l'animation et le suivi des travaux de l'équipe, les relations avec le patient ou parent, notamment pour les aspects pratiques de sa participation aux réunions et le repérage de ses difficultés éventuelles, et la coordination avec l'équipe nationale : cette fonction est subventionnée par l'association Vaincre la mucoviscidose à hauteur d'un temps de 0,20 Equivalent Temps Plein d'infirmière pendant un an ;*

5 *Une incitation des équipes à solliciter* ***les départements qualité hospitaliers*** *pour obtenir un appui par un ingénieur qualité auprès de l'équipe, pour aider dans des analyses statistiques ou dans l'utilisation de certains outils généralistes de la qualité (diagramme des causes, cycles PDSA) ;*

6 *Un* ***accompagnement sur site*** *par la coordinatrice du programme PHARE-M, matérialisée par une visite (a minima) pour analyser les processus (patient shadowing [62]), participer à un staff pluridisciplinaire et une réunion de l'équipe de pilotage, discuter avec le patient/parent et résoudre des difficultés rencontrées par l'équipe avec la mise en œuvre de la démarche ; le cas échéant rencontrer les interlocuteurs du Pôle ou du département qualité pour faciliter la communication sur la démarche et obtenir des arbitrages ;*

7 *Refondre l'outil du* ***site web du PHARE-****M en supprimant l'outil de messagerie intégré et réservé aux équipes engagées (modèle de Dartmouth Listserv.) qui était sous-utilisée car redondante avec la messagerie électronique utilisée par les soignants et fournie par leur établissement ;*

L'article III (Volume 2) présente les étapes de l'introduction de la démarche qualité dans la filière mucoviscidose en France et illustre ***l'interdépendance entre l'implémentation de l'intervention et l'évolution du format de l'intervention***, dans le contexte des forces en présence soumises à la double « *pression* » d'une démarche d'AQ innovante et du rôle des patients et parents dans cette démarche.

L'abstract (*traduction française*) est présenté ci-après. L'article complet est intégré à la suite.

Introduction of a Collaborative Quality Improvement Program in the French Cystic Fibrosis Network: the PHARE-M* initiative

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Introduction

Une charte signée en 2007 entre les deux centres de référence maladie rare mucoviscidose (CRMR-M), l'association Vaincre la Mucoviscidose et les 49 centres de ressources et de compétences de la mucoviscidose (CRCM) en France, prévoyait l'engagement de participer, dans les 5 prochaines années, à un programme d'amélioration de la qualité des soins.

Objectif

Déployer dans la filière mucoviscidose un programme d'amélioration de la qualité des soins inspiré du programme américain développé par The Dartmouth Institute Microsystem Academy (TDIMA) et adapté pour la mucoviscidose par la Fondation américaine (US CFF) entre 2002 et 2013.

Méthode

L'équipe du CRMR-M de Nantes-Roscoff s'est formée au TDIMA et a visité des centres impliqués dans le programme qualité de la Fondation américaine, en vue de le transposer en France en traduisant le Guide d'action et les outils de formation. Une Session1 du PHARE-M†† a inclus sept centres en 2011 pour tester le programme dans le contexte français. Elle a fait l'objet de 2 évaluations externes. Des ajustements ont été effectués avant que la Session2 du PHARE-M ne soit déployée dans sept autres centres. L'accompagnement des équipes sur site a été renforcée. La satisfaction des équipes a été évaluée et quelques ajustements complémentaires ont été réalisés. En 2014, le programme a été déposé auprès de l'organisme de formation continue hospitalière pour demander sa reconnaissance comme programme de développement professionnel continu (DPC).

Résultats

Quatre-vingt-seize personnes, dont 14 patients et parents, ont participé aux sessions 1 et 2 du PHARE-M dans les 14 CRCM volontaires. La comparaison des indicateurs de santé des patients à partir du Registre par centre, l'analyse des meilleures pratiques de soins, la sélection par chaque équipe d'un thème d'amélioration prioritaire, la mise en œuvre d'actions et les échanges entre équipes ont permis de développer l'adhésion à la démarche. Le programme a amélioré la qualité des soins, notamment le fonctionnement interdisciplinaire, la pratique de l'éducation thérapeutique et la collaboration avec les patients et parents. La satisfaction des équipes a augmenté dans le temps. Un cycle post-PHARE-M a été mis en place à la demande des équipes pour soutenir l'amélioration continue de la qualité. En 2015, PHARE-M a reçu l'agrément de programme de DPC pour les professionnels médicaux et paramédicaux.

†† Programme Hospitalier d'Amélioration des Résultats et de l'Expertise en Mucoviscidose

Conclusion

PHARE-M est une intervention complexe dans les équipes multidisciplinaires des CRCM de divers contextes hospitaliers. Des facteurs multiples ont motivé les équipes à s'engager. L'implication des patients et parents et le développement de l'éducation thérapeutique ont contribué à soutenir la démarche qualité. La reconnaissance du programme par la formation continue hospitalière favorise sa pérennisation. La transparence des indicateurs du Registre par centre est nécessaire pour améliorer continuellement la qualité des soins. L'impact du PHARE-M sur les résultats cliniques des patients après 3 ans est l'objet d'un programme de recherche dont les résultats seront disponibles fin 2017.

Mots-clés : *mucoviscidose, amélioration de la qualité des soins, microsystèmes cliniques, démarche collaborative, maladies rares*

Article III

Introduction of a Collaborative Quality Improvement Program in the French Cystic Fibrosis Network: the PHARE-M initiative

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Abstract

Introduction

An agreement, signed in 2007 by the 49 French Cystic Fibrosis Centers, included a commitment to participate, within the next five years, in a care quality assessment and improvement program (QIP).

Objective

To roll out in the French Cystic Fibrosis (CF) care network a QIP adapted from the US program for Accelerating Improvement in Cystic Fibrosis Care developed by The Dartmouth Institute Microsystem Academy (TDIMA) and customized by the US CF Foundation between 2002 and 2013.

Method

The French national team at the Nantes-Roscoff CF Center of Expertise was trained at TDIMA and visited US CF centers involved in US Learning and Leadership Collaboratives (LLCs). It introduced the PHARE-M[‡‡] in France by transposing the Action Guide and material. A PHARE-M LLC1 including seven centers, underwent two external assessments. Adjustments were made, then a PHARE-M LLC2 was rolled out at seven more centers in two regions. On-site coaching was strengthened. The teams' satisfaction was assessed and further adjustments were made. In 2014, the program sought recognition as a continuing education program for healthcare professionals.

Results

Ninety-six trainees including 14 patients/parents from the 14 CFCs volunteered to participate, test and adapt the program during LLC1 and LLC2 sessions. Comparison of patient outcomes collected in the Registry report by CF center, reflection on potential best practices, selection by each team of an improvement theme, implementation of improvement actions, and exchanges between teams fostered the adhesion of the teams. The program strengthened quality of care, interdisciplinary functioning and collaboration with patients/parents at the centers. The satisfaction expressed by the teams increased over time.

[‡‡] Programme Hospitalier d'Amélioration des Résultats et de l'Expertise en Mucoviscidose – A hospital-based program for improvement of results and expertise in cystic fibrosis care

A post-PHARE-M cycle maintains the focus on continuous quality improvement (CQI). In 2015, PHARE-M was recognized as a continuing professional development program in healthcare.

Conclusion

The PHARE-M is a complex intervention in multidisciplinary teams working in a variety of hospital settings. A confluence of factors motivated teams to engage in the program. Involving Patient/Parent in quality improvement (QI) work and developing patient therapeutic education for self-management appeared to be complementary approaches to improve care. Incorporating the program into hospital continuing education insures its sustainability. Transparency of Patient Registry indicators per center published in a brief lapse of time is required to effectively support CQI. The impact of the PHARE-M on patient outcomes after three years is the subject of a research program funded by the French Ministry of Health whose results will be available in 2017.

Keywords: cystic fibrosis, quality improvement program, clinical microsystem, learning and leadership collaborative, rare disease, patient registry

Introduction

The follow-up of cystic fibrosis (CF) patients in specialized care centers has been shown as an independent factor for patients better outcomes and longer survival in patients [63; 64]. In the 21st century Quality Improvement Programs (QIPs) have emerged as new strategies to reduce variability of care and facilitate the implementation of best practices across centers. Following the publication in 2001 of the report entitled *Crossing the Quality Chasm* [65], the US CF Foundation (US CFF) launched a benchmarking study to analyze the differences in patient outcomes across the CF care network. This study highlighted differences in median survival between the 10 best centers and all other centers. The decision was made to design and implement Learning and Leadership Collaboratives (LLCs) with an overarching goal of delivering the best possible care to all patients and improving clinical outcomes [66]. This program was developed by the Dartmouth Institute Microsystem Academy (TDIMA) [67], then adapted, tested and implemented into the CF network starting in 2002 [[68]].

The cystic fibrosis care center network in France was formalized in 2002, following generalization of systematic newborn screening for CF, to deliver specialized CF care from the diagnosis to adulthood [[69]]. In 2006, the French National Authority Health published a CF Diagnosis and Treatment Protocol for CF [[70]]. The French National CF Observatory, modelled on the CF American Patient Registry questionnaire, was established in 1992. Its objective has evolved into taking a comprehensive census of the population [71]. It is now known as the French CF Registry [72] and was certified by the French National Committee of Rare-Diseases Registries in 2007. It is fed into the European CF Registry and contributes to European epidemiologic studies [73]. Within the framework of the first French National Plan for Rare Diseases, the French Ministry of Health designated two CF Centers of Expertise in 2006 to carry out national action plans across the CF care network. The Nantes-Roscoff Center of Expertise action plan featured the following priorities: health information and communication systems, therapeutic patient education, clinical research in the social sciences and transplantation, and a care QIP. An agreement prepared in 2007 and signed by the heads of all CF centers included a commitment to "*participate, within the next five years, in a care quality assessment and improvement program to be offered by the Centers of Expertise in collaboration with the French CF Society, the French Ministry of Health and patient organizations*."

Since 2006, communications at the North American CF Conference and the European CF Conference have reported successful experiences on the part of centers engaged in the US CF LLCs. At a conference in France in 2008 by the French CF patient organization Vaincre la Mucoviscidose and the French CF Society, results of the US LLCs on CF care and

patient outcomes were presented to an assembly of clinicians, care providers, patients and parents. A working group including representatives of the patient organization and of the Nantes-Roscoff EC was formed to reflect on a method for developing and implementing a QIP in France inspired from the US CF QIP. With the support of the CF Foundation, a training for the lead physician of the Nantes-Roscoff EC at The Dartmouth Institute as well as visits to centers engaged in the US CF QIP were organized in 2008. These confirmed the interest of transposing this program to France in order to benefit from this experience and reduce the time taken to develop a QIP in France [74]. A team including a parent (an engineer by training) and a physiotherapist was formed at the Nantes-Roscoff Center of Expertise. A presentation by the US QIP coordinator at the Vaincre la Mucoviscidose General Assembly (Reims 2011) was made to inform the French CF community of the importance and feasibility of such a QIP in CF care in France. Both the physiotherapist and the parent went to TDIMA for training and to US centers engaged in LLCs to observe the results achieved following the implementation of a QIP. This was made possible by a grant from the patient organization. Under the supervision of experts from Dartmouth and the CFF, the French team began the translation of the CF Action Guide and educational tools, registered on the Dartmouth CF network's collaborative website, and reflected on the resources needed to implement the program in France. When the program started in France in 2011, some differences between the two countries, such as certain characteristics of the French healthcare system and unique features of the French CF care model and the French cultural context, questioned the success of transposition of the program, the adherence by stakeholders and the achievement of results on the level reported by the United States.

The aim of this article is to report and reflect on the experience of introducing the PHARE-M†† QIP in France, between 2011 and 2015, through two annual LLCs leading to the standardization of the final program as a continuing professional development training program on the French hospital continuing education website. We present the factors that gained the teams' adherence, the synergies at work and the adaptations that led to the adoption of the program in the French CF network. Based on our experience, we discuss the elements that we believe to be essential in transposing this CF LLC QIP to the context of another country, since the European CF Society have paved the way for care quality improvement initiatives across the CF care center network in Europe.

Method

This QIP, designed according to the systematic approach described by Nelson, Batalden, and Godfrey [75], is focused on the clinical microsystem, which includes the multidisciplinary care team, patients and their family. The LLC QI format has been adopted by the CF Foundation in 2002 *to support the CF centers' work to reduce the variation in patient outcomes across the US network. This adoption included adaptations to the specificities of the care center network, such as local culture, patient population and multidisciplinary staff and the healthcare system in which it existed,* as described by Godfrey and Oliver [68]. The French program is derived from the 2011 US LLC program and benefitted from the experience with and customization of the program in the US CF care network.

French national team responsible for transposing of the US CF LLC

A French national team was formed comprising the lead physician at the Nantes-Roscoff Center of Expertise, his assistant, a parent of an adolescent with CF (an engineer by training), a physiotherapist and the head of information and communication system projects. The physician, physiotherapist and parent had been trained in a quality course at TDIMA, and had visited several CF centers involved in the CF LLCs for years [74]. The physician in charge of the French national therapeutic patient education program (TPE) and director of the pediatric CF center in Nantes, was closely associated with the team and led its testing at her center. This team is hereinafter referred to as the "national team". Due to its composition, the national team included two main features unique to French CF model of care: 1) the CF therapeutic patient education program, validated in 2005 by the French health authorities and structured according to developmental stages in children and needs in terms of management of complication in adults (http://etp.centre-reference-muco-nantes.fr), and 2) respiratory physiotherapy care, delivered to patients at home according to the French National Diagnosis and Treatment Protocol and reimbursed by the French national health insurance system. The national team also strongly emphasized the involvement of patients and parents in the QIP at each center. A recruitment procedure was put in place to identify in the patient caseload at each center individuals with CF or parents of children with CF who were motivated, available, at ease in their relationships with professionals, capable of self-expression in a group, able to communicate via Internet with the team. The patient or parent was enlisted as a full member of the local quality improvement team and their travel expenses were reimbursed by the patient organization Vaincre la Mucoviscidose.

Transposition of the US CF LLC into a first version of the PHARE-M LLC

Training materials were provided free of charge by the US CFF and access to TDIMA's electronic resources was authorized. Resources were developed before the program started in France (September 2011). They included:

- the translation of training materials, including the Action Guide for Accelerating Improvement in Cystic Fibrosis Care [76] under a Dartmouth Director supervision;
- the drafting of a French national report entitled "Registry, a Tool for Quality Improvement" (RTQI), to inform patients and parents and present the usefulness of the French CF Registry to assess improvement on patient outcomes; "The 10 Goals of the PHARE-M" (see Box 1); and an itemization of each goal with the respective roles in a for care improvement partnership to be played by the patients, their family and the healthcare providers;
- the creation of a website dedicated to the PHARE-M [77] containing tools, training materials and updates and serving as a messaging tool dedicated to the teams engaged in the PHARE-M; and
- the selection of a web conference tool for remote training meetings.

Box 1: The 10 Goals of the PHARE-M

1 Parents and patients are full partners of the healthcare team. Each patient/family has a right to clear and understandable information.

2 Each patient, regardless of his or her geographical, social, and cultural circumstances, enjoys effective multidisciplinary care.

3 Each patient/family has a right to therapeutic education to aid in acquiring or strengthening the skills required to best manage life with cystic fibrosis.

4 Patients grow normally and have a normal nutritional status.

5 Respiratory infections and exacerbations thereof are detected as early as possible, and appropriate treatments are started without delay.

6 Physical and sports activities are encouraged from an early age and adapted to each patient throughout his or her life.

7 Suitable measures are put in place and hygiene advice is given to prevent cross-contamination.

8 Complications, including diabetes, are diagnosed and treated early.

9 All patients who progress to a state of severe respiratory failure are informed of their therapeutic alternatives, then either supported in their decision to undergo transplantation or accompanied at the end of life.

10 Post-transplant care aims at sustainable improvement in quality of life and in physical, psychological, and social health.

The Pilot PHARE-M LLC1 (September 2011 – June 2012)

The PHARE-M LLC1 enrolled 7 volunteer centres, including four CF centers from the two national French national Centers of Expertise of Nantes-Roscoff and Lyon, thanks to close professional networking. A multidisciplinary "quality improvement team" was formed at each center included a physician leader, four to five professionals and a parent or a patient. Vaincre la Mucoviscidose agreed to reimburse the travel fees of the teams – including those of the patients/parents – and give each center a grant covering a 0,20 FTEs for a nurse for one year, corresponding to the extra time required for data analysis and teamwork management.

Four Face-to-face LLC meetings were organized. At these meetings, theoretical presentations of the method illustrated with examples drawn from the American teams were alternated with practical exercises by the French center teams. Each team analyzed its patient outcomes and selected a theme for improvement for a target patient population. Patient data was available for each center from the 2009 Patient Registry report by center; however, some indicators presented weaknesses such as body mass index (BMI) being expressed for children as an absolute value and not as a percentile or Z-score. This forced the teams to collect specific data from their patient electronic records. The teams were offered Action Guide tools (satisfaction surveys, activity analysis grids, communication tools, etc.) and took advantage of the opportunity to adapt them to their setting. International experiences published in the literature were presented [78;79] and the teams were reminded of CF care guidelines [80]. Each team identified actions to redesign its processes, in line with its theme for improvement, to be tested according to successive PDSA cycles. The teams' satisfaction and suggestions were recorded at each meeting and an overall score was displayed on the PHARE-M website.

Close collaboration with the TDIMA and the CFF was sustained over the course of LLC1 through:

- the participation of members of the national team, as well as physicians at several pilot centers, in the adult LLC session at the North American conference in Anaheim (October 2011);
- the participation of the Director of TDIMA Clinical Microsystem Group in the third face-to-face meeting to supervise the poster session meeting (PHARE-M LLC1, Marseille, March 2012);

- the trainings for the physiotherapist and the parent on the national team in the TDIMA's "eCoach the Coaches" course at the same time as the PHARE-M LLC1.

Assessments of the pilot PHARE-M LLC1

The PHARE-M being an innovative approach to QI in France, some key stakeholders were dubious as to its applicability in the French CF care network. The head of the Nantes-Roscoff Center of Expertise asked a Nantes Mines Engineering School sociological researcher to perform a first assessment of the program to analyse the factors for its success and barriers to its adoption, and the patient organization asked a consulting a firm to perform a second assessment to inform its decision as to whether to continue to fund the program.

The first assessment took place during LLC1. The assessor participated as an observer during two web meetings and the third Face-to-Face meeting. The assessment included familiarization with PHARE-M documents, interviews with a panel of professionals and patients/parents on the quality improvement teams, an interview with the members of the national team, an interview with the Director of TDIMA, and a visit to one site. All interviews and focus groups were recorded and fully transcribed. The data was exploited (coding, categorization), processed (analysis, validity) and interpreted according to the standard thematic content analysis protocol (Miles & Huberman, 2003 [81]). This was followed by manual grouping and counting within an analysis framework with the following dimensions: process applicability (terminology, formalization, tools, distance web meetings); incorporation of patients and parents (roles, time spent, barriers); national/regional coordination (roles, nature of support, incorporation mechanisms); process adoption (perceived benefits and costs, working atmosphere, engagement, acquisitions); and impact (operation, working practices, cooperation with the stakeholders). The report was submitted in July 2012 for consideration to adjust the PHARE-M LLC2.

The second assessment was contracted at the end of LLC1 to evaluate the effectiveness of this QI method in France, and to perform a comparative analysis between aims and outcomes achieved (efficiency) and between actions performed and expenses (efficacy). The study methodology included: familiarization with the PHARE-M documents and the literature on CF (French National Diagnosis and Treatment Protocol, French National Registry, etc.); investigations into four engaged CFC sites (Versailles, Lyon pediatric, Reims, and Roscoff) with professionals and patients/parents; telephone interviews with the members of the national team and patients/parents. The report was submitted during the October 2012 meeting of the board of directors of the patient organization, and the decision as to whether to continue funding was voted on in December 2012.

Main adjustments in the PHARE-M LLC2

Following these two assessments, the national team made adjustments to the program, thus further customizing the second version of the PHARE-M (see Box 2). The patient organization continued to fund the travel fees of the teams and the extra-time worked by a referent professional on the team at each center. No funding was allocated to the national team for intensive coaching of the teams at each center.

Box 2: Main adaptations in the PHARE-M LLC2

1 Drafting of a second version of the Action Guide illustrated with examples from the French teams in LLC1 instead of examples borrowed from the American teams;

2 Reduction of certain theoretical presentations in the training materials in favor of more exercises during face-to-face meetings;

3 Updated and revised version of the RTQI with was more systematically offered to patients/parents and professionals, either in its entirety or as separate chapters focusing on the goal chosen by the team at the center;

4 Formalization of the "PHARE-M referent" role on each quality improvement team, for a non-physician professional subsidized by the patient organization;

5 Incentive to enlist a quality engineer from the hospital quality department on the quality improvement team at the center, this professional sometimes becoming the PHARE-M referent;

6 One on-site coaching of the team at each center, offered during a visit by the program coordinator and focusing on mapping the clinic process with the “Shadowing a Patient” method [82]; and

7 Simplification of the PHARE-M website by withdrawing the PHARE-M specific messaging tool for the teams engaged in the PHARE-M as they did not use it in addition to their existing messaging tool.

Inter-regional rollout of the PHARE-M LLC2 (September 2012 – June 2013)

A second PHARE-M LLC session was planned to enroll the centers in the two French inter-regions of Rhône-Alpes-Auvergne and Grand-Ouest belonging to the regional care network of the two CF Centers of Expertise of Nantes-Roscoff and Lyon that could not have been included in the first session.

The teams' satisfaction and suggestions were recorded at every face-to-face meeting and web conference during LLC2. They led to two more adjustments to the training material:

- rearrangement of the content of the third and fourth face-to-face sessions by moving up the benchmarking visit and delaying the poster at the end of the LLC session; and

- strengthening of the link with TPE, underlying the importance of programming time for educational sessions during the clinic visit, focusing on the improvement goal and particular needs of the patient.

The teams also requested that a "post-PHARE-M cycle" be established to maintain a focus on quality improvement and have CFCs continue to exchange experiences after the LLC until they achieved their goal for improvement (two to three years after the training year). This was discussed with the patient organization for purposes of obtaining additional funding to organize an annual CQI meeting at a CF center for benchmarking and sustaining QI work.

Standardization and sustainability of the PHARE-M

The growing difficulty of enlisting new CFCs and the risk of jeopardizing patient organization funding led the national team to conceive of different avenues for perpetuating the PHARE-M and its rollout throughout the CF network.

First, a research project was drawn up in an attempt to respond to the recurrent request for evidence of the PHARE-M's positive impact on patient outcomes. The PHARE-M Performance project was submitted at a call for projects by the French Ministry of Health in February 2012. The project was selected by the Ministry on 5 December 2012 and funded for a three-year study. Its protocol combined a quasi-experimental evaluation of the effectiveness of the program to change patient outcomes over the course of three years with a process evaluation [83]. Following a realistic approach, the latter was designed to understand what works, for whom and under which circumstances (context) [84]. The success of the PHARE-M performance project at this call for projects was seen as a means to give credibility and recognition to the PHARE-M as well as funding to the national team for further interventional research.

Second, systematic efforts were made to incorporate the PHARE-M's into hospital accreditation process. The announcement of certain professional practice evaluation (EPP) actions for improvement and the participation of a hospital quality engineer on the quality improvement team at several centers were actively sought to improve the acceptability of the program in hospitals alongside more traditional certification methods.

Finally, continuing professional development in the field of hospital continuing education, which started in 2013 [85;86;87], offered an opportunity to standardize the PHARE-M into a hospital continuing education program without modifying its content or curriculum except to have it take place during a calendar year (January through December). Recognition by the hospital continuing education authority of the PHARE-M as a CPD program was sought as it was key to further roll-out.

Results

Results of PHARE-M LLC1 & LLC2

Seven centers volunteered to test and propose improvements to the program in the PHARE-M LLC1: four pediatric centers (Lyon, Nantes, Paris Robert Debré, and Versailles), one adult CFC (Lyon), and two pediatric teams at mixed centers (Reims and Roscoff) following up a total of about 1,200 patients out of the 6,500 patients in the Registry in 2011. Seven more centers from the two French inter-regions of Rhône-Alpes-Auvergne and Grand-Ouest engaged in the PHARE-M LLC2: three pediatric centers (Angers, Grenoble, and Rennes), two adult centers (Nantes and Rennes), and two mixed centers (Clermont-Ferrand and Morbihan), to which the adult team at the Roscoff center was added, following up about 800 more patients.

Ninety-six trainees from the 14 CFCs participated in the two annual PHARE-M sessions. More than half of the participants (54%) belonged to the multidisciplinary "core" team and 15% were patients or parents of patients. Healthcare providers on the quality improvement teams represented a total of 75 people, patients/parents represented 15 people, and non-healthcare professionals represented six people. Psychologists and dieticians were particularly strongly enlisted to the quality improvement teams (9/75 (12%) and 7/75 (9.3%) respectively).

Among those 14 centers (out of 45 CF care centers in France), three elected a theme for improvement related to adult care, one chose a theme related to transition to transplantation, one chose a theme related to transition to adult care, and nine chose a theme related to either respiratory or nutritional pediatric care. Four of them worked closely with the Quality Department at their hospital. Companion articles in this supplement present the changes in processes and clinical outcomes achieved in some centers between 2012 and 2015 and the links developed between the program and the general quality process at the hospital [88;89;90]. They show that working in QI has allowed these teams to achieve their goals and even exceed them on various themes of improvement such as FEV1 for adolescents, BMI for children 2 to 12 y.o. or time on the lung transplant waiting list. The statistical analysis of the PHARE-M Performance research project, which will assess the effectiveness of the program to change patient outcomes at centers involved in LLC1 & 2, will be performed on the Registry data from 2011 to year 2015 and results will be available by the end of 2017.

The assessment of the teams' satisfaction showed an increase between LLC1 and LLC2, as expressed at each training meeting and for the LLC overall, reflected in the median of all the participants' scores on a scale from 0 to 10, where 10 represented maximum satisfaction (median score = 7.48) and the LLC2 (median score = 8.16).

The final PHARE-M curriculum is presented in Box 3.

Box 3: PHARE-M Curriculum

Phase	Activity: 44 hours, 32h face-to-face meetings, 8h web conf. **ESE:** expertise and sharing of experience face-to-face meeting **Web Conf.:** remote conference organized via internet **PDSA:** plan-do-study-act
Phase 1: Organization of the quality improvement teams at the centres	**Information meeting on the PHARE-M**
	Organization of the quality improvement teams at the CFCs and enrollment in continuing education
	Web conf.: progress report on the preparatory phase
Phase 2: Analysis of the clinical microsystem	**ESE1: Presentation of the methodology and analysis tools (5Ps) and initialization of the analyses in practice**
	Analysis of the clinical microsystem by the quality improvement team at the CFC
	Web conf.: progress report on the analyses at the CFCs
Phase 3: Planning of the actions for improvement in the clinical microsystem	**ESE2: Presentation of the results of the analyses, selection of the themes for improvement and quantified objectives, examination of the ideas for change and foreshadowing of the actions for improvements (PDSA cycles)**
	Organization of the actions and preparation of the PDSA
	Web conf.: progress report on the definition of the PDSA cycles
Phase 4: Implementation of the actions for improvement according to the PDSA cycles and measurement of the outcomes	**ESE3: Benchmarking visit, incorporation of best practices into the actions for improvement, and review of the schedules for implementation of the PDSA cycles**
	Implementation of the first PDSA cycles and operational measurement indicators
	Web conf.: progress report on the implementation of PDSA cycles
	ESE4: Presentation of the teams' posters and presentations

At the teams' request, two post-PHARE-M cycles were offered in 2014, one pediatric and the other adult, consisting of one meeting per year at a CFC, including a benchmarking visit, an account of the progress and outcomes of the teams' actions, exchanges between the teams, and reminders fundamental aspects of the QIP.

Thirteen teams prepared their poster at the end of the PHARE-M session, and these posters were presented at the 1st CF Francophone Conference (2014). Three posters and their updates after three years were presented at the European CF conference (2012, 2014 and 2015) and the North American CF conference (2012). Videos featuring best practice recommendations concerning respiratory physiotherapy, physical and sports activities were prepared.

Improvement of the patient Registry

The French Registry contains one value in a given year for patient health outcomes and long-term treatments, while patient data are recorded at each clinic visit in the electronic patient record within the hospital information system. The Registry Committee establishes rules to select the clinic visit in a given year from which the FEV1, height and weight values are taken to be transmitted to the Registry.

In 2011-2012, the histograms presenting the median values of the centers remained anonymous in the Patient Registry report by center. The transparency brought in the PHARE-M meetings opened up discussions between the teams, leading them either to focus on the themes of improvement when the centers presented unsatisfactory results compared to national median values, or to question the measurement processes at the center. An on-site quality audit of the data transmitted to the Registry was organized in 2014-2015 pointed to variability in the measurement processes and in the application of the selection rule [91]. Avenues for improvement have been identified to support quality improvement of the data transmitted to the Registry by the centers.

To respond to the requests were made to the Registry team, the body mass index (BMI) for children was presented in Z-score value for LLC2. The lag between the year to which the data refer and the time of publication of the report (approximately two years in 2011) led the teams to supplement the Registry data with more recent data pulled directly from their patient records. The 2015 Patient Registry report has been issued by the end of 2016 and then provide more actual data for the PHARE-M LLC5.

Sociological assessment of PHARE-M introduction

The assessment pointed to themes related to cultural acceptance of the PHARE-M at the time of its introduction:

1) the progressive adherence by the teams at the centers to the different steps of the program, taking into account initial feelings of resistance towards administrative hospital quality processes and the associated system of formalization. Putting patient outcomes at the different centers into perspective sparked interest in the process and clarified its purposes. The rapid consensus reached on the priority theme for improvement and the preparation of the poster were unifying;

2) the successful organization of the PHARE-M project, i.e. at national level (program coordinator and program management) and at local level (quality improvement team). However, on the local level, the specific difficulty and required skills of the “referent” position suggested that the role of the “referent” should not be taken by the physician in the quality

improvement team and that the functioning of the physician leader/referent tandem is essential for the dynamic of the team.

3) the innovation consisting of patient or parent participation on the quality improvement teams, alongside their care providers, and their presence at the national face-to-face meetings as well as several local meetings was well perceived [92].

4) the gains for the functioning of the center teams were identified:

- a "collective enlisting of the team" for a unifying, energizing project for which the team learns to work together on what can be improved, thereby creating a "professional dynamic" in which professionals give new meanings to collective and profession-specific work practices;
- "reflexivity" on practices and relationships with patients/parents;
- a "calling into question" of care processes in front of other teams and transparency of outcomes, which may be sustained in a spirit of humility and desire to improve
- a "chance to speak" for all participants, which was possible in the melting pot of the face-to-face meetings;
- "rationale work" around the tools and processes, which objectivized and formalized practices and established a discourse to patients and parents;
- "dissemination" among the teams regarding quality management and tools;
- a "small-gains approach," which allowed pragmatic actions to be implemented with often limited resources and outcomes to be measured to consolidate practices.

The assessment for the patient organization funding recommendations

The consultant highlighted factors related to the feasibility and satisfaction regarding the PHARE-M training year:

- the 5P diagnosis phase faced challenges of feasibility within the training year with respect to 1) analysis of patient data, as Registry indicators were published with a two-year lag and BMI was expressed as an absolute value and not as a Z-score, and 2) analysis of patient satisfaction, as it took longer than expected for patients and parents to return their responses to the questionnaire*;*
- acceptance of the method was overall good, with the teams affirming that they were able to use the tools effectively and will be able to continue to do so beyond the training;
- team satisfaction was high concerning the consensus choice of a theme for improvement, the ability to comment on how they dealt with their work at sometimes difficult times (departures and reduced team), and the enlisting of the team around a joint project to improve patients' outcomes; and
- implementing the actions at the centers met with several difficulties: the building of a consensus on the choice of priority and feasible actions, for example, therapeutic patient

education, which does not always build a consensus on the teams; the availability of the resources to perform certain actions, for example, dieticians who cannot always be enlisted to abide by reconfigured care processes; cultural differences between teams that acted as obstacles to disseminating potential best practices.

Finally, the consultant assessed the effectiveness of the program (see Box 4) and concluded that PHARE-M mainly impacted care quality by allowing centers to use existing resources and innovative actions to comply with CF care recommendations, and that such an impact on quality of care should improve other aims, including the partnership with families and patients, provided that the patient organization support is strengthened.

Box 4: Training's effectiveness after one year assessed according to four criteria

1) sustainable care improvement: **high,** due to adoption of perpetuated tools or practices;

2) improvement in patient health outcomes: **weak after one year**, except in a limited sample of patients included in the new process of care related to improvement actions;

3) development of professional expertise: **average**, especially when there was a slow start; and

4) development of a partnership with patients/parents and care providers: **limited** to the patients involved in the new process of care.

Clinic visit process redesign

During the on-site coaching visits, the clinic visit process was analyzed at most centers by the program coach coordinator according to patient shadowing and process mapping. Multidisciplinary team (MDT) staff meetings, at which patients' situations and treatment plans were determined, were also analyzed. Observation of the multidisciplinary consultation process enabled identification of seven key steps of an "optimal" process (Figure 1) and description of the tasks corresponding to each step (Table I).

Implementation of the process first of all depends on the configuration of spaces. It also incorporates a therapeutic patient education session into the visit. It is linked to multidisciplinary staff meeting at which team members exchange information and hold discussions to ensure that the patient receives genuinely interdisciplinary care and that essential organizational aims are achieved: i) anticipating the consultations scheduled for the following week and having the professionals confirm their planning for these visits by specifying their aims for the patient; ii) drawing conclusions on the situation of the patients seen in the past week and establishing actions to be coordinated before the next visit by the professional in charge of monitoring them; and iii) preparing the visit report and scheduling the next visit.

Most coaching visits pointed out difficulties in sticking to this optimal process. At several centers, there was not enough time to review the situation of all patients seen the past week; as a solution to this problem patients having had an Annual Review or patients with specific needs were prioritized. It was sometimes difficult to get the entire MDT to meet at the same time. Patient records could not always be displayed during the staff meeting. Time was wasted on sharing data rather than making decisions. Effective meeting skills were developed and actions were taken according to a Professional Practice Evaluation process in order to improve the clinic visit process and the staff meeting.

PHARE-M standardization into a CPD program

The PHARE-M was approved as a multidisciplinary CPD program in 2014, and the 2015 PHARE-M LLC3 could be offered as a CPD program (see Box 4).

Box 4: Features of the PHARE-M CPD program

1 The PHARE-M as a CPD program received the approval of the Medical and Paramedical Independent Scientific Committees and will be re-evaluated prior to the extension of this approval (2021); formalized evaluation of each PHARE-M annual session is the responsibility of the hospital continuous education authority.

2 The training center at the Roscoff Foundation runs the PHARE-M CPD program, and the teams' registration fees provide the national team resources to continue to assess, improve and up-date the program and its website.

3 An annual request for application from the director of the Roscoff Center of Expertise, sent in May, invites and reminds the centers to register for the PHARE-M on a volunteer basis; an information meeting is organized in October to present the program and provide documentation to hospital continuing education directorates and quality departments.

4 The professionals on the team at the centers take administrative steps at their hospital to apply for the multidisciplinary PHARE-M CPD program to register for the next year and earn further CPD credits; the professionals on the CF team who are registered must include a lead physician lead and four to five multidisciplinary professionals.

5 The professionals on the teams at the centers are authorized to be absent from their posts for CPD training meetings, both face-to-face and web meetings, and another professional should replace them in their absence.

6 The professionals on the teams at the centers are reimbursed for their travel fees by hospital continuing education.

7 The patient organization is asked to reimburse the travel fees of the patients/parents and for the professionals unable to register to the PHARE-M CPD program.

8 The patient organization is continuing to fund 0.20 FTEs for the extra-time required for a PHARE-M referent on each team during the training year.

Discussion–Conclusion

The PHARE-M represented a "complex intervention" in clinical microsystems embedded in hospital systems marked by their diversity, their constant evolution, and the current economic pressure on the health care system. The various aspects of the program, essentially putting patient outcomes at the heart of quality improvement efforts and involving patients and parents on the quality improvement teams, led to a rapid consensus on the priority theme for improvement and identification of improvements on the process of care. Barriers linked to cultural differences between the United States and France were overcome by “Frenchifying” the Action Guide and the training material. This went beyond translating them into French, and involved searching for synergies with the quality departments. The PHARE-M contributed to the hospital certification process, and thanks to hospital continuing education reform, it was recognized as a multidisciplinary CPD program.

Limitations of the program roll-out

The pace of the roll-out of the PHARE-M throughout France could be accelerated by identifying sources of leverages. This would require professionals and patient organization representatives to pool their efforts (Box 5).

Factors for success in replicating the US CF LLC program

Developing an understanding of the initial model of improvement…

The 2011 Dartmouth and CF LLC model included involving patient and family on CFC improvement teams, using standardized evidence and practice-based ideas for change, preparing regular CF center progress reports, coaching teams, actively using the Patient Registry and applied measurement, and getting to know patients and families through observation and inquiry skills [68]. The following actions laid the foundations for an in-depth understanding of the method and its effects and dynamics: training the physician leader, the physiotherapist and the parent engineer on the national team at the Dartmouth Institute, giving them the opportunity to closely observe US CFCs with a long history of engagement in LLCs, increasing their awareness and energizing them through participation in several US LLC face-to-face meetings at the annual North American CF Conference, and training the parent to the “Coach the coaches” course. The method cannot be learned in its entirety from books, and the practical experiences of the US centers were enlightening. The supervision of the translation by the Dartmouth Institute and the CFF ensured that the training material

initially conformed to the improvement model. The humility of the national team, who recongnized its inability to understand the whole QI approach in depth through training and visits to centers alone, led them to stick to the US Action Guide and training materials during the French LLC1.

… And then adapting the model to the French context

Inevitably, the first LLC had to face the cultural gap between the US and France. This would have led to a great deal of conflict had the national team not anticipated cultural shock and asked the teams to help adapt the program to the French context. Opening up this opportunity decreased the tensions which arose as much from the program as they did from existing frustrations towards the hospital system: burdensome administrative quality procedures, economic pressure on the teams, inadequate facilities, and insufficient resources in every discipline in the CF team compared to standards of care were some of the issues that made the teams uncomfortable with the program.

The modifications made to LLC2 consisted mainly of replacing examples from US teams with examples from French pilot teams in the Action Guide and simplifying some of the theoretical presentations that the pilot teams had rejected, such as the reminders of QI in industry (e.g., process optimization steps) and statistical measurement techniques (e.g., control limits). On-site coaching was intensified and focused on patient shadowing and process mapping, which appeared to be more relevant and usable for the teams. After three years, as the teams engaged in LLC1 and LLC2 were invited to report their results, measurement became a new priority. This topic was addressed in post PHARE-M cycles while writing for publication was envisaged and SQUIRE guidelines were presented.

Performativity of the process initiated with the PHARE-M

All processes pertaining to care quality are evaluated and judged by the professionals with respect to their performativity[§§], that is to say, their contribution by acts that bring about the reality uttered by this process. "*When the players started to prepare and produce their data and their poster, to exchange and compare experiences, the performative capacity of the PHARE-M was perceived and legitimized. The performativity of the action guide was revealed and rationalized in the eyes of the participants on the teams after a few months, when the results that they had presented and debated highlighted the method's organizing nature".* The salience of the outcomes that are put in perspective, the feeling of having

[§§] The notion of "performativity," borrowed from linguistic pragmatics, shows that the medical and healthcare sciences in particular, in the case examined here, and the sciences in general, are not limited to representing the world: they also make it, cause it, and form it, at least to a certain extent and under certain conditions. In linguistics, an utterance is said to be performative when it establishes that of which it speaks. Extended and adapted to the sciences, this insight allows the classification of situations in which the subject of a methodological work is not merely observed or described, but modified or even called into being.

reinvested in care tasks, and the perception of producing and thinking differently most precisely characterize the program's performance. The medical and healthcare population generally had a negative conception of the quality engineering movement. Its culture is the very opposite of the medical, clinical, and healthcare culture which, from the outset, conceives of quality as something incorporated into individual practice, not something existing outside of individual practice or tied to an organization. PHARE-M partially reconciled these two visions.

On-site coaching

The recommendation concerning the strengthening of on-site coaching was verified to be operative during LLC2, with the establishment of visits by the coach coordinator, which at once allowed process mapping to be performed and organizational problems to be addressed. Team coaching was underlined as the most effective measure to develop the capability for improvement of the multidisciplinary teams at the centers [68]. However, this undertaking is costly and could not be offered to the centers during LLC1, as no specific funding had been obtained from the patient organization. Following the assessment, some funding was offered for LLC2 through a specific grant from the Foundation ildys. This grant acted as an investment in the future development of the PHARE-M as a CPD program supported by the training center at the foundation: on-site coaching could be offered, but not at the level achieved in the US. To compensate for the lack of on-site coaching, it was decided to develop the skills of one member of each CF team, referred to as the PHARE-M referent, and to search for synergy with the hospital quality department.

Synergy between therapeutic patient education and patient/parent involvement in QI

Therapeutic patient education in cystic fibrosis has been developed in French CF care, especially at pediatric centers, as it was recognized by law in 2005 as a right for persons suffering from chronic diseases. In practice, it establishes a lasting alliance between the healthcare team and the patient/parent with a view to developing the latter's autonomy and adaptation skills, adjusting them regularly as their needs evolve, and working to remove obstacles to establishing treatments [93]. On the PHARE-M side, the national team fostered patient and parent involvement as a pre-requisite for participation in the program, integrating them as members in the quality improvement team at their center as members so that they would contribute the user's point of view to QI and potentially co-design care processes [94,95]. This convergence between the two dimensions of patient involvement, in self-care and in the process of care redesign, was innovative in 2011 in France, based on the experience of the national team experience rather than on science.

More specifically, the national team fostered links between care improvement actions and educational interventions during the care process. The participation of the patients/parents on the quality improvement teams made it possible to ensure that their preferences and experiences were taken into account when new processes were proposed or care was intensified (nutritional care). Furthermore, within the framework of the PHARE-M, therapeutic education actions were strengthened as sources of leverage to improve home care and thus improve patient outcomes. Prioritizing certain health aims led to priority education actions. Reorganizing multidisciplinary clinic visits allowed an educational session to be incorporated into the course of the visit. Sharing of educational tools among the teams participating in the PHARE-M was boosted. A tool to identify and react to pulmonary exacerbations (REACT) was developed by the national TPE working group after the teams identified the variability in the practices of diagnosing and treating pulmonary exacerbations. Despite fears of therapeutic education competing for space in the teams' tight schedules, the PHARE-M strengthened the practice of PTE and the use of educational tools.

Prospects for the roll-out of PHARE-M and a CQI process in CF care in France

As of early 2017, the PHARE-M has been implemented at 23 centers (out of 45) and LLC6 is ongoing with adult teams. The teams' satisfaction is still increasing, with a median score of 9.1 for LLC5, which was a pediatric program. The outcomes of the centers will be made transparent among the professionals and the patient organization board only in the next few months. Public transparency will take more time.

The research program is aimed at assessing the impact of the PHARE-M on patient outcomes after three years, though it may be difficult to establish a causal link to the PHARE-M, given the evolving context in which centers operates and CF treatments are provided, and the bias inherent to recruiting centers that volunteer to participate. The realistic assessment will conduct an in depth examination of "how and why" a stronger impact of the PHARE-M may have been observed at certain centers engaged in PHARE-M [96]. Presenting the results of the research program in 2017 and publishing on PHARE-M initiative will definitely increase the visibility of PHARE-M and raise awareness in France on this quality improvement approach.

Six years after the PHARE-M was launched in the CF network in France, half the centers have been trained, and the various stakeholders – professionals, patient organization representatives and hospital quality department members in some hospitals – perceive the strength of this LLC QI approach and wish to participate in it and contribute to rolling it out further. Interest in this approach is growing outside of CF care, for example among hospital quality professionals willing to test patient shadowing in other chronic care departments.

Beyond these short-term contributions, the need for overall reflection to adapt the method to another model of care (translated in a disease specific Action Guide) requires a dedicated task force at an appropriate level of the health system. Experience with the QIP in CF may inspire its application to the care of other chronic diseases, and this article may contribute to its dissemination.

Aknowledgements: We would like to thank Vaincre la Mucoviscidose and the Foundation ildys for their financial support to the PHARE-M program.

Box 5 Next steps to accelerate the pace of the roll-out of the PHARE-M in France

1 Develop the French CF Registry

- Reduce the time taken to produce annual Registry reports;
- Achieve public transparency of the results by center;
- Advance towards an encounter-based national CF database which produces annual Registry reports as well as ongoing (quarterly) results for the monitoring of the QIPs at the centers

2 Strengthen the motivation of the teams to enroll in PHARE-M program

- Report the PHARE-M experience, results and satisfaction during professional conferences and patient organization assemblies;
- Get the CF community leadership, professionals and the patient organization more involved in continuous quality improvement;
- Continue to obtain funding from the patient organization for the extra-time needed for the PHARE-M referent at each center during the training year;
- Validate continuing professional development credits through the PHARE-M;
- Maintain a focus on continuous quality improvement with financial support for post-PHARE-M cycles until other funding is available (see below);
- Develop a convergence between the roll-out of the PHARE-M and other actions to increase the availability of professional resources, access to CF care guidelines translated in French, and tutoring by discipline within the network;

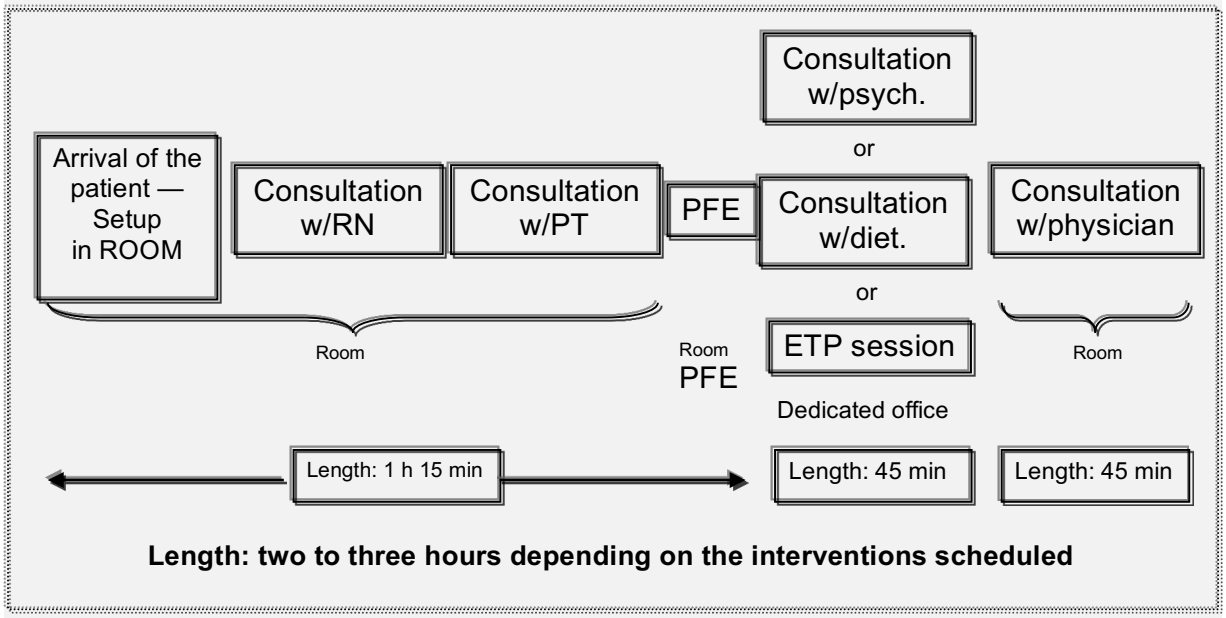
3 Consolidate and develop expertise and resources for the PHARE-M

- Organize a community of PHARE-M referents from the centers for advanced training on measurement, effective meeting skills, quality tools (fishbone diagrams, PDSAs, patient shadowing);
- Develop a culture of publishing QI initiatives according to SQUIRE standards
- Improve and adapt the PHARE-M website to show the various aspects of the program (registration to the CPD program, international research, international community ties, publications, etc…)

4 Build alliances at the hospital and national health system levels

- Continue contributing to the hospital certification process, supporting the hospital quality department through improvement actions, Professional Practice Evaluations, or hospital quality indicators;
- Develop new CPD programs for post PHARE-M cycles focusing on providing reminders of the QI method and tools, benchmarking, measuring and writing for publications;
- Participate in conferences of health authorities or working groups aimed at care quality improvement and patient involvement in healthcare to promote this QI LLC method;

635 **Figure 1 -** Example of multidisciplinary consultation process at a pediatric CFC



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No.	Step	Who	What	Length (min)	Protocol
1	Installation of the patient	RN	- Setup in the dedicated room - Collection of new elements since the last visit - Verification of the results of examinations performed in the community or at the hospital - Needs for administrative documents (transport passes and certificates) - Reminder of the hygiene rules (wearing a mask) - Validation of the day's clinic visit circuit	5 - 10	Hygiene — CR
2	Consultation w/nurse	RN	- Taking of measurements (weight and height) - Recording of the assessment in the patient's electronic record - Taking stock of the treatments prescribed and taken - Care (implantable device, blood draw, etc.) - Events in the life of the patient to be prepared - Responses to the patient's/parent's questions	20 - 30	Measurement protocol (height and weight) according to the patient's age
3	Respiratory assessment	PT	- Implementation of the hygiene protocol - Taking stock of the physiotherapy practiced in the community and review of instrumental aids - Taking stock of physical and sports activities - Physiotherapy session with sputum collection for sputum culture - Assessment of bronchial congestion - Recording of the assessment in the patient's electronic record	40	
4	PFT (pulmonary function test)		- Measurement of respiratory function - Recording in the patient's electronic record	10	Recommendations of the American Thoracic Society
5	Other scheduled intervention		- Psychological assessment (psychologist), social assessment (social worker), or nutritional assessment (dietician) - Or individual therapeutic education session - Recording of the assessment in the patient's electronic record	30 - 40	
6	Medical consultation	Physician	- Additional examination - Clinical examination - Review of all treatment - Response to the patient's/parent's questions - Referral to the referent professional	35 - 45	End of the course of consultation to benefit from assessments performed by the other

			- Planning of the next visit and need for additional examinations to be performed at the hospital or in the community - Preparation of prescriptions - Recording in the patient's electronic record - Signing of medical certificates		professionals recorded in the patient's electronic record
7	Departure of the patient	Admin. Sec. or RN	- Scheduling of the next appointment - Review of organization for departure (transport, nutritional need, and support) - Verification that the patient has all useful documents - Instructions for events by the next visit - Once the patient leaves the room, disinfection before accommodating the next patient.	30	Disinfection protocol

Table I - Description of the steps of the multidisciplinary consultation process

IV- OBJECTIFS DE LA THESE : EVALUER L'APPORT D'UN PARTENARIAT PATIENT DANS LA DEMARCHE D'AQ DES SOINS EN MUCOVISCIDOSE PHARE-M

IV-1. Le programme de recherche sur l'intervention PHARE-M

Les évaluations externes réalisées lors de l'introduction de l'intervention PHARE-M, auprès des équipes de la phase pilote, ont permis d'observer les prémisses de changements dans les pratiques de soin, la satisfaction des équipes des centres engagés, des résistances à la participation des patients et parents, tout en soulignant la nécessité d'un recul de trois années au moins pour en mesurer l'impact éventuel sur les indicateurs de santé des patients suivis dans ces centres.

Un projet de recherche a été élaboré dans le but d'évaluer l'impact, après trois années, du programme qualité PHARE-M dans les CRCM engagés dans la phase pilote (2011- 2013). Il a été soumis à l'AAP du ministère dédié à la Recherche sur la Performance des Soins (PRePS) et a été sélectionné par le ministère de la santé en décembre 2012.

Le projet de recherche, intitulé PHARE-M Performance, a pour objectif principal d'évaluer, en 2015, l'impact de la démarche qualité PHARE-M sur ***l'évolution des indicateurs de santé des patients*** pour le groupe de patients suivis dans les 14 centres formés à PHARE-M, et de la comparer à l'évolution des indicateurs de santé d'un groupe de patients non exposés à la démarche, car suivis dans des centres non formés au programme PHARE-M jusqu'en 2015. Les données de santé annuelles des patients sont issues du Registre Français de la Mucoviscidose pour les années 2011 à 2015.

L'objectif secondaire du projet de recherche est d'analyser les contextes des CRCM et les mécanismes mis en jeu, associés à une ***variabilité d'impact du programme dans les 14 centres formés***. L'impact est mesuré d'une part sur l'évolution des indicateurs de santé des patients (résultats de l'approche quantitative) et d'autre part, sur les caractéristiques de la prise en charge au regard des critères du ***Chronic Care Model*** et des critères d'une prise en charge centrée sur le patient. Cet objectif est réalisé grâce à une évaluation réaliste de l'intervention complexe PHARE-M (45,97).

Du fait de son objet et de son design mixte intégrant une évaluation réaliste d'intervention complexe, ce projet de recherche constitue ***une contribution française à la recherche internationale sur une démarche qualité collaborative dans notre système de soin, pour une maladie rare.*** Il est à noter que le programme américain dont est issu le PHARE-M n'a pas fait l'objet d'un projet de recherche similaire, l'évolution positive des indicateurs de santé des patients observée au cours des dernières décennies dans le registre des patients aux Etats-Unis étant « attribuée » à un faisceau de causes agissant toutes sur l'amélioration des soins – telles que la création du réseau des centres spécialisés, du registre, la diffusion des recommandations de soins, et l'implication des patients et parents dans un partenariat pour leurs soins (98, 99, 100, 101,190, 102). Le programme qualité est considéré comme un ***accélérateur de l'amélioration des soins*** (non publié), et les

facteurs de succès de la démarche identifiés par la méthode du benchmarking dans les centres ayant les meilleurs indicateurs de résultats.

La coordination du projet de recherche PHARE-M Performance a été confiée au parent coordinateur du programme PHARE-M, dans le but de capitaliser sur sa connaissance de la démarche qualité pour ***modéliser l'intervention et construire les instruments de l'évaluation réaliste***. Le cadre théorique de la recherche et son design mixte sont présentés dans la partie suivante du document (§V).

IV-2. Les objectifs spécifiques de la thèse

La thèse a pour objectif principal d'***évaluer l'apport du partenariat patient (et parent) dans le succès de la démarche d'AQ des soins en mucoviscidose PHARE-M***. Pour répondre à cet objectif, la réflexion conduite a pris appui sur l'étude réaliste, à travers la modélisation de l'intervention, la conception des instruments d'évaluation et la définition du mode de recueil des données du projet de recherche. Cette articulation de la thèse avec l'étude réaliste a permis d'analyser le partenariat patient en lien avec les autres éléments du contexte et mécanismes par lesquels l'intervention a opéré dans les CRCM.

L'évaluation de l'apport du partenariat patient vise à répondre aux interrogations soulevées par les acteurs et à élaborer des connaissances utilisables pour une transposition éventuelle à d'autres contextes de pathologies chroniques et/ou rares. Ces interrogations formulées au lancement de la démarche en France, portaient sur l'intérêt d'associer les patients/parents aux différentes étapes du programme, la faisabilité de leur participation sur la durée, son utilité pour le travail des équipes, son acceptabilité par les professionnels, et l'impact éventuellement délétère de cette participation sur les patients et parents eux-mêmes. Dans quelques CRCM pédiatriques où existait un collectif de parents, des craintes sur l'articulation des rôles entre le collectif et le parent membre de l'équipe de pilotage ont été exprimées. Le mode de recrutement par les équipes soignantes et les critères de choix du parent ou patient ont également été questionnés.

Si l'apport du partenariat patient dans la démarche qualité devait être évalué, il importait que cette évaluation :

- s'inscrive dans la modélisation de l'intervention complexe, et s'intègre à l'ensemble des éléments de contexte et des mécanismes explorés dans le cadre de la recherche,
- soit réalisée par l'ensemble des participants, professionnels et patients/parents ayant participé à la démarche.

La participation des patients/parents à la recherche a été organisée dans le même cadre que celle des soignants des équipes de pilotage : il a été demandé à tous les membres des équipes de répondre à l'enquête sur l'ensemble des composantes et des effets modélisés de l'intervention – la qualité des soins, le fonctionnement des équipes et la participation des patients et des parents. L'analyse des réponses a ainsi permis de dégager les consensus et les différences de points de vue entre les professionnels et les patients/parents.

A notre connaissance, c'est la première étude qui interroge le point de vue des patients et parents engagés sur la démarche qualité collaborative à laquelle ils ont participé, recueillant leur opinion sur l'ensemble des domaines de l'AQ au même titre que celui des professionnels.

Quatre objectifs spécifiques ont été ciblés dans la thèse :

OS1 : Evaluer les conditions mises en place pour permettre la participation des patients/parents dans le programme PHARE-M et dans la démarche continue d'amélioration de la qualité *(empowerment)*

Cette évaluation avait pour but de valider (ou ajuster) le ***dispositif d'intégration des parents et patients dans la démarche PHARE-M*** pour les sessions futures du programme, et de ***tester certaines propositions nouvelles***, plus largement débattues dans les domaines de la participation de patients experts formateurs ou éducateurs, pour faciliter leur participation ou en améliorer la contribution effective.

OS2 : Evaluer l'effet de la démarche qualité auprès des professionnels et des patients/parents, à travers la maîtrise des outils et des méthodes de la qualité, le fonctionnement de l'équipe de pilotage et in fine la perception d'utilité d'une telle démarche *(compétences acquises)*

Cette évaluation visait notamment à valider ou infirmer l'intérêt de la ***formation des parents et patients*** à la démarche qualité, au cours des journées nationales de formation organisées pendant l'année du PHARE-M.

En effet, des arguments étaient présentés pour privilégier leur participation à des réunions de travail locales de l'équipe de pilotage, à l'instar de ce qui se fait dans le programme américain, sans que leur formation aux outils et aux méthodes de la qualité apparaisse nécessaire pour leur contribution à la démarche.

Parmi ces arguments, la nécessité d'une formation de l'usager membre de l'équipe de pilotage à une formation « *professionnelle à la démarche qualité* » apparaissait discutable par comparaison avec l'engagement, dans les collectifs des CRCM, d'usagers n'ayant pas bénéficié d'une acculturation préalable à la « *défense des droits des usagers* ».

La formation qualité dispensée aux patients et parents engagés a déstabilisé certains bénévoles des collectifs ou des instances régionales associatives, leur donnant le sentiment de constituer une « *élite* » d'usagers. Elle a aussi rencontré le scepticisme de certains professionnels, doutant de la capacité des patients/parents à prendre suffisamment de recul pour tirer profit de ces apports méthodologiques.

Enfin, des arguments économiques étaient présentés par l'association qui finançait leurs frais de déplacement. L'évaluation de l'efficacité de la démarche a consisté en une auto-évaluation des compétences acquises par les professionnels et les patients/parents dans les différents domaines de la démarche qualité et le recueil de leur point de vue sur le fonctionnement de l'équipe de pilotage - l'organisation du travail, la communication entre ses membres, le mode de prise de décision et les collaborations internes et externes, qui est un facteur de modulation de l'efficacité de la démarche qualité dans le CRCM.

OS3 : Appréhender l'évolution de la représentation de la place de l'usager chez les professionnels et les patients/parents suite à l'expérience de participation des patients/parents au programme qualité PHARE-M *(place de l'usager)*

L'objectif était d'analyser l'évolution de la perception, par les professionnels et les patients/parents, de la participation de ces derniers au programme PHARE-M entre le début de la démarche (évaluations préliminaires) et le moment de l'enquête réalisée après trois années de démarche qualité continue. L'hypothèse était que les tensions observées au début du programme pouvaient se dissoudre dans la pratique durable du travail collaboratif en équipe pluridisciplinaire.

OS4 : Appréhender le niveau de qualité des soins et de culture de l'organisation après trois années de démarche qualité continue, perçu par les professionnels et des patients/parents *(qualité des soins)*

L'instrument utilisé pour l'évaluation de la qualité des soins, à savoir les critères du Chronic care model et ceux d'une prise en charge centrée sur le patient, caractérisait les composantes d'une prise en charge « conforme » à celle attendue dans un CRCM, a fortiori après trois années de démarche qualité.

La liste des items reflétait cette prise en charge attendue, et devait permettre d'atteindre un score de réponses positives au-delà de 80/100 et très voisin de 100/100. Aucun item ne relevait d'une prise en charge « exceptionnelle ». Seule la possibilité de réponses neutres (« ne sait pas ») à certains items pouvait laisser entrevoir d'éventuels scores inférieurs à la cible.

La première hypothèse était donc que tous les CRCM impliqués dans la démarche aient pu mettre en place une prise en charge conforme à ces caractéristiques en 2015. La seconde hypothèse était que la vision des soins dispensés par le CRCM soit partagée par les professionnels et les patients/parents après trois années de travail en commun.

V- CADRE THEORIQUE DE LA RECHERCHE : UN DESIGN MIXTE QUANTITATIF ET QUALITATIF POUR EXPLORER L'IMPACT DE LA DEMARCHE QUALITE

V-1. La modélisation du programme PHARE-M

Le programme PHARE-M a été modélisé sous la forme d'une intervention complexe.

Cette intervention consiste à installer, former et accompagner une équipe de pilotage composée de 4 à 5 membres de l'équipe pluridisciplinaire du CRCM et d'un patient ou parent de la file active. Les membres de l'équipe de pilotage sont formés aux outils de la démarche qualité et à la conduite du changement dans le CRCM.

L'impact de l'intervention est mesuré sur ***l'évolution des résultats de santé*** des patients et sur ***la qualité des soins après 3 ans***.

L'effet direct de la démarche est évalué en termes d'appropriation de la méthode et des outils de la qualité, le transfert de connaissances et les modalités de mise en œuvre de changements dans l'organisation, ainsi que la perception de l'utilité de la démarche par les équipes. Cet impact direct est intitulé « ***effet de la démarche*** ».

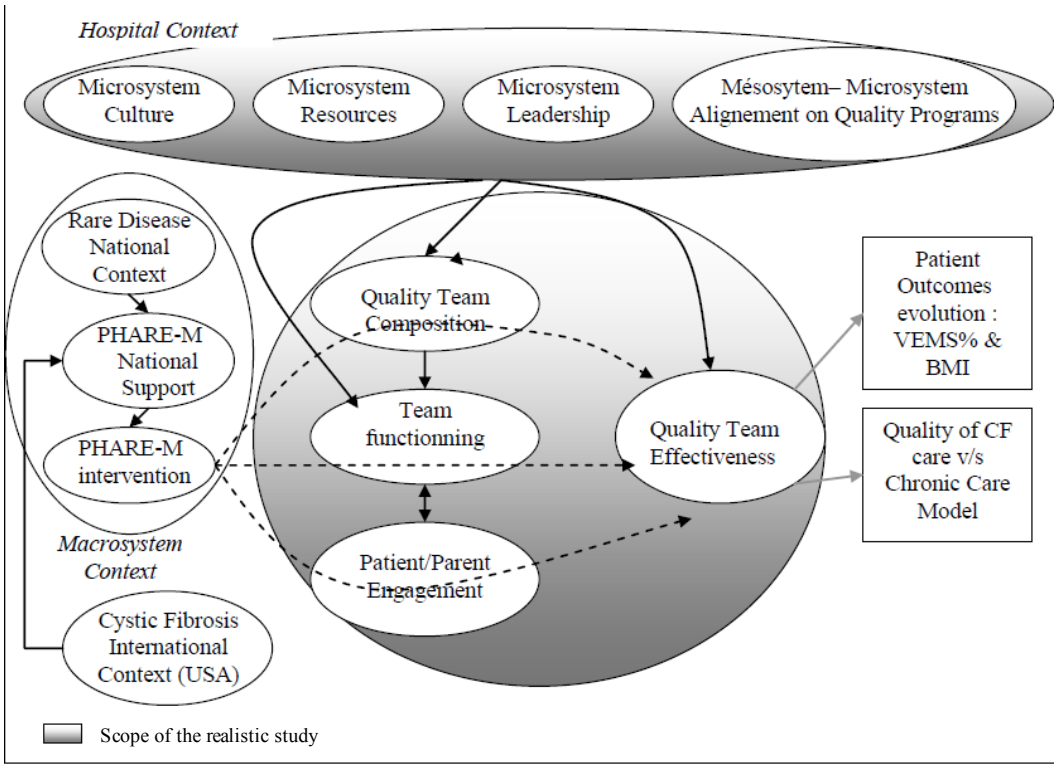
L'effet de la démarche dans le CRCM est potentiellement modulé par des « mécanismes », tels que le ***fonctionnement de l'équipe de pilotage*** du PHARE-M dans le CRCM (rigueur du travail, processus de prise de décision, clarté des rôles et responsabilités…) et ***l'engagement du patient ou parent***.

Des ***éléments de contextes*** extérieurs à l'intervention peuvent conditionner à la fois l'adhésion de l'équipe au programme (« effet de la démarche ») et son impact (« résultats » et « qualité des soins »).

Les éléments de contexte retenus dans la modélisation incluent la ***composition de l'équipe pluridisciplinaire*** (disponibilité des ressources professionnelles) qui a influencé la composition de l'équipe de pilotage, le ***leadership***, la ***culture de l'organisation*** centrée sur le patient et son ouverture à l'innovation, ainsi que le ***support apporté par le département qualité*** de l'hôpital.

La figure 3 ci-après représente la modélisation de l'intervention, du contexte et des mécanismes explorés.

Figure 3 : Modélisation de l'intervention, du contexte et des mécanismes.



V-2. Le design mixte de la recherche

Le design repose sur :

1) une étude quantitative d'une cohorte fermée de patients sur la base des données de santé collectées dans le Registre de 2011 à 2015 : les deux groupes de patients, exposés à l'intervention et contrôle, ont été appariés par tranche d'âge, type et taille de CRCM ;

2) une étude qualitative basée sur les hypothèses décrites dans le modèle de l'intervention : un questionnaire a été élaboré par un groupe d'experts coordonné par la doctorante, testé auprès des équipes des CRCM du centre de référence, ajusté avant déploiement auprès de l'ensemble des personnels des 14 CRCM formés y compris les patients et parents engagés ; ce questionnaire couvre :

- ***L'impact en termes de qualité des soins*** décrite selon les caractéristiques du CCM (103) décliné à la mucoviscidose en une liste de 47 items élaborée dans le cadre de la recherche ;
- ***L'effet en termes d'appropriation de la démarche*** par l'équipe de pilotage décrite selon les items validés par les travaux de Lemieux-Charles (104) et Shortell (105) ;
- ***les mécanismes internes*** modulant l'appropriation de la démarche par l'équipe de pilotage (EP) décrits par Lémieux-Charles : 1) l'organisation du travail de l'EP, 2) le processus de décision au sein de l'EP 3) les objectifs partagés d'amélioration, et 4) la communication et le support externe et 5) l'engagement des patients/parents caractérisé à partir de la publication de Carman (106) en une liste de 31 items, élaborée dans le cadre de la recherche ;

- ***les éléments de contexte*** : 1) la composition des équipes pluridisciplinaires en 2011, qui a pu influer sur la constitution des équipes de pilotage du PHARE-M ; 2) la culture de l'organisation ; 3) le leadership (décrits dans les travaux de Shortell) ; 4) l'alignement entre la politique qualité de l'établissement et le PHARE-M à l'aide 8 questions ouvertes adressées au responsable du département qualité (décrites dans l'étude européenne QUASER (107)).

En complément du questionnaire, des ***Focus groups*** ont été conduits sur les 14 sites avec les membres des équipes de pilotage pour explorer la perception des freins et des succès de la démarche qualité par les équipes, autour de 4 questions ouvertes : 1) Quels sont les changements dans l'organisation du CRCM qui résultent du programme qualité PHARE-M ? 2) Quelles ont été les difficultés rencontrées au cours du PHARE-M ? 3) Quels sont les succès notables que vous attribuez à la démarche qualité ? 4) Quelles sont les leçons de cette expérience que vous souhaiteriez transmettre aux prochains CRCM ? Les résultats de ces focus group ont été mis en perspective des résultats de l'enquête conduite en 2012 par l'évaluateur externe dans le cadre de l'évaluation du processus de transposition de la démarche en France.

Les autorisations règlementaires ont été délivrées par le Comité d'Ethique du CHU de Brest et par la CNIL (DR2015040).

L'article IV (Volume 2) décrit le protocole de recherche. L'abstract (*traduction française)* est présenté ci-après. L'article complet est intégré à la suite.

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Introduction

Le programme d'amélioration de la qualité des soins PHARE-M, inspiré du programme américain, a été introduit dans 14 CRCM de la filière mucoviscidose entre 2011 et 2013. Les évaluations de la phase pilote ont attesté l'adhésion progressive des équipes et des améliorations dans la prise en charge. Le projet de recherche PHARE-M Performance vise à évaluer en 2015 l'impact du programme PHARE-M sur les indicateurs de santé des patients dans les centres formés versus les centres non formés. Il vise aussi à identifier des éléments contextuels et les mécanismes mis en jeu qui pourraient expliquer la variabilité dans la performance du PHARE-M parmi les centres formés.

Méthode

Une méthodologie mixte combinant :

- une étude expérimentale quantitative : comparer, à l'aide d'un modèle pour données répétées (de 2011 à 2015), l'évolution des valeurs moyennes du volume expiratoire maximal en une seconde (VEMS) et de l'indice de masse corporelle (IMC) entre deux groupes de patients inclus dans une cohorte fermée, l'un étant suivi dans l'un des 14 CRCM ayant bénéficié du programme PHARE-M et l'autre dans des CRCM non formés jusqu'en 2015, et

- une étude réaliste : 1) modéliser l'intervention complexe ; 2) caractériser l'impact du programme sur la qualité des soins en 2015 selon les critères du Chronic Care Model et, 3) explorer les mécanismes par lesquels l'intervention PHARE-M a contribué à l'efficacité de la démarche qualité dans les différents contextes de CRCM ; un questionnaire d'enquête a été développé pour administration aux professionnels et aux patients et parents ; des focus group ont été menés en complément.

Discussion

Malgré la constitution d'une cohorte contrôlée et appariée entre les deux groupes, il peut être difficile d'établir une relation de causalité entre l'évolution des indicateurs de santé des patients entre les deux groupes de patients et l'intervention PHARE-M, comme c'est souvent le cas dans les interventions complexes introduites dans des environnements adaptatifs. L'analyse des facteurs associés aux variations de l'impact du PHARE-M entre les différents CRCM engagés dans la démarche a nécessité l'adoption et l'adaptation à la mucoviscidose d'instruments validés dans d'autres contextes ; ceux-ci pourraient être utilisables à l'avenir pour évaluer la performance d'autres types d'interventions complexes dans les CRCM en France.

Mots-clés : *mucoviscidose ; programme qualité des soins ; étude quantitative ; registre patient ; étude réaliste ; intervention complexe.*

Quality of care in cystic fibrosis: assessment protocol of the French QIP PHARE-M*

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Abstract

Background

The PHARE-M care quality improvement program, modeled on the US Cystic Fibrosis Quality Improvement Program, was introduced at 14 cystic fibrosis centers (CFCs) in the French Cystic Fibrosis Network between 2011 and 2013. The pilot phase assessments attested the progressive adherence of the teams and improvements in care management. The PHARE-M Performance research project aims at assessing in 2015 the impact of the PHARE-M program on patient health indicators at trained versus untrained centers. It also sought to identify contextual factors that could account for variability in the performance of the PHARE-M among the trained centers.

Method

A mixed methodology combining:

- a quantitative experimental study: a comparison, using a mixed model for repeated data (from 2011 to 2015), of the average changes over time in forced expiratory volume in one second (FEV1) and body mass index (BMI) between two groups of patients included in a closed cohort (non-transplant patients, continuous follow-up at one participating CFC, and a CF-causing mutation), one having benefitted from the PHARE-M program and the other not having done so, and

- a realistic study: a characterization of the impact on care management and an identification of mechanisms through which the PHARE-M intervention improved the team's effectiveness in different CFC contexts; this required modeling the intervention, context, and impact on care management with respect to the criteria of the chronic care model (CCM); this was done using a self-administered questionnaire given to professionals and patients/parents supplemented with focus groups.

Discussion

Although the study population was controlled, it may be difficult to establish a causal relationship between the differences in the changes over time in patient health indicators in the two groups of patients and the PHARE-M intervention as it is often the case in complex interventions rolled out in adaptive environments. The analysis of factors associated with variations in the impact of the PHARE-M at the different trained CFCs required the adoption of instruments validated in other contexts; these could be useful for assessing the performance of other interventions in healthcare practices at CFCs in France.

Keywords: cystic fibrosis; quality improvement program; quantitative study; patient registry; qualitative study;

Background

Cystic fibrosis is the most common rare disease affecting the Caucasian population; it afflicts around 6,500 individuals in France, 29,000 in the United States, and 11,000 in the United Kingdom. It is an autosomal recessive genetic disease caused by mutations in the cystic fibrosis transmembrane conductance regulator (*CFTR*) gene. Among all identified *CFTR* gene mutations, a list of mutations responsible for cystic fibrosis symptoms has been established and is regularly reviewed by the CFTR2 expert group [108]. Cystic fibrosis mainly affects the respiratory and digestive systems. The thick mucus in the bronchi brings about chronic inflammation and repeated infections, leading to chronic respiratory failure, the major cause of death. The majority of patients have pancreatic insufficiency and show poor nutrient absorption, resulting in an at-risk nutritional status associated with a poorer respiratory state [109]. Since the 1960s, the US Cystic Fibrosis Foundation (CFF) has identified multidisciplinary patient management at specialized centers as an essential factor in care improvement; this has led it to establish criteria for the accreditation of cystic fibrosis centers [110]. In the late 1990s, an increase in the number of adults suffering from cystic fibrosis led the CFF to clarify certain criteria for adult centers by stipulating care management by specialized physicians and a specialized team and a formalized process of transition from a pediatric center to an adult program. The accreditation process not only validates centers but also *"fosters continuous improvement efforts within care centers,"* as *"the expectation that each care center have a QI program in place was added to the accreditation and oversight process in 2004."* In the 2000s, following the publication by the US Institute of Medicine, of the report on the Quality Chasm [[111]], the CFF launched a benchmarking study across the US CFCs, which showed a difference of several years in the median survival age between the ten centers having the best patient outcomes and the other centers (unpublished study). This led the CFF to develop and implement a Quality Improvement Program (QIP) in the form of Learning and Leadership collaboratives [[112], 113, [114]] with the academic support of The Dartmouth Institute Microsystem Academy (TDIMA). A supplement in BMJ Quality and Safety has been published in May 2014 to present the success of this QI initiative [[115]].

In 2002, following the generalization of newborn screening in France, the French Ministry of Health designated 49 cystic fibrosis centers (CFCs) [[116]] and in 2006, the French National Authority for Health (HAS) published the National Diagnosis and Treatment Protocol (PNDS) in Cystic Fibrosis to establish a framework for multidisciplinary care at CFCs. The French public health insurance guarantees that every CF patient is reimbursed

100% for care and authorized drugs related to cystic fibrosis. In 2006, within the framework of the 1st National Plan for Rare Diseases, two centers of expertise for cystic fibrosis were labelled (CF-CERDs), in order to implement six priorities across the CF Network: care expertise, information systems and epidemiology, quality of care, clinical research, network organization and coordination. The Nantes/Roscoff CF-CERD, consisting of the CFCs at the two hospitals in Nantes and Roscoff as well as the transplant center in Nantes and the rehabilitation center in Roscoff, developed its action plan contributing to 5 out of the 6 priorities, covering themes such as therapeutic patient education (care expertise), quality improvement in care processes, information and communication systems, and clinical research on transplantation and in human and social science. The agreement signed by the heads of all CFCs in 2007 included a commitment to *"participate in a quality assessment and improvement program to be offered by the CF-CERDs in collaboration with the French Cystic Fibrosis Society (SFM) and the patient organizations in the next five years"*.

In 2011, the French national team at the Nantes/Roscoff CF-CERD transposed the PHARE-M quality improvement program from the US CFF QIP model. It was launched in September 2011 with a pilot phase (2011-2012) involving seven volunteer CFCs, which underwent two external assessments, leading to certain adjustments to the initial program. This adjusted version was deployed during a regional expansion phase (2012-2013), including seven more CFCs before its national deployment [[117]]. The main adjustments consisted in more practical exercises during face-to-face meetings (less theoretical presentations), more on-site coaching to help the quality teams analyze their processes of care, and the designation of a PHARE-M referent in each local team to keep focused on the QI work. These two years are called the "experimental phase", which involved 14 CFCs.

The two evaluations at the end of the one-year pilot phase showed the progressive adherence of the teams and improvements in care management, but a limited impact on patient health outcomes. They also highlighted that the adherence to the program mainly depended on the motivation of the multidisciplinary team (MDT), especially its lead physician. The lack of resources at some CFCs was raised to account for variations in the teams' engagement as the level of available staff seemed to influence the extent to which the team was effectively enlisted. The participation of a patient or parent in each local quality team varied depending on the cultural context of the centers, some being used to share information with patients/parents, having a patient group in the CF center for years, others being involved in patient therapeutic education while others were acting in a more partenalistic model of care. The support received from the hospital quality department in two hospitals was emphasized as

a factor that facilitated the adoption of quality tools by the teams. The recommendation of the assessor was to evaluate the impact of the program on patient outcomes by 2015.

Given the innovative nature of the QIP PHARE-M in France, the cultural differences and various organizational contexts at the CFCs, an assessment of the impact of PHARE-M at the CFCs engaged in the experimental phase was expected after three years to continue the enrollment in the program. Will it show favorable changes in the patient outcomes in the group of CFCs engaged in the PHARE-M compared to the other CFCs? What impact on care management can be observed in 2015? Was the period sufficient to show improvements in the two areas? In which contexts is the impact of PHARE-M observed to be the strongest? The PHARE-M Performance research project, submitted at a call for projects of the French Ministry of Health and selected for funding in December 2012, aims at providing answers to these questions.

Method

1- A mixed methodology

The rationale of the PHARE-M Performance project is to show evidence of the performance of the PHARE-M program on patient outcomes and care management.

The study is based on a mixed methodology inspired on the one hand by epidemiology, using data from the French Cystic Fibrosis Registry, and on the other hand by the British guidelines on "Process evaluation of complex interventions" [118] :

1) *a quantitative study* to compare the changes over the 4 years in the patient health indicators of a closed cohort, using data from the French Cystic Fibrosis Patient Registry, between CFCs having benefitted from the intervention during the experimental phase and CFCs not having benefitted from the intervention up to 2015; and

2) *a qualitative study* to analyze the contextual elements and mechanisms brought into play by the PHARE-M intervention that could account for a difference in impact among trained CFCs either on patient health indicators or on care management assessed according to the criteria of the chronic care model [119].

2- Quantitative Study

2-1- Design

- observational,
- national and multi-center, and

- before/after and here/elsewhere: a comparison of patient health indicators before and after the "PHARE-M training" program at "PHARE-M Group" centers versus "Control Group" centers.

2-1-1- Primary and secondary endpoints

- FEV1%
- BMI as an absolute value and as a Z-score (standardized normal distribution of the BMI for children under two years of age)

For this research in particular, the value selected for these indicators is the only value appearing in the French CF Registry for a given patient and a given year. It will be analyzed by category of patients defined by age, sex, age at diagnosis, and possibly severity of disease expression, treatment, and certain social characteristics (data appearing in the Registry).

2-1-2- Study population

A closed cohort was formed to identify the study population including the patients followed up at CFCs who met the following inclusion criteria according to the 2012 Registry data:

- patients seen at a CFC in 2012
- patients having two of the CF-causing mutations of the CFTR2 list published on Feb 2012
- patients not having received a transplant in 2012

A patient left the cohort if he or she no longer met the inclusion criteria after the annual data were updated in the Registry (2013, 2014, and 2015), i.e.: if he or she was a carrier of a mutation excluded from the CFTR2 list updated on 13/08/2015 [108]; if he or she was followed up at a CFC engaged in the PHARE-M in 2014 or 2015; if he or she changed CFC in the course of the study and in doing so, changed CFC group; if he or she received a transplant between 2013 and 2015 (data up to the transplantation were taken into account), or if the patient died between 2013 and 2015 (data up to the death were taken into account).

The cohort was divided into two groups: the "PHARE-M Group" and the "Control Group":

- The "PHARE-M Group" consisted of the patients followed up at one of the 14 CFCs trained in the PHARE-M in the experimental phase (1,309 patients).
- The "Control Group" consisted of the patients followed up at the CFCs not having benefitted from the intervention in the same period of time (2,490 patients).

2-2- Pairing of the two "PHARE-M" and "Control" Groups

A preliminary analysis of the cohort formed from the 2012 Registry data showed significant differences between the two groups of patients, before the PHARE-M intervention, in terms of: 1) distribution by age, 2) distribution by age at diagnosis, and 3) distribution by FEV1% value (see Table I).

Consequently, a 1:1 pairing of the patients from the Control Group was decided in an attempt to eliminate certain confounding factors that could be attributed to the type and size of the CFC to which the patient was assigned: each "PHARE-M patient" was associated with a "control patient" followed up at a center of the same type (pediatric, adult, or mixed) caring for a total number of patients belonging to the same interval ([1;50], [51;100], [101;150], [151;200], or [> = 200]). Reunion island CFCs were excluded from the Control Group to reduce heterogeneity in CF care. All "eligible" control patients for each patient in the PHARE-M Group were selected, and one control patient was randomly drawn from that group of eligible control patients (without replacement). The patients in the PHARE-M Group were paired in a random order.

At the end of the process, 1,104 patients remained in each of the two paired groups. The Control Group included 20 CFCs. No paired control patients were found for 205 "PHARE-M patients". As data are collected in the French Cystic Fibrosis Registry for all patients, exposure variables are identical in both groups. Completeness is similar: for FEV1, 20.2% and 24.5% of missing data corresponding to the children below 6 y.o., for whom this measure is not taken, and 0.6% and 3.5% for ZBMI, in the PHARE-M group and the Control group respectively. The two groups had a similar distribution by age (see Fig. 1). However, there remained a significant difference in average age at diagnosis (PHARE-M paired group: 1.9 years; control paired group: 2.5 years; p value: 0.0123); this could be due to the fact that newborn screening was implemented in the 1990s in Brittany, and that seven (out of the 14) CFCs in the PHARE-M Group are located in this region. Furthermore, a significant difference in FEV1% of +3.89% (p value = 0.0012) remained in favor of the PHARE-M patient group before the intervention (see Table II).

2-3- Analysis of the primary endpoint between the two groups

Changes over 5 years in patient health indicators are measured for 2011 (baseline), 2012, 2013, 2014, and 2015; each patient served as his or her own control. A difference in the rate of decline is expected between the two population groups, PHARE-M and control (see Fig. 2). Changes over time in FEV1% will be modeled and compared in the two groups using a mixed model for repeated data with adjustments for potential confounding variables. Measurements for a subject i at time j is given by the following model, where ε_{ij} are the normally distributed residual components with mean zero and covariance structure Σ :

$Y_{ij} = \beta_0^P + \beta_1^P t_{ij} + \varepsilon_{ij}$ for the PHARE-M group

$Y_{ij} = \beta_0^C + \beta_1^C t_{ij} + \varepsilon_{ij}$ for the CONTROL group

$cov(\varepsilon_{ij}, \varepsilon_{ik}) = \sigma_{jk}$

The covariance structure Σ is given by the σ_{jk}. It allows taking into account correlation between measurements on a same subject. Correlation is assumed to be null between subjects. The choice of a covariance structure will be data driven, but we can expect that the correlation between two measurements will only depend on the time lag between them. The most realistic covariance structure should be the so-called Toeplitz covariance matrix. A special case of the Toeplitz model is the first-order autoregressive model.

The question here is to investigate whether the two slopes are parallel or not, that is to test whether $\beta_1^P = \beta_1^C$ (H_0) versus $\beta_1^P \neq \beta_1^C$ (H_1).

Using this model, the slopes (i.e. decline in FEV1) in the two groups will be calculated and compared. Changes over time in BMI will likewise be analyzed by comparing the changes in the two groups from 2011 to 2015, taking into account the Z-score for children under two years of age. The average trends will be calculated and analyzed for different patient categories (such as age, sex, age at diagnosis, severity of disease expression, treatment, and certain social characteristics in the Registry). The changes over time in indicators will be presented for the "PHARE-M Group" population by CFC for crossing with the results of the qualitative study.

2-4- Audit of the quality of the data included in the primary endpoints' calculation

The patient data measured by the CFCs (height, weight, and FEV1 [per L]) for 2012 and 2013 underwent an on-site quality audit at the 14 CFCs in the PHARE-M Group. It was the first on-site audit ever performed to establish the quality of these indicators. The objective was not to comprehensively audit all data for the patients included in the study. Rather, the objective was to comprehensively identify the different causes of error due to failures in the processes of measuring and/or selecting the values transmitted to the Registry in order to identify avenues for improvement of the quality of the data in the Registry. The sample of patients whose data were audited thus had to reflect the distribution by age range of the patients at each CFC (20 records/CFC) in order to cover the different measurement procedures defined by international benchmarks [120, 121, 122] and the data selection rules defined by the French Patient Registry Steering Committee, and to offer every opportunity to reach saturation of the various causes of error [123]. They will be taken into account in the interpretation of the results of the quantitative study.

3- Qualitative Study

3-1- Design

The design refers to the modeling of the intervention [97] including the contextual elements and the mechanisms shown in Figure 3.

The PHARE-M intervention consisted of establishing, training and coaching a quality team (QT) at each CFC comprising a number of professionals from the multidisciplinary CF team and 1 parent or patient from the CFC's caseload. The members of the QT have been trained in quality methods and tools and coached in changing care processes. The PHARE-M intervention should have directly impacted the ability of the local QT to master QI methods and tools, lead changes in the care processes, and should have generated good appreciation of the utility of the QT efforts. This direct impact of PHARE-M is identified under the heading "QT effectiveness". QT effectiveness may not only be the result of the PHARE-M intervention but may have been modulated by internal mechanisms, such as the composition of the QT (number of members and disciplines enlisted), its functioning (rigor in the QI work, decision-making, clarity of the roles…) and the parent or patient engagement. Those mechanisms are represented as impacting QT effectiveness (Fig. 3). Beyond the ability to master the QI methods and tools, the PHARE-M intervention was expected to have an impact on the quality of CF care delivered at the CFC. The Chronic Care Model [119] was deemed appropriate to account for quality of CF care across the 6 dimensions: existing improvement goals, multidisciplinary care, self-management support, decision support (use of evidence-based guidelines), use of information system and electronic patient record, and organization of resources in the patient's community of life. Finally, an indirect impact of the PHARE-M intervention is expected on the trend in patient outcomes' evolution as measured in the quantitative part of this study. Moreover, some elements in the CFC contexts, which are external to the PHARE-M intervention and preexisted to its introduction, may have had a major impact both on the adherence of the team to the QI work and on its outputs. The contextual elements that have been brought in this study include the composition of the MDT, the leadership, the patient-centeredness of care, the innovative culture of the team, and the support from the hospital quality department.

The qualitative study will test these hypotheses using a questionnaire to be self-administered, in 2015, to all members of the MDT at the 14 CFCs and to the patients/parents participating in the quality teams.

Quality of care has been defined according to the criteria of the Chronic Care Model [119]; as this model has not been popularized in France nor in cystic fibrosis, we adapted it with 47 items aimed at characterizing CF care. *Table III presents a list of these items.*

QT effectiveness has been described in the studies by Lemieux-Charles [124] and Shortell [125]: it is characterized according to 27 items *(see Table IV).*

QT Internal factors that may have modulated the QT effectiveness: QT functioning [124] is characterized by 22 items classified in 4 categories 1) the organization at work, 2) the decision-making process, 3) the shared improvement goals, and 4) the ability to communicate and get external support. Studies by L. Lemieux-Charles defined these items to analyze the impact of adopting quality improvement practices on the internal functioning of a team. We use the same items to analyze if the team's functioning could modulate its effectiveness *(see Table V).*

The engagement of the patient/parent as characterized in Carman's framework [126] is assessed by a list of 31 items, prepared as part of this research *(see Table VI).*

The context elements include: the composition of the multidisciplinary team at the beginning of the PHARE-M intervention (2011) because it might have been a limiting factor in assigning staff to the QT; the culture of the microsystem to which the QT belongs [125] i.e. the organizational culture *(see Table VII)* and patient centeredness and leadership style *(see Table VIII);* the alignment of the PHARE-M QIP with the hospital quality policy as described within the framework of the European QUASER study [127] using eight open questions in an interview with a head of the hospital quality department *(see Table IX).*

Focus groups with the members of each QT were conducted by the Clinical Research Assistant, designed around four open-ended questions: 1) What changes in the organization of the CFC can be attributed to the PHARE-M? 2) What difficulties were faced at the CFC? 3) What successes were achieved? and 4) What lessons from this experience after 3 to 4 years? The results of these focus groups involving the 14 CFCs will be put in perspective with the results of the survey conducted by one assessor of the pilot phase who interviewed the 7 first CFCs on the following themes: 1) PHARE-M applicability, 2) participation of patients and parents, 3) functioning and coordination, 4) perceived benefits and costs, 5) effect on the team, 6) effect on care management, and 7) recommendations for PHARE-M national deployment.

3-2- Development of the instruments of the realistic study

The self-administered questionnaire was developed from the instruments (cited above) translated into French, and new items prepared as part of this research to characterize quality

of CF care and the degree of engagement of the patients or parents. The whole questionnaire is proposed to the members of the quality teams. A limited part of the questionnaire is proposed to the members of the MDT not on the quality team. The questionnaire has been prepared from January to June 2014 with clinicians from the Nantes/Roscoff CF-CERD and experts from the Health Education and Practice Laboratory (LEPS) at the Sorbonne Paris Cité University - Paris 13 Bobigny. It has then been tested between July and September 2014 in three teams from the Nantes/Roscoff CF-CERD (pediatric, adult, and mixed) with 29 respondents from all disciplines and the patients/parents participating in the QT. As a result of these tests, the questionnaire has been slightly adapted, essentially by rewording parts of the French translation and adding free text fields (*Questionnaire available upon request to the corresponding author*).

4- On-site investigations

The investigations conducted by the clinical research assistant at the 14 PHARE-M centers take place over the course of 2.5 consecutive days per CFC. The questionnaire is self-administered successively under the supervision of the clinical research associate according to a schedule established with the team at the site, with no possibility of communication or consultation among respondents. The questionnaires and responses are managed in SurveyMonkey Software and subsequently exploited using SAS and Excel Software. The focus group is conducted at the end of the visit. Each focus group is recorded using audio equipment and transcribed in writing.

4-1- Analyses of responses and validation of the questionnaire

Responses to the items of the questionnaire are processed anonymously. Each item receives a score on a Likert scale from one to four based on the degree to which the respondent agrees or disagrees with the proposition: “Completely disagree; Disagree; Agree; Completely agree”. "No" and "Unknown" responses are assigned a score of 0. The score is reset to 100 points and can thus be totaled by theme of the questionnaire and category of respondents. An initial descriptive analysis of the responses by CFC is returned to each quality team in the month following the on-site investigation, via a web conference, in order to validate the interpretation of the scores for the different themes and identify avenues for or obstacles to continuous care quality improvement at the CFC.

A Cronbach's alpha test will be performed on all responses collected at the centers. Since the anticipated number of respondents is around 130 people in total for the 14 teams, this test will not allow the questionnaire to be modified for use in a larger population of respondents. It

mainly aims to validate the French translations of the parts of the questionnaire coming from previous studies in English and discuss the use of the parts created within this research study.

A second level of descriptive analysis will be performed by aggregating the responses (all CFCs, by professional discipline, for resource patients/parents, and for professionals) to search for potential associations between quality of care at the CFC 3 years after the PHARE-M intervention and the effectiveness of the QT and/or the engagement of parents/patients and/or contextual elements.

After the publication of the Registry report presenting the 2015 data, changes in indicators from 2011 to 2015 will be crossed with the results of the realistic part of the study, in an attempt to identify any association in relation with more favorable changes over time in patient outcomes. A "signature" set of factors associated with a maximum/minimum impact of the PHARE-M will be sought.

4-2- Analyses of the content of the focus groups

The content of the focus groups will be exploited (coding, categorization), processed (analysis, validity), and interpreted according to the standard thematic content analysis protocol [128]. This will be done by grouping and counting within the framework developed during the pilot phase assessment.

4-3- Regulatory matters

Regulatory authorizations were granted for the quantitative research part focused on the patients' personal health data: a favorable opinion from the Ethics Committee of the Brest University Hospital (CHU) (session on 13 May 2014) and a notification of authorization by CNIL for a change in data processing stipulating the addition of a new recipient of the Registry data within the framework of a care quality improvement program (DR2015040 on 16 February 2015).

Conclusion/Discussion

Scope of the study and generalization

The research program aims at identifying the impact of the PHARE-M quality improvement program three years after the intervention at the 14 trained CFCs, situated in different organizational and cultural contexts. It uses a mixed methodology crossing the results of a quantitative analysis based on registry data and the results of a qualitative study designed in accordance with the recommendations for research on complex interventions.

The scope of the PHARE-M intervention and thus of the research concerns the management of a singular disease in a care network organized since 2002, which represents a relatively

controlled scope. Therefore, the influence of contextual elements on the PHARE-M program's impact can be analyzed independently from other confounding factors associated with different organizations for the management of various diseases or different hospital departments running diverse specialties.

Fourteen centers volunteered to engage and test the PHARE-M program; they were not randomized. Moreover, initial assessment highlighted that team motivation is a determinant of the speed of adherence to the program. This pattern of our research, focusing on an experimental phase having enlisted volunteer centers, is to be considered in interpreting the results and developing recommendations for a successful roll- out of the PHARE-M program in the national network.

Finally, the research study on the PHARE-M intervention has a study design that could be applied in the assessment of other complex interventions at healthcare settings. Hence, this research study could inform the assessment of interventions concerning the care of rare and/or chronic diseases and the instruments needed for such assessment.

Limitations identified and initial lessons

As a result of the experimental study based on Registry data, a study population paired between two groups (intervention and control) was defined to eliminate certain confounding factors, especially factors linked to patient age distribution. Despite this pairing, significant differences remained in terms of patient age at diagnosis and primary endpoint (FEV1%) between the two groups before the intervention, in favor of the intervention group. These initial differences could have a favorable effect for the rate of decline in FEV1% in four years in the intervention group [[129], [130]]. The question is to investigate whether the slopes are parallel or not. The difference in FEV1% will be taken into account using two different intercepts in the model, one for the intervention group and one for the control group. The patients belonging to either the "PHARE-M" group or the "Control" group will be identified in the Patient Registry with respect to their group for further analysis of their health outcomes.

Moreover, on-site quality audits of the Registry data included in the calculation of the primary endpoints showed discrepancies, mainly due to the CFCs' interpretation of the rule for selecting the values to transmit to the Registry [123]. The volume of the discrepancies identified in the data audited could be attributed to the change of the rule applied from the 2011 registry survey. This audit points out the need for a certification process to enable a larger use of this database in epidemiologic studies or for public health or pharmacovigilance purposes.

The survey conducted for the qualitative study of the multidisciplinary teams at the 14 centers should include around 130 respondents, including at most 14 patients/parents. This number of respondents might seem low for having enough statistical power in the statistical validation of the survey instruments, especially for the parts of the questionnaire developed within this research. The survey instruments could be improved within the framework of subsequent research studies aiming, for example, at comparing quality of care between centers trained in the PHARE-M and centers untrained in the program, or at making an assessment of the quality of care before/after another intervention. Therefore, this questionnaire represents an instrument that could have further uses in the network.

Expected results in terms of quality improvement of care

If the research study enables to identify factors promoting the adoption of the PHARE-M QIP and the maximization of its impact at CFCs, attention must be paid to the contextual elements to be worked on before or in parallel with the introduction of this program at the remaining CFCs. In the United States, the CFF has conducted "Leadership Collaborative" programs to develop leadership on multidisciplinary teams. The availability of the MDTs staff at the European standards for the number of patients followed could also represent a pre-requisite for their participation in the PHARE-M. The quality of care assessed after three years within the CFCs trained to PHARE-M might also enable to identify new avenues for improvement, including some beyond the scope of the clinical microsystem such as the Information System or the generalization of Guidelines.

Table I — Distribution by age, age at diagnosis and FEV1% of the 2012 study population between the two groups of the study cohort before pairing.

Comparison of the two groups	PHARE-M (N=1051)			Control (N=1962)		
Comparison of Ages	Avg.	Med.	Max.	Avg.	Med.	Max.
Age of patients (years)	15.0	13.0	62	18.0	17.0	74
Age at diagnosis (years)	2.0	0.1	51	3.2	0.2	71
Comparison of FEV1%	Avg.	LLM	ULM	Avg.	LLM	ULM
FEV1%	83	81,55	84,45	75,48	74,33	76,64

Table II — Comparison between the PHARE-M Group and the paired Control Group

Comparison between PHARE-M Group and Control Group		PHARE-M (N=1104)	Contrôles (N=1104)	Patients PHARE non paired (N=205)	Comparison between PHARE-M Group and Control Group (proc TTEST)
Gender	Men *n (%)*	582 (52.72)	564 (51.09)	93 (45.37)	
	Female *n (%)*	522 (47.28)	540 (48.91)	112 (54.63)	
Age	Average	15.57	16.05	14.48	
	Std Deviation	10.73	11.00	10.51	
Age (classes)	00-04 *n (%)*	182 (16.49)	175 (15.85)	32 (15.61)	
	05-09 *n (%)*	209 (18.93)	206 (18.66)	42 (20.49)	
	10-14 *n (%)*	213 (19.29)	204 (18.48)	48 (23.41)	
	15-19 *n (%)*	169 (15.31)	168 (15.22)	38 (18.54)	
	20-24 *n (%)*	125 (11.32)	130 (11.78)	19 (9.27)	
	25-29 *n (%)*	93 (8.42)	84 (7.61)	10 (4.88)	
	30-34 *n (%)*	53 (4.80)	68 (6.16)	4 (1.95)	
	35-39 *n (%)*	36 (3.26)	34 (3.08)	6 (2.93)	
	40-44 *n (%)*	7 (0.63)	18 (1.63)	1 (0.49)	
	45-49 *n (%)*	9 (0.82)	11 (1.00)	4 (1.95)	
	50-54 *n (%)*	4 (0.36)	3 (0.27)	0	
	55-59 *n (%)*	4 (0.36)	2 (0.18)	0	
	60-64 *n (%)*	0	0	1 (0.49)	
	70-74 *n (%)*	0	1 (0.09)	0	
VEMS	Nmiss	223	270	49	
	Average	83.00	79.11	85.06	p=0.0012 (S)
	Std Deviation	23.96	25.81	21.92	
ZBMI	Nmiss	7	39	2	
	Average	-0.17	-0.14	-0.18	p=0.5171 (NS)
	Std Deviation	1.05	1.15	1.11	

Table II (Followed): Comparison of Age at diagnosis between PHARE-M and Control

Age at diagnosis (years)

	Control	PHARE-M	Patients PHARE non paired
Nmiss	33	39	2
Average	2.49	1.85	2.47
Std Deviation	6.34	5.33	6.30

	P-value*
Comparison of Age at Diagnosis between PHARE-M and Control Groups	0.1317

***Test de Wilcoxon**

394 Table III — Criteria for quality of CF care derived from the chronic care model

IG — Improvement Goals at the CFC	1 — There are improvement goals at the CFC
	2 — These goals, if they exist, are the subject of both indicators and an action plan at the CFC
	3 — The CFC has tools to follow up this action plan in the form of a dashboard
	4 — To your knowledge, this action plan has been discussed with management and validated
SMS — Self-Management Support - Therapeutic Patient Education	1 — To your knowledge, there is a therapeutic education program for patients at the CFC authorized by the French regional health agency (ARS)
	2 — In your opinion, the professionals at the CFC are well trained in TPE
	3 — More than 80% of the patients/parents attended at least one TPE session in the last year
	4 — The total time spent by the professionals on TPE is sufficient
	5 — There are no obstacles to implementing TPE at the CFC
	6 — The team is involved in the studies of one of the French national groups on therapeutic education via face-to-face participation or regular reporting of information
	7 — The CFC has priority objectives for developing TPE
	8 — If yes, the CFC has indicators to follow up the achievement of these priority objectives
MM — Multidisciplinary management	1 — To your knowledge, the multidisciplinary team at the CFC comprises all the disciplines recommended by the French National Diagnosis and Treatment Protocol (PNDS): specialist physician, nurse, physiotherapist, psychologist, secretary, and social worker
	2 — The number of staff in all disciplines is sufficient for the number of patients followed up
	3 — In your view, the multidisciplinary team seems stable over time (the professionals' turnover rate is below 20% in a year)
	4 — The members of the multidisciplinary team have a great deal of expertise in managing cystic fibrosis
	5 — The multidisciplinary team meets often enough to perform a summary of the records of the patients who have come to the CFC
	6 — During these multidisciplinary meetings, the team generally reviews the records of the patients with a scheduled visit to the CFC
	7 — During these multidisciplinary meetings, the team regularly examines the patients' educational needs and the outcomes of the educational sessions held
	8 — The scheduled consultation is genuinely multidisciplinary: the patient meets with at least the physician, the nurse, and the physiotherapist
	9 — The scheduled consultation allows the patient to meet with a professional other than the ones mentioned above, as required (dietician, psychologist, or social worker)
	10 — The scheduled consultation allows the patient to benefit at least once per year from a TPE session on a priority objective for him or her
	11 — When a patient requires it, the CFC is able to call upon a network of referent professionals in other disciplines with knowledge of cystic fibrosis (geneticist, endocrinologist, ENT, gastroenterologist, etc.)

	12 — It is possible to be managed at the CFC on a 24/7 basis
	13 — Patients who arrive at the hospital emergency department are managed in accordance with a protocol established by the CFC with the emergency department for patients suffering from cystic fibrosis
	14 — The team regularly holds a meeting to discuss its functioning and the problems at the CFC in order to improve care management
DS — Therapeutic decision support (guidelines)	1 — The team manages the availability of guidelines (nutritional, respiratory, hygienic, etc.) in a way that they are accessible to all professionals
	2 — The team has defined an internal reporting procedure to insure that care management recommendations (guidelines) updates are accessible to the team
	3 — The team systematically verifies for each patient that the latest recommendations are applied and/or offered to him or her
	4 — The team uses alerts on the population followed up to verify that the latest recommendations for care are applied to the eligible patients (e.g. glucose tolerance test alert, vaccination alert, examination alert, etc.)
	5 — The team has optimally organized the multidisciplinary consultation process (circuit, schedules, chain of professionals, cross-contamination, hazards, etc.) to deliver high quality of care.
	6 — The team has optimally organized the process of responding to telephone or email messages from the patients and families
IS — Patient information system	1 — The team uses an electronic cystic fibrosis patient record
	2 — The team has an electronic patient record system that allows it to view changes in the patient health outcomes (nutritional and respiratory outcomes) over the course of several years
	3 — The team uses the electronic patient record system during the multidisciplinary staff meetings
	4 — The team displays information from the electronic patient record during the multidisciplinary meeting (graphs of changes over time, reports from previous consultations with different professionals, etc.)
	5 — The team uses the electronic patient record system both to create alerts on applying recommendations for the patient and to compile statistics on the population followed up
	6 — The team uses the electronic patient record system to include biology results
	7 — The team uses the electronic patient record system to include imaging results
	8 — The electronic patient record system helps in selecting patients for clinical trials
	9 — The electronic patient record data are automatically transmitted with a good degree of reliability (minimal verifications, corrections, and additions) to the French Cystic Fibrosis Registry
SN — Staff	1 — The CFC has organized a network of professionals in the patient community for managing care at home

	2 — The CFC organizes regular trainings for professionals in the patient community
	3 — The CFC regularly evaluates the professionals caring for CF patients in the community
	4 — The CFC assesses the health providers of devices managing CF patients
	5 — The CFC assesses the needs for home care and its distribution between professionals and carers for a balanced organization of home care
	6 — The CFC provides the patients with offers of sports activities, creative activities, and psychological support near their place of residence

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396 Table IV — Effectiveness of a quality team (QT)

Command of the quality process and tools	1. The teams that implement a quality process have a clear vision of the area on which to focus their improvement efforts and the expectations to be met. When you started the project, did you have such a vision?
	2. The quality teams sometimes use a method for making progress, such as a guide to follow step by step which helps them organize their work. Did your team use such a structured method?
	3. Did your team make one or more changes in its way of working?
	4. Did the team analyze data to ensure that such change(s) indeed represented an improvement?
	5. Did the team try to understand variations in the CFC processes and the reasons that could account for them (variations over time or between professionals, time of year, patient characteristics, etc.)?
	6. Does the team routinely have data allowing it to make a state of play and identify problems?
	7. Did the team have to develop a system to collect specific data (such as questionnaires, audits, interviews, or measurements) to identify problems and assess the responses provided?
	8. Did the team establish a data collection system to continue to manage quality or monitor the new processes established?
	9. Was the team able to rely on a referent professional to coordinate the meetings and work of the quality team?
	10. Was the team able to rely on a referent professional to collect and analyze data?
	1. The team was able to perform measurements to define and assess changes within the framework of tests.
Capacity to drive change	2. After testing a change, the team succeeded in discussing the outcomes observed and learning from this test.
	3. The team succeeded in analyzing the outcomes of the test to propose new changes or adjustments to be tested.
	4. During the process, the team was able to easily incorporate and adapt ideas for changes to meet the organization's needs.
	5. The team was able to enlist sufficient knowledge and skills to drive change under good conditions.
	6. The team could find sufficient assistance in the hospital to support changes.
	7. The team could sufficiently rely on the support of the French national team to make changes at the CFC.
E f	1. The performance of the PHARE-M steering team met my expectations.

	2. I was satisfied with my experience as a member of the quality team.
	3. I believe that my participation was useful and positive for the work of the team.
	4. I would be willing to participate again on a similar team to work on quality improvement.
	5. I believe that the work of the quality team was useful for improving quality.
	6. The outcomes achieved through the work of the quality team meet the organization's needs for improvement.
	7. It is necessary to maintain an ongoing quality improvement process to continuously improve care at the CFC.
Effectiveness perceived by the	1. I believe that the work of the steering team was useful for improving quality at the CFC.
	2. I believe that the entire team at the CFC was enlisted and contributed to quality improvement.
	3. I believe that the outcomes achieved collectively meet the organization's needs for improvement.
	4. I believe that it is necessary to maintain an ongoing quality improvement process to continuously improve management at the CFC.

397 Table V — Internal functioning of the quality team (QT)

Strictness of organization and clarity of roles	1. The leader was clear and explicit on how he or she wanted the team to work.
	2. The leader reviewed the steering team's work and asked how we were going to go about it.
	3. The leader also requested the opinion of the other members of the team.
	4. The leader's behavior reflected the importance he or she placed on the quality team functioning well.
	5. Our team could have been better at seeking help and securing more skills to do the work.
	6. Sometimes it seemed that we were working or going about the matter in the wrong way.
	7. Roles were so unclear that the work of different individuals seemed to overlap.
	8. The members of the team had different outlooks and experiences and came from different disciplines.
Decision- making on the QT	1. Most of the members of the team had an opportunity to participate in decision-making.
	2. We appreciated our differences, which shaped our decisions.
	3. The contribution of each member of the team was heard and taken into consideration.
	4. We examined many different ideas before making a decision.
	5. Our team possessed sufficient resources and skills and applied them well enough to work properly.
	6. Our team worked well enough to accomplish its mission satisfactorily.
Clarity of objectives	1. The members of the team were in agreement on the objectives of the project.
	2. The achievement of the objectives guided the activities of the members of the team.
	3. The members of the team did what was expected of them.
	4. The members of the team were all focused on the achievement of the same objectives.
Communication and cooperation	1. There was a great deal of cooperation between the different hospital departments.
	2. In this hospital, most departments and services have a hard time sitting down at a table and solving problems together.

	3. The people I worked with were comfortable with suggesting changes and improvements.
	4. Our team received all the information required to plan and organize its work.

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399 Table VI — Engagement of the patients/parents on the quality team (QT)

Information and activation of the patients/parents	1. The patients and parents are educated regularly (annually or more often) by the team about general subjects concerning cystic fibrosis care and research.
	2. The patients and parents are rather familiar with general cystic fibrosis information: research, progress made, and Registry data.
	3. The CFC team has educated the patients and parents about the PHARE-M's importance and aim.
	4. A good relationship between the patient or parent recruited and the team is indispensable for the patient or parent to participate in the PHARE-M.
	5. The patient or parent recruited is well informed of the challenges (10 commitments) of management quality.
	6. The presence of a patient or parent on the steering team is a given and an asset.
	7. The place of a parent or patient is not on a quality team, because he or she does not have enough training or education.
	8. The place of a parent or patient is not on a quality team, because he or she already has too many personal problems to manage.
	9. The patient or parent recruited possesses the qualities to become a member of the steering team.
	10. The patient or parent recruited must have developed coping skills (see therapeutic education standard: knowing how to manage emotions and stress; solving problems, making decisions, and making choices; knowing how to communicate and being adept in relationships with others; and knowing how to put oneself in the place of others).

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Empowerment of patients/parents to allow them participate in the QT	1. The participation of a patient or parent depends on the systematic reimbursement of his or her travel expenses.
	2. The participation of a patient or parent should be facilitated by the reimbursement of other expenses: child-care, lost working hours, etc.
	3. The participating patient or parent does not represent all patients.
	4. The patient or parent was selected by the team based on a list of specific criteria (cultural level, capacity to communicate, availability, etc.).
	5. The patient or parent is motivated to improve management for all.
	6. The patient or parent is also motivated to improve his or her own management by participating in the program.
	7. It is important to communicate with the other patients or parents concerning the role of the patient or parent on the steering team.

	8. It would be necessary to include several patients or parents to ensure that more different points of view are represented.
	9. The patient or parent must be knowledgeable about the disease and its management beyond the requirements of his or her own care.
	10. The patient or parent must be knowledgeable about the general functioning of the hospital.
	11. The patient or parent must know how to communicate with the professionals by taking a step back and drawing general lessons from his or her own experience.
Capacity for effective contribution of the patients/parent	1. The PHARE-M national organization created good conditions for incorporation of the patient or parent.
	2. The participation of a patient or parent on the team at French national training and information meetings (four French national face-to-face "EPE" meetings) is indispensable.
	3. The patient or parent participated and contributed as much as the professionals during the French national "EPE" meetings.
	4. The patient or parent's regular participation at quality team meetings at the CFC is indispensable.
	5. The patient or parent participates in and contributes significantly to the work of the steering team.
	6. The patient or parent's ideas and proposals are generally taken into account by the steering team.
	7. The atmosphere of work of the steering team meeting is better and more productive when the patient or parent is present.
	8. The pace of work is slower when the patient or parent is present at the steering team meeting.
	9. Certain decisions made by the steering team are inspired by the patient/parent.
	10. The process of incorporation and participation of the patient or parent should be reviewed and improved for the continuation of the PHARE-M.

Table VII — Organizational culture

Organizational culture: Research studies have defined four types of organizational culture, arising from both the organization's external environment and internal management: a "familial" type, an "entrepreneurial" type, a "prescriptive" type, and a "productive" type. The five rubrics below describe the characteristics associated with these different types of organization. You have 100 points to distribute among the four proposals based on the degree to which they resemble your organization. For example: If the CFC resembles Description A a great deal and Description B a little, and does not resemble Description C or Description D at all, assign 70 points to Response A and the 30 remaining points to Response B.	
§1. Character	1. Organization A is very familial, like a big family. People seem to share a lot of themselves.
	2. Organization B is very dynamic and entrepreneurial. People seem to want to venture off the beaten path and take risks.
	3. Organization C is very structured and formalized. Procedures govern people's work.
	4. Organization D is very focused on production, with the concern being that the work gets done. Individuals are not very personally involved.
§ 2	5. Organization A's director(s) are warm and attentive. They try to develop people's potential and act as

	mentors or guides.
	6. Organization B's director(s) take risks. They encourage people to be innovative and to try out new ideas by taking risks.
	7. Organization C's director(s) enforce rules. They expect people to strictly apply policies and procedures.
	8. Organization D's director(s) resemble coordinating coaches. They help people achieve the organization's objectives.
§3. Cohesion	9. Organization A's factors for cohesion are loyalty and tradition. Dedication to the organization is high.
	10. Organization B's factors for cohesion are the race for innovation and development. There is a desire to be the first.
	11. Organization C's factors for cohesion are hierarchical rules and establishment policies. Maintaining suitable functioning is important here.
	12. Organization D's factors for cohesion are the achievement of objectives and the performance of required tasks. This vision of production is shared.
§4. Emphasis placed on...	13. Organization A emphasizes human resources. Having strong cohesion and a high sense of morale are important.
	14. Organization B emphasizes growth and acquisition of new resources. Being ready to rise to new challenges is important.
	15. Organization C emphasizes permanence and stability. Complying with rules and performing operations smoothly are important.
	16. Organization D emphasizes competition to achieve objectives. Measuring results is important.
§5. Recognition of efforts	17. Organization A recognizes all its members' efforts equally. It is important that everybody in the pyramid, from the very top to the very bottom, is treated as equally as possible.
	18. Organization B rewards individual initiative. Those who have the most ideas and perform the most innovative actions receive the most recognition.
	19. Organization C modulates recognition based on rank. The higher your position, the more your efforts are recognized.
	20. Organization D rewards the achievement of objectives. Individuals who demonstrate leadership and thus help achieve objectives are recognized.

Table VIII — Patient-oriented culture and leadership

Patient-oriented organization	1. Our organization works to properly identify patient needs and expectations.
	2. The professionals handle patient requests promptly.
	3. Patient complaints are analyzed to identify recurring causes and prevent problems from being replicated.
	4. The organization uses data from the patients themselves to improve services.
	5. The organization uses data regarding patient satisfaction and/or patient expectations to improve services.
Leadership at the CFC	1. The leader develops interesting/exciting opportunities for our organization.
	2. The leader proposes new and even innovative ideas to improve management services and processes.
	3. The leader drives the organization to meet patient needs and ensures management/care safety.
	4. The leader takes into account the needs of both the service and the staff during major changes within the organization.
	5. The leader builds close, positive relationships with the other departments in the hospital.
	6. The leader builds close cooperative relationships with other organizations outside the hospital.

Table IX — Open-ended questions to the hospital's quality department

1. What are the priorities of the hospital's quality department?
2. Support for care services in quality improvement: was another quality program developed for another disease or another care service?
3. How are patients included in the different committees and groups working to improve quality in the hospital?
4. How is quality measured (main indicators)?
5. What training programs in quality tools and methods are promoted by the hospital?
6. How was the quality department informed of the PHARE-M (by whom and when)?
7. What were the reasons for the quality department's engagement (or non-engagement) in the PHARE-M, in support of the CFC? In the case of engagement, what resources and time were dedicated to supporting the CFC?
8. How is the PHARE-M perceived by the quality department management in terms of coherence with hospital policy, perceived effectiveness, and other matters? If necessary, the example of another quality improvement program rolled out in the hospital can be cited.

Figure 1 — Distribution by population age between the two groups (PHARE-M and control), paired in 2012 data.

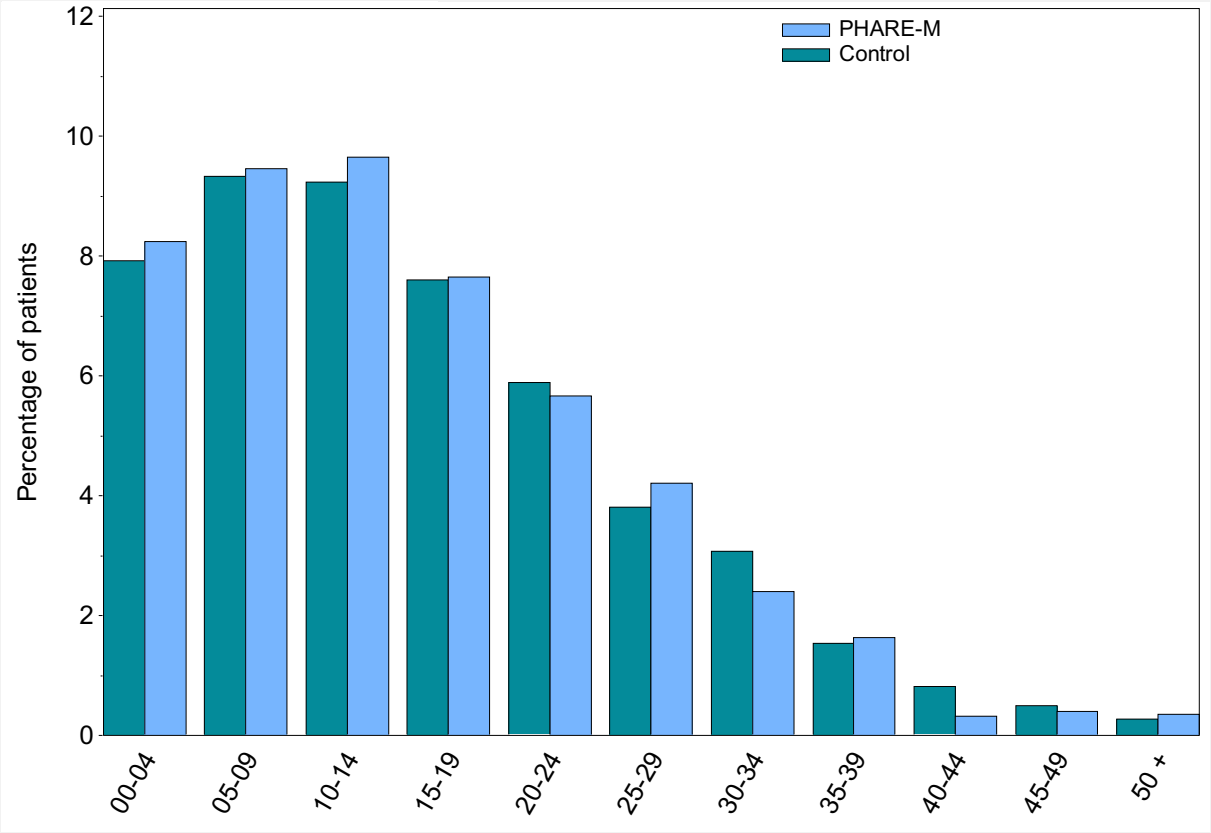


Figure 2 — Representations of the analysis of the primary endpoint

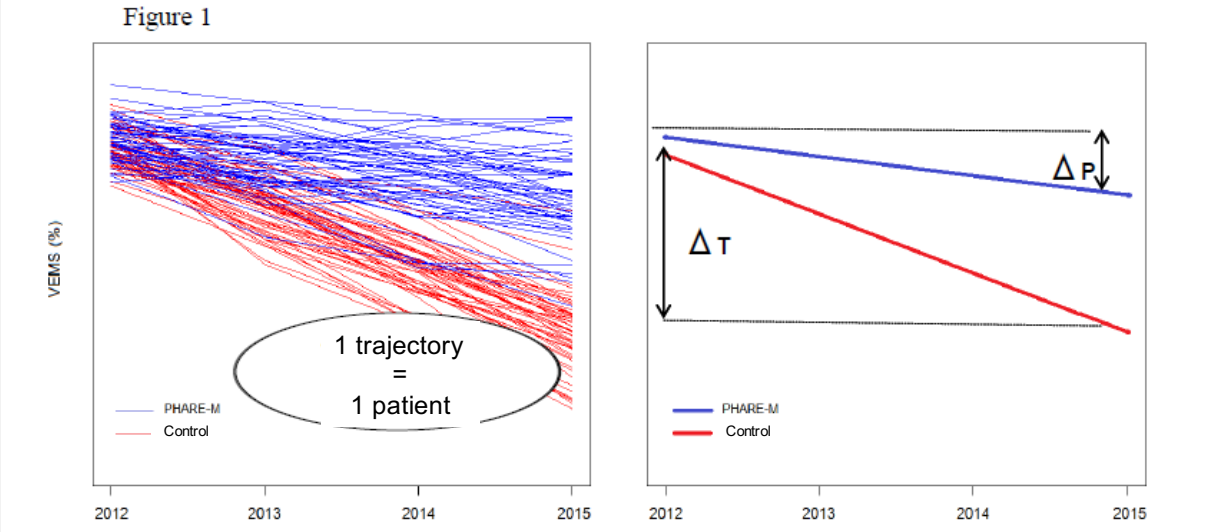
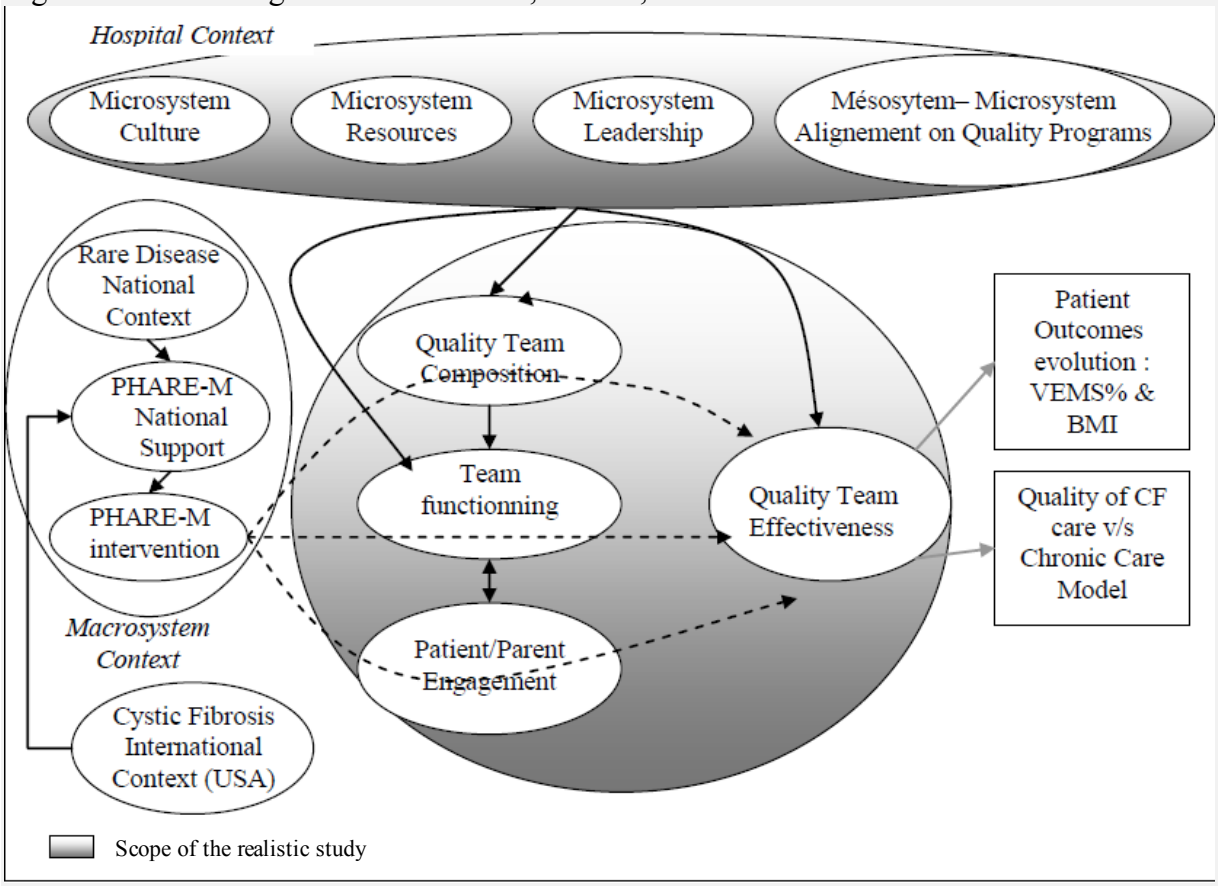


Figure 3 — Modeling of the intervention, context, and mechanisms.



V-3. L'analyse de la participation des patients/parents

La participation des patients/parents a été analysée dans le cadre général de la recherche sur l'évaluation de la performance de la démarche qualité PHARE-M.

Deux aspects spécifiques du dispositif de l'enquête réaliste ont été intégrés pour permettre cette analyse : 1) les ***conditions de la participation des patients et parents*** au cours du programme ont été explicitement analysées en tant que mécanisme pouvant moduler l'efficacité de l'équipe de pilotage ; 2) les ***réponses des patients et parents des équipes de pilotage à l'ensemble des items du questionnaire d'enquête*** ont été recueillies au même titre que les réponses des professionnels de ces équipes.

Ces dispositions ont permis d'analyser les réponses des professionnels et des patients/parents à l'ensemble des items et en totalisant les 14 CRCM : le groupe des patients/parents (N=12 répondants) et le groupe des professionnels (N=64 répondants dans toutes les disciplines). Ainsi les réponses apportées aux questions de recherche sur la participation des patients et parents font état des ***consensus et dissensus*** au sein de chaque groupe et entre les deux groupes de répondants.

Un premier niveau d'analyse a permis d'identifier les assertions recueillant un fort consensus dans chacun des deux groupes de répondants (unanimité ou > 80% de voix), qu'il soit positif (accord), négatif (désaccord) ou neutre (ni-ni ou NSP).

Un second niveau d'analyse a permis d'identifier les items faisant l'objet d'un consensus ou un dissensus significatif entre les deux groupes de répondants d'après le Test Exact de Fisher (131).

Les résultats obtenus ont pu être classés en 4 catégories (cf. Tableau II) :

1) items recueillant un consensus dans chaque groupe de répondants et un consensus entre les 2 groupes de répondants, dans le même sens positif (+) ou négatif (-) ou neutre (N) ;
2) items recueillant un consensus dans le groupe patients/parents uniquement, ces items n'ayant pas obtenu de consensus dans le groupe des professionnels (mention NC) ;
3) items recueillant un consensus dans le groupe des professionnels uniquement, ces items n'ayant pas obtenu de consensus dans le groupe des patients/parents (mention NC) ;
4) items ne recueillant aucun consensus dans aucun des deux groupes (NC,NC).

Tableau II : Présentation des items selon le degré de consensus

Classement des Items selon le degré de consensus	Consensus entre P&P	Pas de consensus (NC) entre P&P
Consensus entre Professionnels	1) (+,+) or (-,-) or (N,N)	3) (NC, +) or (NC, -) or (NC, N)
Pas de consensus (NC) entre Professionnels	2) (+,NC) or (-,NC) or (N,NC)	4) (NC, NC)

L'absence de consensus sur un certain nombre d'items n'a pas été explorée dans le cadre de ces analyses, mais renforce l'hypothèse d'un « effet centre » qui sera analysé dans la suite de l'étude réaliste, sans distinction des deux groupes de répondants. Compte-tenu du petit nombre de répondants et leur appartenance à différents CRCM, les réponses **du** patient ou parent n'ont pas été comparées aux réponses des professionnels de l'équipe du CRCM.

V-4. Les analyses complémentaires de la recherche

L'analyse ***statistique quantitative*** des effets biocliniques est prévue en 2017, suite à la publication en mars 2017 des données 2015 du Registre Français de la Mucoviscidose : une communication écrite préliminaire au congrès nord-américain sera présentée en octobre 2017.

L'analyse ***réaliste*** sur le périmètre des 14 CRCM PHARE-M sera finalisée en 2018 et deux communications sont prévues :

1) la présentation de la validation statistique du questionnaire de l'enquête suite à l'analyse Alpha de Cronbach, réalisée à partir de l'ensemble des réponses des CRCM PHARE-M ;
2) l'analyse de la variabilité d'impact du programme entre les CRCM PHARE-M en 2015, croisée avec les éléments de contexte et les mécanismes observés pour cette intervention.

VI- LA CHRONOLOGIE DES TRAVAUX REALISES

Le tableau III ci-après résume les travaux réalisés entre 2010 et 2017, concernant :

- l'intervention PHARE-M,
- les travaux de recherche dans le cadre du programme PHARE-M Performance
- les travaux spécifiques pour la thèse,
- les diverses communications à des congrès nationaux et internationaux et la liste des articles du supplément de l'Orphanet Journal for Rare Diseases, relatant l'expérience du PHARE-M, publiés en 2017 (ou acceptés pour publication par la revue).

L'intervention PHARE-M

La phase d'***introduction*** de la démarche qualité en France, par transposition du programme mis au point par la CF Foundation et le Dartmouth Institute Microsystem Academy, s'est déroulée de septembre 2011 à Juin 2013. Elle a concerné 14 équipes de CRCM, volontaires pour tester et ajuster la démarche au contexte français. Suite aux deux évaluations externes réalisées en 2012, et à la contribution des équipes engagées au cours de la phase expérimentale, une version finale du programme de formation a été élaborée fin 2013 avec un calendrier de formation repositionné sur l'année civile.

En vue de pérenniser le programme au sein de la filière mucoviscidose, il a été décidé de le normaliser dans le contexte de la formation continue hospitalière et dans le cadre des orientations et méthodes de développement professionnel continu définies par la HAS. Une candidature pour la reconnaissance du PHARE-M en programme DPC a été déposée dès la phase transitoire du DPC, en 2014, par l'organisme de formation de la Fondation ildys.

Cette candidature a reçu fin 2014 un accord transitoire qui a permis de réaliser la session 2015 du programme au titre de la formation continue hospitalière. La Commission Scientifique Indépendante paramédicale a donné son agrément définitif en février 2015.

La mise à jour des orientations et méthodes de DPC par la HAS en décembre 2015 n'a pas permis de remplir les démarches administratives pour la session annuelle 2016 du programme PHARE-M, qui s'est déroulée hors DPC. Suite à l'agrément définitif de la CSI médicale en janvier 2016 et à la réponse positive de l'OGDPC sur les éléments du programme DPC 2015, le PHARE-M a pu être inscrit comme programme de DPC pour les sessions 2017 et suivantes (agrément délivré jusqu'en 2021).

L'objectif de l'inscription du PHARE-M en DPC est prioritairement d'inciter les équipes à s'inscrire en permettant la reconnaissance de leurs compétences et de leurs efforts pour améliorer la qualité des soins, tout en étant un moyen de financer les coûts de la formation. L'association Vaincre la Mucoviscidose accorde en complément un financement pour un temps supplémentaire (0,20 ETP) de soignant paramédical chargé d'animer l'équipe de pilotage du CRCM et les frais de déplacement des personnels qui n'ont pas obtenu l'autorisation de s'inscrire au DPC.

Le projet de recherche PHARE-M Performance

Les travaux ont été structurés dans le cadre du programme de recherche déposé et financé par le PRePS avec le design mixte de l'étude exposé plus haut.

Pour mener à bien l'ensemble des travaux liés à la recherche, la doctorante a initié une collaboration avec :

- le Bureau méthodologique et statistique du CIC du CHU de Brest, pour la méthodologie et les validations statistiques
- une Attachée de Recherche Clinique (ARC) recrutée, pour les investigations sur site des CRCM grâce au budget du programme de recherche
- un groupe d'experts du LEPS et du Centre de Référence Mucoviscidose de Nantes-Roscoff, pour l'élaboration du questionnaire d'enquête de l'étude réaliste
- un groupe d'experts internationaux issus du groupe éditorial du BMJ Quality & Safety et du Dartmouth Institute, pour la formation aux Standards for QUality Improvement Reporting Excellence (SQUIRE) et au coaching d'Ateliers d'écriture

Les principales étapes de l'étude réaliste qui fait plus particulièrement l'objet de la thèse ont été :

- l'élaboration du questionnaire d'enquête, à partir de :
    - questionnaires déjà publiés pour certaines parties, et en développant une adaptation de cadres publiés sur d'autres parties (CCM et Patient Engagement)
    - la relecture et validation par un groupe d'experts constitué autour de la doctorante par des représentants du LEPS et du Centre de Référence Mucoviscidose de Nantes-Roscoff
    - le test de ce questionnaire par une Attachée de Recherche Clinique (ARC) auprès des équipes des CRCM faisant partie du CRMR de Nantes-Roscoff (4 CRCM ; professionnels et patients-parents engagés dans les équipes de pilotage)
    - l'ajustement du questionnaire et la validation de sa structure finale par des experts des questionnaires d'enquête
- la passation de l'enquête par l'ARC auprès de l'ensemble des professionnels et des patients et parents engagés dans les équipes de pilotage ainsi que les membres des équipes pluridisciplinaires de ces CRCM, entre Octobre 2014 et juin 2015 :
    - l'ARC dédiée à l'étude a organisé ses visites sur site, pour mener le contrôle qualité des données du CRCM transmises au Registre (volet quantitatif de la recherche) et la passation du questionnaire, ainsi que la tenue des focus group,
    - les conditions de réponse à l'enquête en présence de l'ARC devaient garantir l'expression indépendante et sans influence réciproque des points de vue des interviewés : planification de l'ordre de passage individuel par l'ARC (durée aménagée 1h), réponse sur Survey Monkey avec possibilité de poser une question à l'ARC sur la compréhension des items si besoin, pas d'échanges entre les répondants avant leur passage, pas de retour aux répondants sur leurs réponses individuelles, anonymat garanti de l'exploitation des réponses ;
- la restitution des résultats globalisés à l'équipe de pilotage de chaque CRCM, par une réunion Webex environ un mois après la visite sur site, en vue d'obtenir leur réaction sur la fidélité de la représentation de leur groupe, et de dégager de nouvelles pistes d'amélioration de la qualité des soins dans le cadre de la démarche qualité continue
- la validation du questionnaire menée à l'issue de la passation de l'enquête avec l'ensemble des réponses recueillies, par le statisticien du CIC du CHU de Brest

afin de dégager les dimensions et la cohérence interne (alpha de Cronbach) des parties du questionnaire, notamment des parties créées pour la recherche
- L'analyse des résultats quantitatifs à partir des données du Registre Français de la mucoviscidose, dès la mise à disposition des données 2015 en 2017 ainsi que les résultats complets de l'étude réaliste seront produits courant 2018.

La thèse de doctorat

La thèse de doctorat, engagée dès l'année de démarrage du projet de recherche, est centrée sur l'apport du partenariat des patients et parents dans le cadre du programme PHARE-M. Les échanges scientifiques au sein du LEPS au cours des années de doctorat ont permis d'approfondir la réflexion et d'enrichir l'analyse. Les principaux échanges ont concerné :

- les réunions mensuelles des doctorants du Laboratoire sur divers sujets en lien avec les recherches en cours sur la place et le rôle des usagers dans le système de soins, l'éducation thérapeutique et l'empouvoirement des patients, l'implication des patients dans les cursus de formation des soignants
- la prise de connaissance et mise en œuvre des UK MRC Guidelines pour l'évaluation des interventions complexes (97)
- une journée d'échanges scientifiques en Ateliers autour de la participation des patients à l'amélioration des soins (132) et plus particulièrement l'animation d'un Atelier sur la participation des patients aux démarches qualité des soins
- l'analyse prioritaire des données de l'enquête de l'étude réaliste au regard du sujet de la thèse, en vue de la publication des résultats dans le délai de la thèse
- une journée de réflexion scientifique commune avec les représentants du CIUSSS de Sherbrooke autour des théories et des pratiques entourant a) la participation et l'engagement des usagers et de la population au sein des services de santé et des services sociaux et b) l'accompagnement à l'autonomisation des usagers en santé.

Valorisation et Communication

L'objectif de publication des travaux relatifs au programme qualité PHARE-M et à la recherche sur la performance de ce programme, y compris les travaux menés dans le cadre de la thèse, a été essentiel dans le cadre de ce cheminement de 4 années.

L'expérimentation de cette démarche unique en France, dans une filière maladie rare intégrée dans une communauté internationale dynamique, l'implication de parents et patients dans cette démarche, l'application de la méthodologie d'évaluation réaliste à une intervention complexe dans le cadre d'un design mixte d'étude et l'utilisation de cadres d'évaluation peu utilisés en France (le Chronic Care Model, le fonctionnement du travail en équipe, l'efficacité de la démarche et le degré d'engagement des patients et parents), sont apparus très vite comme autant d'éléments originaux devant être portés à la connaissance de la communauté scientifique ainsi que de la communauté des soignants et des représentants des patients.

Cet objectif s'est concrétisé par :

a) Des communications, écrites ou orales, dans des congrès nationaux ou internationaux, permettant des rencontres avec la communauté scientifique et soignante, sur la synergie entre l'éducation thérapeutique et la démarche qualité

des soins, la démarche qualité dans la communauté mucoviscidose internationale, ou la recherche sur les démarches qualité des soins

b) ***La contribution à un groupe de travail de l'European Cystic Fibrosis Society sur la révision des Standards of Care*** datant de 2005 a inclut un groupe de travail sur l'amélioration de la qualité des soins incluant des patients et des parents et donné l'occasion de présenter l'approche collaborative développée en France aux autres membres de la société européenne (133)
c) ***Une intervention dans le QM Training Course*** lors des conférences européennes annuelles organisées par l'ECFS (2015, 2016) et la contribution au QM e-training course (2017)
d) ***Une formation de la doctorante aux standards internationaux de publication SQUIRE 2 :*** International Writing Conference, Hanover, USA) animée par BMJ Quality & Safety et le Dartmouth Institute en novembre 2015
e) ***L'animation d'ateliers d'écriture par la doctorante*** avec les auteurs des autres articles de la revue OJRD, pour l'aide à l'écriture et la révision d'articles dans le processus de revue
f) ***La préparation d'un numéro spécial de l'OJRD*** (Orphanet Journal for Rare Diseases – BioMedCentral online) relatant l'expérience de la phase expérimentale du PHARE-M (Tab III):
   - 2 articles de présentation du programme national et de sa transposition à partir du programme développé aux USA,
   - 3 articles de présentation d'expériences d'équipes de CRCM centrées sur des objectifs d'amélioration différents et variés avec les résultats obtenus à 3 ans,
   - 2 articles décrivant le protocole de recherche et le contrôle qualité des données sur site,
   - 1 article de synthèse sur la participation des patients et parents

Tableau III : Liste des articles du supplément PHARE-M de l'OJRD (Volume 2)

N°	Article
Edito	History, context and spirit of the French CF QIP
1	Trans-Atlantic Collaboration: Applying Lessons Learned from the US CF Foundation Quality Improvement Initiative
2 (III)	Introduction of a Collaborative Quality Improvement Program in the French Cystic Fibrosis Network: the PHARE-M initiative
3	A Quality Improvement Program to improve nutritional status of children with Cystic Fibrosis aged 2-12 years old over a 3 year period at CF center Roscoff , Brittany
4	A quality improvement program for adolescents with cystic fibrosis: focus on psychosocial skills.
5	A Quality Improvement Program to Reduce the time on the lung transplant waiting list at the Nantes University Hospital
6 (IV)	Quality of care in cystic fibrosis: assessment protocol of the French QIP PHARE-M
7	Lessons learned from on-site quality control of data transmitted to the French CF Registry
8 (V)	Lessons from patient and parent involvement in a Quality Improvement Program in Cystic Fibrosis care in France

Tab IV : Chronologie des travaux réalisés

<table>
<tr><th colspan="19">CHRONOLOGIE DES TRAVAUX - DEMARCHE QUALITE PHARE-M & RECHERCHE PHARE-M PERFORMANCE</th></tr>
<tr><th>2010</th><th colspan="3">2011</th><th colspan="3">2012</th><th colspan="2">2013</th><th colspan="2">2014</th><th colspan="3">2015</th><th colspan="2">2016</th><th colspan="2">2017</th><th>2018</th></tr>
<tr><td colspan="19">Programme de formation-action PHARE-M</td></tr>
<tr>
<td>Mobilisation des acteurs de la Filière: Association et Société Française Mucoviscidose</td>
<td>Formation Démarche Qualité LLC - Dartmouth Institute - Avril-Mai</td>
<td>Traduction-Adaptation des documents de formation Mai-Août</td>
<td colspan="2">PHARE-M Session1: 7 équipes de CRCM formées Sept 2011-Juin 2012</td>
<td>Suite à évaluation externe: Révision des documents et du curriculum de formation Juil-Août</td>
<td colspan="2">PHARE-M Session2: 7 équipes de CRCM formées Sept 2011-Juin 2012</td>
<td colspan="3">Standardisation du programme PHARE-M: Curriculum année civile, Candidature OGDPC, Agrément DPC transitoire</td>
<td colspan="3">DPC PHARE-M Session1: 3 équipes pédiatriques de CRCM formées et 1 équipe mixte/adulte Janv-Déc 2015</td>
<td colspan="2">PHARE-M Session3: 3 équipes de CRCM pédiatriques formées Janv-Déc 2016
Hors DPC : Révision des orientations du DPC par la HAS

Agrément définitif par les CSI médicale et paramédicale du DPC PHARE-M jusqu'en 2021</td>
<td colspan="2">DPC PHARE-M Session2: 2 équipes de CRCM adultes formées Janv-Déc 2017</td>
<td>DPC PHARE-M Session3: 3 équipes de l'Ile de La Réunion engagées; équipes de Métropole en cours de recrutement Janv-Déc 2018</td>
</tr>
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<td rowspan="2">Design étude quantitative avec le CIC CHU Brest</td>
<td colspan="10">Thèse de doctorat</td>
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<td colspan="2">Design Evaluation Réaliste: Définition du questionnaire d'enquête, Test auprès de 4 CRCM et Elaboration du questionnaire définitif</td>
<td colspan="2">Investigations sur Sites CRCM & Restitutions aux équipes de leurs résultats: Contrôle qualité des données transmises au Registre et réponses au questionnaire d'enquête & Identification de nouvelles pistes d'amélioration de la qualité</td>
<td colspan="2">Validation Alpha-Cronbach du questionnaire d'enquête

Formation SQUIRE International Writing Conference, Hanover, USA</td>
<td colspan="2">Analyse des résultats de l'enquête pour la participation des patients et parents au programme PHARE-M

Animation de l'Atelier LEPS sur la collaboration des patients dans la qualité des soins</td>
<td colspan="2">Etude quantitative: Analyse des données 2011 à 2015 du Registre Français de la Mucoviscidose

Rencontre Scientifique LEPS - CIUSSS Sherbrooke: Participation des usagers et de la population au sein des services de santé et des services sociaux et accompagnement à l'autonomisation des usagers</td>
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<td>Publication Revue des Maladies Respiratoires: Programme d'accompagnement des patients et familles par les pairs</td>
<td>Congrès SETE, Bobigny: Poster PHARE-M & Education Thérapeutique du Patient</td>
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<td>BMJ Quality & Safety, Londres: Poster Phare-M Performance Protocol; Revue Santé Publique Analyse de la prise en chaege ambulatoire de la mucoviscidose</td>
<td>Congrès de la Société Française de Pédiatrie, Tours: communication orale & publication Ethique de la coopération parents-soignants</td>
<td>NACFC, Phoenix: Poster Phare-M Performance Study design</td>
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Préparation de 4 articles de synthèse dans le supplément1 de l'OJRD;

Rédaction de l'article Revue SOINS Vers une participation active des usagers dans les démarches qualité</td>
<td>Finalisation des articles de synthèse du supplément1 OJRD</td>
<td>NACFC, Indianapolis: Poster PhareM Performance on Patient Health Indicators;

Préparation de l'article de synthèse de l'étude quantitative</td>
<td>Préparation des articles de synthèse de l'étude réaliste: Validation Alpha-Cronbach du questionnaire; Article de synthèse des résultats</td>
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VII- SYNTHESE DES RESULTATS

Les résultats résumés ci-dessous proviennent d'une part des observations réalisées au cours de la première session pilote du PHARE-M par l'évaluateur externe, d'autre part des réponses au questionnaire d'enquête des membres des équipes de pilotage, professionnels et patients ou parents. Les affirmations rapportées ont recueilli un fort consensus auprès du public indiqué (> 80% de réponses concordantes sur l'item) : pour alléger le texte, l'indication de « *fort consensus* » n'est pas répétée, seuls les *non consensus* sont explicitement mentionnés comme tels. Pour une présentation plus détaillée de ces résultats, se reporter à l'article V (Volume 2) et page et suivantes

Les résultats sont présentés ci-après rattachés à l'objectif spécifique auquel ils se rapportent : pour chaque objectif spécifique, ils éclairent une thématique explorée dans le travail de recherche.

OS1 : Evaluer les conditions mises en place pour permettre la participation des patients/parents dans le programme PHARE-M et dans la démarche continue d'amélioration de la qualité *(empowerment)*

Les conditions de la participation proposées dans le cadre du programme ont été analysées selon trois rubriques, correspondant au ***cadre de Carman*** (106).

VII-1. L'activation et le recrutement des patients et parents

Si les professionnels indiquent informer régulièrement (a minima une fois par an lors d'une réunion formelle) les patients ou parents du CRCM sur les sujets concernant la maladie, la recherche et les traitements, les patients et parents des équipes de pilotage ont des avis partagés sur le sujet. Les patients et parents s'informent donc sur ces thèmes par d'autres canaux – associations via des publications papier ou internet ou l'Assemblée Générale (AG) annuelle de Vaincre la Mucoviscidose, réseaux sociaux, site e-corn CF qui propose des réponses d'experts à des questions individuelles, ou au cours d'échanges individuels avec leurs soignants lors des consultations. Une présentation du programme PHARE-M avait été réalisée devant les adhérents de l'association réunis en AG en mars 2011, mais cette information n'avait pas été relayée ultérieurement dans les différents canaux destinés aux patients et parents.

La procédure de recrutement d'un patient ou parent par les soignants, à l'aide d'une feuille de candidature explicitant les critères à remplir (disponibilité, nécessité de participation à la formation nationale, accès à internet, participation à distance ou physique aux réunions locales…) a facilité cette étape pour l'équipe soignante. Les qualités jugées nécessaires par les soignants pour le choix du patient ou parent ont été en premier lieu, la « qualité de la relation avec l'équipe pluridisciplinaire » (critère partagé par les patients et parents), puis des « qualités particulières » dont le fait « d'avoir pris du recul par rapport à son expérience singulière avec la maladie ». Les patients et parents ont exprimé la nécessité d'avoir développé des « compétences d'adaptation » telles que décrites par l'éducation thérapeutique (ces compétences étaient explicitées dans la rédaction de l'item), alors que ce critère n'a pas rencontré de consensus chez les soignants. Il faut noter qu'entre 2011 et 2013, tous les soignants n'étaient pas formés à l'éducation thérapeutique et donc tous ne la pratiquaient pas. Les patients et parents recrutés ont été reconnus par les deux

groupes de répondants comme ayant une forte motivation pour améliorer la qualité des soins pour TOUS les patients, au-delà de leur motivation à améliorer les soins pour eux-mêmes – assertion consensuelle uniquement chez les professionnels.

Tous ont exprimé l'importance de bien informer le patient ou le parent, préalablement à son recrutement, sur les objectifs du programme PHARE-M et sur les conditions de sa participation. Aucune équipe n'a mentionné de difficulté au recrutement. Certaines ont mentionné avoir dû choisir entre plusieurs candidatures exprimées et n'avoir associé qu'un seul des candidats « officiellement », mais plusieurs candidats lors des réunions locales, selon les sujets abordés.

VII-2. L'empouvoirement des patients et parents

L'empouvoirement a été caractérisé par : la participation des patients et parents aux réunions de formation nationales – afin de leur donner la même compréhension de la démarche qu'aux soignants ; la présentation, aux autres patients/parents, de leur rôle au sein de l'équipe de pilotage PHARE-M – afin d'éviter de créer des tensions entre les autres patients ou parents et l'équipe soignante ; le remboursement de leurs frais de déplacement (par l'association) pour les déplacements aux réunions de formation à Paris ou sur un site de CRCM (*benchmarking visit*) – dans les mêmes conditions que pour les professionnels des équipes.

La nécessité de leur participation aux réunions nationales de formation n'a pas recueilli de consensus dans aucun des deux groupes. Précisons néanmoins que les réunions de formation sont aussi le lieu d'exercice de la méthode, en prenant la situation du CRCM comme objet d'étude de cas. De nombreuses idées sont ainsi débattues en suivant la méthode, idées qui seront approfondies ensuite dans les réunions locales. Sur cet item, une maman qui a succédé à un parent qui s'était retiré au bout de l'année de formation a exprimé, lors du focus group avec l'équipe, qu'elle aurait mieux compris la démarche si elle avait pu participer à cette formation nationale – elle est par ailleurs ingénieur qualité dans l'industrie.

Les deux groupes ont souligné l'importance d'informer les autres patients ou parents du CRCM sur la participation et le rôle du patient ou parent recruté dans l'équipe. Certains CRCM animaient depuis plusieurs années un « collectif de parents » et des informations régulières ont été réalisées avec ce groupe de parents actifs. Il est arrivé qu'un parent du collectif soit le parent recruté dans l'équipe de pilotage, sans que ce soit le cas général. A l'inverse certains parents recrutés dans PHARE-M se sont par la suite investis dans le collectif de leur CRCM ou dans une instance régionale de l'association.

La nécessité de rembourser les frais de déplacement du patient ou parent n'était pas consensuel parmi les soignants – alors qu'elle l'était parmi les patients et parents. Cette disposition avait été adoptée par l'association Vaincre la Mucoviscidose afin d'assurer l'équité de traitement entre les professionnels et les patients et parents participant aux journées de formation nationales (4 journées dans l'année). Les équipes des CRCM n'ont par ailleurs pas disposé de budget pour indemniser les frais de transports du patient ou parent lorsqu'il venait en réunion dans le CRCM.

D'autres items étaient présentés à titre de proposition pour, à l'avenir, renforcer la capacité de participation des patients et parents au PHARE-M : d'autres formes d'indemnisations complémentaires (garde d'enfant, heures de travail perdues), une information/formation sur fonctionnement général de l'hôpital ou une information plus générale sur la maladie et les traitements, au-delà des connaissances du patient ou

parent acquises pour sa propre prise en charge. Ces items n'ont pas rencontré de consensus dans aucun des deux groupes. En réponse à un item du questionnaire, les répondants ont indiqué que le programme avait créé de bonnes conditions pour la participation des patients et parents dans les équipes de pilotage.

VII-3. L'évaluation de leur contribution individuelle dans le fonctionnement de l'équipe de pilotage

La contribution des patients et parents a été jugée ***significative*** dans les réunions locales avec l'équipe de pilotage. Leur participation à ces réunions est jugée unanimement ***indispensable*** pour apporter une contribution dans le cadre du programme PHARE-M. Les idées et propositions du patient ou parent ont été ***généralement prises en compte par les équipes de pilotage***.

Les avis ont été partagés sur les items relatifs aux effets de la présence du patient ou parent dans les réunions d'équipe de pilotage, sur l'atmosphère de travail (ni meilleure et ni plus productive), le rythme de travail (pas plus lent). Certains parents ou patients ont mené des actions spécifiques bien identifiées au sein de leur CRCM – conception et réalisation d'un tableau de smileys pour que les enfants expriment leur satisfaction à la fin de la visite ; rédaction d'une gazette interne au CRCM pour annoncer les changements et informer les autres parents ; proposition d'un questionnaire de satisfaction pour les parents du CRCM et gestion informatisée des réponses ; conception et réalisation d'un carnet de liaison avec le médecin traitant en ville ; création d'une valisette pour les documents de l'enfant tels que les ordonnances, les imagiers d'éducation…). Toutefois, aucun consensus ne se dégage sur l'item « certaines décisions de l'équipe ont été inspirées par le patient ou parent ». Ceci nous semble à mettre en perspective des réponses concernant le mode de fonctionnement interne des équipes de pilotage, qui témoignent d'une cohésion et d'une solidarité entre les membres de l'équipe et rendent plus difficile l'identification de l'effet de la présence ou de l'absence d'un individu en particulier et la capacité à attribuer la paternité d'une décision à l'un de ses membres. (cf. §5).

OS2 : Evaluer l'effet de la démarche qualité auprès des professionnels et des patients/parents, à travers la maîtrise des outils et des méthodes de la qualité, le fonctionnement de l'équipe de pilotage et in fine la perception d'utilité d'une telle démarche *(compétences acquises)*

L'effet de la démarche qualité a été analysé au regard des catégories de Shortell (105) et Lémieux-Charles (104). Les items de leurs questionnaires respectifs ont été utilisés sans modification, après traduction en français. L'analyse de la validité interne (Alpha de Cronbach) réalisée sur la base de la totalité des réponses exprimées a montré la bonne cohérence interne et le maintien des dimensions de ces questionnaires après traduction et dans le contexte de l'enquête française (résultat non publié, publication à venir en 2018).

VII-4. Appropriation de la démarche qualité

L'effet du programme de formation-action en termes d'appropriation de la démarche qualité a été évaluée par des items caractérisant la ***maîtrise de la méthode et des outils de la qualité*** par les membres de équipes de pilotage.

Selon les deux groupes de répondants, PHARE-M a permis de dégager une vision claire des domaines sur lesquels faire porter les efforts d'amélioration, a fourni une

guidance pour organiser le travail de l'équipe de pilotage, et a permis à l'équipe de modifier ses façons de travailler et de s'assurer, grâce au suivi de données ciblées collectées dans le cadre du programme, que ces changements étaient des progrès. L'outil plébiscité par les équipes pour une utilisation courante était le diagramme des causes (arêtes de poisson).

Les deux groupes ne paraissaient pas avoir acquis, de leur point de vue, la maîtrise des « cycles PDSA » pour tester, analyser et ajuster les idées de changement. Cette technique a posé le plus de difficultés aux équipes de pilotage au cours du programme : le choix d'un périmètre de test (volontairement limité), la réalisation (DO) du test tel que prévu et décrit (PLAN) pour permettre d'en évaluer le plus fidèlement possible les conséquences (STUDY) avant ajustement éventuel de l'action (ACT), a paru être en tension avec la culture auto-adaptative de l'équipe et avec une tendance à la sur-réaction face à un événement jugé insatisfaisant par le soignant ou à un imprévu dans l'organisation. La planification et la mise en place de changements globaux (tous patients inclus dans l'objectif d'amélioration de l'indicateur de santé et tous soignants concernés par l'objectif), avec des réajustements successifs discutés lors des réunions d'équipe, a été la méthode le plus souvent suivie. Ceci semble faire écho aux constats de Tucker sur la difficulté culturelle de distinguer la boucle de réaction/adaptation (premier niveau) de la boucle d'analyse et résolution des problèmes (second niveau) (33). Il peut en résulter une difficulté à interpréter l'évolution des mesures de process suivies du fait de processus en adaptation continue et à fixer la version du processus à standardiser dans l'organisation.

Les autres items relatifs à la disponibilité de données en routine, pour suivre la qualité des processus de soin au-delà de la durée du programme, identifier l'émergence de nouveaux problèmes et maintenir les nouveaux processus dans la durée, n'ont pas recueilli de consensus dans aucun des deux groupes de répondants. Toutefois, le besoin de mettre en place un tel suivi de données en routine a été jugé nécessaire pour maintenir le travail sur la qualité des soins dans la durée. L'effectivité du soutien des autres départements de l'hôpital (le département qualité était cité dans la rédaction de l'item) pour soutenir l'amélioration continue de la qualité au-delà de la durée du programme n'a pas recueilli de consensus parmi les répondants.

VII-5. Fonctionnement interne de l'équipe de pilotage

Lémieux-Charles a montré que la participation à une démarche qualité améliorait le fonctionnement du travail en équipe. Nous avons considéré la proposition inverse, à savoir que le bon fonctionnement de l'équipe de pilotage, dans les 4 domaines identifiés par Lémieux, permettrait une meilleure efficacité de la démarche qualité. Les items analysés concernent donc : 1) l'implication du leadership dans l'organisation du travail de l'équipe de pilotage, 2) le processus de prise de décision partagé, 3) la régulation normative par les objectifs et les tâches allouées à chacun, 4) les collaborations internes ou externes traduisant les solidarités.

L'image du fonctionnement de l'ensemble des équipes, consensuelle pour les deux groupes de répondants, a rendu compte de : 1) un leadership témoignant de l'importance accordée au programme et au bon fonctionnement de l'équipe, leur affectant des ressources et des compétences adaptées aux besoins et partageant les informations pour la bonne organisation du travail de l'équipe ; 2) un processus de prise de décision partagée, avec une attention portée à l'expression des idées de

chacun en veillant à créer une ambiance propice à la proposition d'idées ; les professionnels ont en outre exprimé leur satisfaction d'avoir pu « *construire à partir de nos différences* » 3) une régulation normative par les objectifs partagés et l'implication de tous les professionnels pour le bon achèvement de ces objectifs ; 4) une bonne collaboration interne entre les membres de l'équipe, notamment pour suggérer des idées d'amélioration. Les items non consensuels dans les deux groupes portaient d'une part sur une régulation par le respect des tâches allouées aux différents membres, d'autre part sur la collaboration avec les autres départements de l'hôpital. Il existe une forte variabilité entre les centres sur les réponses à ces items.

En conclusion, les visions sont très consensuelles entre les deux groupes de répondants sur le fonctionnement interne de l'équipe de pilotage, dont il ressort une cohésion, une bonne écoute des idées de chacun et une prise de décision partagée en vue de la réalisation des objectifs communs.

VII-6. Utilité de la démarche qualité

La perception de l'utilité de la démarche qualité a été exprimée de façon consensuelle par les patients et parents et les professionnels de équipes de pilotage à travers les assertions suivantes : ***une satisfaction sur la participation*** en tant que membre de l'équipe de pilotage et le ***souhait de rester dans une telle équipe*** pour travailler à l'amélioration de la qualité des soins dans le CRCM ; ***l'utilité du travail*** réalisé par l'équipe pour améliorer la qualité des soins et le fait que ***ce travail a répondu aux besoins de l'organisation*** ; la nécessité de ***maintenir une démarche qualité continue*** pour continuellement améliorer la prise en charge.

OS3 : Appréhender l'évolution de la représentation de la place de l'usager par les professionnels et les patients/parents suite à l'expérience de participation des patients/parents au programme qualité PHARE-M *(place de l'usager)*

VII-7. L'évolution au fil du temps de la représentation de la place de l'usager dans cette démarche qualité

Entre 2011 et 2013, la démarche qualité collaborative incluant la participation de patients et parents était tout à fait innovante dans la filière mucoviscidose en France, bien qu'étant l'objet de communications par les équipes américaines ou canadiennes dans les congrès nord-américains depuis plusieurs années. Des différences notables de culture et de pratiques pluridisciplinaires et collaboratives avec les patients et parents, existaient entre les équipes engagées dans le programme : certaines équipes étaient bien formées à l'éducation thérapeutique, participant au groupe national de développement de l'ETP et ayant développé un programme local (pédiatrie), certaines équipes animant un collectif de parents d'enfants pour partager des idées d'amélioration des pratiques, d'autres équipes étant davantage dans une culture médico-centrée prescriptive associant ponctuellement des paramédicaux selon les besoins perçus par le médecin et la disponibilité des soignants lors des consultations.

Lors de la session 1 du PHARE-M, l'évaluation externe a identifié des tensions entre les médecins, les paramédicaux et les patients ou parents sur la participation de ces derniers à la démarche qualité. Si les parents manifestaient leur volonté d'apporter un témoignage de leur expérience des soins et de la vie avec la maladie – en veillant à ne pas paraître « donneurs de leçons » vis-à-vis des soignants, les patients

exprimaient une prudence vis-à-vis de l'équipe et des médecins pour s'exprimer en tant qu'usager, en étant néanmoins motivé par la curiosité pour l'organisation de leur CRCM et de l'équipe, ainsi que par la comparaison des indicateurs de santé des patients des différents centres. Le niveau de tension chez les patients et parents était plus fort sur des sujets ravivant une insatisfaction pour des soins reçus dans le passé, ou pour des annonces difficiles. Du côté des professionnels, des différences entre les équipes sont apparues, avec en point saillant partagé : le fait d'être confronté à un « vrai » patient (ou parent) et de ne plus s'autoriser à parler au nom « des patients », à leur place. De plus, la présence du patient ou parent a obligé à prendre en compte ses réactions et ses suggestions lors des échanges sur les objectifs d'amélioration et les idées de changements. Les soignants ont indiqué avoir dû modifier leur façon de s'exprimer et d'envisager les choses, dès les réunions nationales de formation puis lors des réunions locales en équipe, tout en invoquant parfois la question de la *représentativité* du point de vue du patient recruté dans l'équipe.

Après trois années de travail en commun au sein des équipes de pilotage, les réponses ont été unanimes sur l'item : « ***la présence d'un patient ou parent dans l'équipe de pilotage est une évidence et un atout*** ». Pour rappel, leur contribution a été jugée forte au cours des réunions locales de l'équipe, leurs idées ont été généralement prises en compte, et tous, professionnels et patients ou parents, souhaitent prolonger leur implication dans une telle équipe pluridisciplinaire pour l'amélioration de la qualité des soins. Le patient ou parent a développé une relation de confiance privilégiée avec l'équipe sur les sujets touchant à l'amélioration des soins et à l'organisation du CRCM et est sollicité plusieurs années après pour avis, partage des informations sur les résultats obtenus ainsi que pour sa participation à l'enquête. A noter : un parent a cessé son implication pour raison d'aggravation de la santé de son enfant – il a pu être remplacé sans délai ; un CRCM a souhaité revenir à son fonctionnement antérieur avec le collectif plutôt que de prolonger la participation du parent à l'équipe de pilotage.

OS4 : Appréhender le niveau de qualité des soins et de culture de l'organisation après trois années de démarche qualité continue, perçue par les professionnels et des patients/parents *(qualité des soins)*

VII-8. La vision de la qualité des soins & de la culture de l'organisation après trois années de démarche qualité

La caractérisation de la qualité des soins a utilisé le Chronic Care Model décliné à la prise en charge de la mucoviscidose, dans ses 6 dimensions, à l'aide de 44 items, ainsi que le questionnaire de Shortell concernant le leadership et la culture centrée sur le patient.

Les deux groupes de répondants ont souligné les caractéristiques suivantes après 3 années de programme PHARE-M : l'existence *d'objectifs de progrès* dans le CRCM et le *suivi d'indicateurs* s'y rapportant ; une *équipe pluridisciplinaire stable et experte* dans la prise en charge de la mucoviscidose ; une équipe bien formée à l'éducation thérapeutique et l'existence d'un programme ETP ; des *processus optimisés* de consultation et de réponse aux appels téléphoniques entre les visites et *l'existence d'un dossier électronique du patient.* Le leadership est perçu comme conduisant l'organisation à *satisfaire les besoins des patients et assurer la sécurité des soins*. La culture du CRCM est qualifiée comme prenant en compte *les besoins et les*

demandes des patients et analysant *les causes des réclamations* pour éviter que les problèmes ne se reproduisent.

Les professionnels indiquent en outre *revoir régulièrement en staff pluridisciplinaire* les situations des patients, disposer de *l'accès aux recommandations de soin* et *organiser des ressources de soin dans l'environnement du patient* – alors que les patients et parents n'ont pas de réponses consensuelles sur ces items. Le consensus des professionnels n'est plus acquis sur *l'utilisation du dossier électronique du patient lors des revues* de leurs situations au cours des staffs pluridisciplinaires, ni sur *l'accès aux mises à jour récentes des recommandations* de soin– les patients et parents affichant leur méconnaissance sur ces questions. Enfin, aucun consensus ne se dégage dans les deux groupes sur les items : les patients sont éduqués à hauteur de leurs besoins ; le dossier électronique contient les résultats de biologie et de radiologie ; des données provenant des patients sont utilisées pour améliorer les services.

Les visions des deux groupes convergent donc pour les aspects de la qualité de la prise en charge qui ont été ciblés au cours du programme, à savoir les processus organisationnels, notamment les processus en contact avec les patients, la culture centrée sur les besoins du patient, et la mise en valeur de l'ETP. Toutefois la mise en pratique de l'ETP, dans le cadre des venues en consultation, reste insuffisante par rapport aux besoins des patients avec des effets centre à rechercher. Les patients et parents ne sont pas informés des processus internes à l'équipe professionnelle tels que le fonctionnement des staffs pluridisciplinaires, la gestion du dossier électronique ou l'information sur les mises à jour des recommandations de soin.

En conclusion, les résultats de l'étude montrent la faisabilité de la participation des patients et parents dans le cadre structuré de la démarche qualité PHARE-M, l'appropriation de cette démarche par les professionnels et les patients/parents, son utilité perçue pour améliorer la qualité des soins et l'évolution de la représentation de la place de l'usager dans l'amélioration de l'organisation et des processus jusqu'à la considérer comme une évidence et un atout.

L'article IV (Volume 2) présente les résultats de l'apport du partenariat patient dans la démarche qualité PHARE-M. L'abstract (*traduction française)* est présenté ci-dessous. L'article complet est présenté à la suite.

Lessons from patient and parent involvement (P&PI) in a Quality Improvement Program in Cystic Fibrosis care in France

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Introduction

Les démarches qualité des soins en mucoviscidose ont émergé comme de nouvelles stratégies pour réduire la variabilité des soins et des indicateurs de santé des patients en diffusant les meilleures pratiques dans tous les centres. La Fondation américaine a développé un programme qualité collaboratif qui a été introduit en France en 2011. La participation de patients et de parents dans les équipes de pilotage de la qualité des centres est une dimension innovante de cette démarche.

Méthode

Un patient ou parent a été recruté par chaque CRCM pour participer à l'équipe de pilotage de la qualité. Tous les membres des équipes de pilotage ont été formés à la démarche et ont apporté leur expertise pour améliorer les processus de soins. La participation des patients et parents a été analysée dans le cadre de l'évaluation de l'efficacité de l'intervention PHARE-M. Les observations et les interviews menées au cours de la première année de formation PHARE-M ont pris acte des motivations des patients et des parents et de la vision des professionnels sur leur participation. Dans le cadre de la recherche, un questionnaire d'enquête a été développé pour analyser les différentes composantes de contexte et les mécanismes de l'intervention PHARE-M, et recueillir les opinions des professionnels et des patients et parents, après 3 années sur les changements intervenus dans les soins, les effets sur l'équipe et la représentation de la participation des usagers à cette démarche qualité. Les réponses sur l'ensemble des items ont été analysées en vue d'identifier les consensus et dissensus entre les 2 groupes de répondants.

Résultats

L'évaluation de la session1 du PHARE-M a mis en évidence des tensions liées à la participation des patients et parents, entre leurs positions respectives de patients et d'usagers du système de soins. Les patients étaient motivés par la curiosité vis-à-vis du fonctionnement des équipes, la vision des diverses organisations ainsi que les résultats des CRCM. Soixante-seize personnes, dont 12 patients ou parents des 14 CRCM pilotes, ont répondu au questionnaire d'enquête après 3 ans. Un consensus s'est dégagé sur les items caractérisant la performance du programme, l'efficacité des équipes de pilotage et leur fonctionnement interne, ainsi que sur les caractéristiques de la prise en charge, notamment l'optimisation des processus, la multidisciplinarité et la délivrance de soins centrés sur les besoins du patient. Concernant l'utilisation des dossiers électroniques des patients, l'application des recommandations ou l'organisation de ressources dans la communauté du patient, les réponses n'étaient pas consensuelles et sources de dissensus entre les deux groupes. Les conditions créées par le PHARE-M pour la participation des patients et parents ont été jugées bonnes.

Discussion

Des facteurs de réussite de la participation du patient/parent à la démarche qualité ont été identifiés. Des réponses ont été apportées aux questions de recherche concernant la faisabilité, l'efficacité et l'utilité de la participation des patients et parents au PHARE-M. De nouvelles questions ont été posées sur la soutenabilité de la démarche d'amélioration continue de la qualité. Des pistes de réflexion ont été tracées : 1) un cadre de formation pour les professionnels impliqués dans des pratiques collaboratives en équipe pluridisciplinaire et avec des patients et parents, afin de développer leurs compétences d'animation et de gestion d'équipe, de résolution des conflits et de motivation au changement ; 2) le développement d'enquêtes sur l'expérience patient dans le système de soins pour obtenir une vision plus représentative des processus pour la démarche qualité.

Mots-clés : *engagement des patients, mucoviscidose, maladie rare*

Lessons from patient and parent involvement (P&PI) in a Quality Improvement Program in Cystic Fibrosis care in France

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Abstract

Introduction

Quality Improvement Programs (QIP) in cystic fibrosis (CF) care have emerged as strategies to reduce variability of care and of patient outcomes among centres facilitating the implementation of Best Practices in all centres. The US CF Foundation developed a Learning and Leadership Collaborative program which was transposed in France in 2011. Patient and parent involvement (P&PI) on the local quality teams (QTs) is one dimension of this complex intervention. The conditions and effects of this involvement needed to be evaluated.

Method

In all settings, patients and parents were recruited by their centre care team. They were trained to QI method and tools and contributed their own expertise to improve the process of care. This involvement has been analyzed in the frame of the whole process evaluation. Observations and interviews conducted during the course of the first PHARE-M*** training year explored the motivations of the patients and parents to participate and the vision of the health care teams. A research study was carried out after three years with the patients/parents and the professionals to assess the PHARE-M's effectiveness using a questionnaire to report their opinions on various components of the program, including their experience of P&PI. Responses were analyzed in view of identifying consensus and dissensus between the two groups.

Results

At the introduction of the program, P&PI was an opportunity for healthcare providers to reflect on their conceptions of these individuals both as patients and as healthcare system users. Curiosity about the teams' functioning, the various center organizations and outcomes led patients to overcome their initial barriers to participation. Seventy-six people including 12 patients/parents from the 14 pilot centres responded to the questionnaire after 3 years. Consensus between professionals and patients/parents was high on most items characterizing the performance of the QIP, QT effectiveness and QT functioning. Patients, parents and professionals agreed on the main characteristics of care such as an optimized organization, multidisciplinary care and patient-centredness. Regarding the use of patient electronic records, the use of care guidelines or the organization of support in the patient community, responses were not consensual amongst

*** Programme Hospitalier d'Amélioration des Résultats et de l'Expertise en Mucoviscidose – A hospital-based program for improvement of results and expertise in cystic fibrosis care

patients/parents and a source of dissensus between the two groups. All agreed that the PHARE-M organization created good conditions for their involvement. In the end, both groups agreed that it was difficult to attribute the paternity of some changes specifically to any member in the team.

Discussion

Success factors for patient/parent long-term involvement in QIP have been identified. Answers to questions raised by the stakeholders about the feasibility, efficiency and usefulness of P&PI in PHARE-M could be given but new questions arose about the sustainability of continuous quality improvement over time. Perspectives such as an educational framework to develop the skills and behaviors of professionals engaged in collaborative practice with patients and families and large patient experience surveys could be used to capture patients' experience of care in the improvement work.

Key words:
Quality improvement, patient involvement, cystic fibrosis.

Introduction/Background

Patient involvement in quality of care improvement is discussed in various ways depending on the perspective and the point of care delivery.

Regarding self-management of care, strategies have been developed and evaluated to inform, educate, and involve patients in their direct care [134]. A new model of care for persons with chronic diseases has been conceptualized that focuses on their experience and knowledge, and endeavors to shift from paternalism to a care partnership [135 ;136]. Formalized processes such as shared decision making have been developed to support patient engagement in their own options for care [137;138]. In several countries, the movement to empower chronically ill patients has given rise to specific trainings to involve them in mentoring or in peer-to-peer programs in order to support other patients with the disease [139]. Experience with patients as teachers at schools of medicine or interprofessional healthcare programs is ongoing [140;141;142].

Quality of care in hospital settings was defined by the US Institute of Medicine in 2001 as clinical effectiveness, safety and patient centredness [143]. Clinical effectiveness is generally viewed as too technical to accommodate patient contributions and the usefulness of patient surveys in assessing medical quality of care remain debatable [144]. However, it is widely accepted that patients may make significant contributions to non-clinical aspects of care [145]. Many opportunities have been identified for patients to contribute to the safety of the care they receive at the hospital [146]. Moreover, reporting of safety information on medical errors and adverse events through patient interviews or surveys may also aid in identifying failures in every stage of the care process, from diagnosis to medication or clinical services [147]. Therefore, patients are recognized as being capable of contributing substantially to safety in the care by identifying care factors that potentially lead to harm or helping to learn from an incident to avoid it in the future [148]. Beyond matters of safety, the involvement of patients or their representatives in the organization of hospital care is usually associated with activities related to planning services, designing processes or assessing quality management. Groene and Sunol

proposed a conceptual framework for patient involvement in quality management comprising 5 stages: criteria development, process design, quality committees, improvement projects and discussion of quality improvement project results [149]. Their literature review and a cross-sectional survey at hospitals in the DUQuE project [150] reported experiences of patients involvement across these stages [151]: 1) on guideline development to address the needs of chronically ill patients as well as aspects of continuity of care and integration of service; 2) in assessing care preferences and designing process through surveys, focus groups and observations ; 3) in regular formal meetings to ensure quality and safety ; 4) in establishing a partnership with the QI team to plan and deliver a QI intervention in a series of plan-do-study-act (PDSA) cycles ; 5) more rarely in discussing quality improvement project results.

The history of cystic fibrosis (CF) care has been one of continuous improvement, led by the worldwide combined efforts of patient organizations, researchers and clinical teams. Therapeutic advances associated with the implementation of CF specialized care centres have brought about a dramatic increase in life expectancy and quality of life for people with CF. In the new century, Quality Improvement Programs (QIP) have emerged as new strategies to reduce variability in care as well as in patient outcomes across centres facilitating implementation of Best Practices in all centres. In this rare disease, QI is driven by comparisons of patient outcomes between national patient registries at national and centre levels [152]. Since the 2000s, the US CF Foundation and the Dartmouth Institute have developed a CF Learning and Leadership Collaborative (LLC) program to accelerate the improvement of CF care across the US centres [153].

France is a country of major prevalence of this genetic disease with 6,585 patients recorded in the national Registry in 2013, 53.7% of whom were adults. Since newborn screening became generalized in France in 2002, the French CF care network has been organized into specialized CF centres (CFCs). In the frame of the second French National Plan for Rare Diseases two centres of expertise were designated in order to develop French national action plans. The US CF QIP was

transposed to France by the Nantes-Roscoff centre of expertise, and the PHARE-M[10] program was launched in September 2011 through a pilot phase involving 14 centres volunteer to test and adapt the method to the French CF care organization (Table I) [154]. This QI approach is innovative in France as it installs a quality improvement dynamics and culture among the health care teams focusing on disease specific care practices and patient health outcomes improvement [155] when most QI interventions are framed by the French National Health Authority certification process. PHARE-M intends to involve patients and parents on a long-term collaboration with their care teams (nearly 3 years) to take into account their experience and preferences along the successive PDSA cycles for the redesign of the care process at their centre. The attempt to establish this long-term partnership to improve the care process is part of the innovation of this QI approach in France which needed to be evaluated. Some aspects were particularly questioned from the point of view of the patients/parents and the professionals: how did they perceive the conditions in place to allow the participation of patients and parents in the program? How did the quality team's professionals perceive this participation and what were the feelings of the participating patients and parents? Is the quality of care appreciated in the same way by patients and professionals after three years of joint work? How effective were the quality teams perceived in organizing the QI work and mastering the QI method and tools to which they had been trained? How effective was the participation of all members in the discussions and in decision-making? In the end, was the contribution of patients / parents perceptible in the quality improvement work and on the results on the process of care?

The objective of this article is to report and reflect on patient and parent involvement at the 14 centres engaged in the pilot phase of the PHARE-M program from the perspective of the patients and parents and from the perspective of the professionals on the quality teams. By illustrating Groene's conceptual framework regarding *the partnership between patients and the QI team to plan and deliver a QI intervention in a series of plan-do-study-act (PDSA) cycles*, we intend to contribute to the field with

[10] Programme Hospitalier d'Amélioration des Résultats et de l'Expertise en Mucoviscidose – A hospital-based program for improvement of results and expertise in cystic fibrosis care

our experience of patient/parent involvement in a learning and leadership quality improvement program within a rare disease network in France.

Method

We present successively the conditions set up for patient and parent involvement in the PHARE-M program then how this involvement has been analyzed, first in the evaluation of the transposition process of the US QIP to France, then in the assessment of the program's effectiveness after three years [156].

Setting: Patient and Parent involvement in the PHARE-M

The PHARE-M was developed and adapted to the French setting by the senior pediatrician director of the centre of expertise, and a parent of an adolescent with CF, an engineer by training. Both attended the quality course in The Dartmouth Institute Microsystem Academy. The parent became the teacher and coach in the QI program.

The PHARE-M is a one year training program that follows a step by step curriculum known as the Dartmouth Microsystem Improvement Ramp [157]. This curriculum consists of multiple steps described in this OJRD supplement [154] including the declaration of a theme for improvement, the identification of leverage factors and the establishment of PDSA cycles to implement changes in the care process. As many changes require two to three years to be fully implemented, post PHARE-M sessions have been organized at the request of the teams, consisting in an on-site benchmarking visit each year, allowing to review methodological points, follow up the CFCs' actions, analyze the results achieved, and prepare publications of QI experiences.

The quality team (QT) formed at each CFC involves 4 to 5 professionals from the multidisciplinary team and is led by a physician. The recruitment of a parent (pediatric program) or a patient (adult program) in the quality team is a prerequisite to engage in PHARE-M. It has been operated by the physician leader following a recruitment procedure including a list of criteria on an application form. The consent form specifies that neither their participation nor their withdrawal would have any impact on their own care or their child's care and that their participation in the QT can cease at any time they wish. One « correspondent » professional is in charge for liaising

with the patient or parent to regularly share information, answer their questions and solve practical issues. When recruited, patients and parents are enlisted in the PHARE-M training sessions as QT members. They exercise the method with their team during the face-to-face-meetings. Patient outcomes as well as key process indicators are transparently shared with them, those regarding their centre as well as those regarding the other centres involved in the training session. Patients or parents are also invited to participate in the PHARE-M web conferences every 4 to 6 weeks. Their travel fees are reimbursed by the national patient organization. They are invited at the local QT meetings which are generally hold every 2 to 3 weeks. If they can't attend these meetings, they are updated on the work done by their correspondent on the QT. All personal health information from patients included in redesigned care processes are anonymized before being discussed at any QT meetings attended by the patient or parent. Ethical rules are established in relation to the information shared at the meetings. When a patient or parent group is active at the centre, rules are defined for communication with the group.

P&PI analysis as part of the transposition process evaluation

An evaluation was requested by the leader of the Centre of Expertise as part of the transposition process of the US CF QI program to France [154]. It was conducted by a sociologist from Mines Nantes School on the PHARE-M pilot session in order to investigate requirements for a successful national roll-out of the PHARE-M, identify the possible technical or cultural barriers and propose possible adjustments to the program to adapt it to the French context.

The assessor participated as an observer in two web meetings and one Face-to-Face meeting. The assessment included becoming familiar with PHARE-M documents, interviews with a panel of professionals and the patients/parents on the QTs, the members of the national PHARE-M team, the American supervisor from the Dartmouth Institute, and visiting one CFC site. All interviews and focus groups were recorded and fully transcribed. The data was managed (coding, categorization), processed (analysis, validity) and interpreted according to the standard thematic content analysis protocol (Miles & Huberman, 2003 [[158]]). This was followed by manual grouping and counting within a framework for analysis with the following dimensions: process applicability (terminology, formalization, tools, remote coordination); patients and parents involvement (roles, time spent, obstacles); French

national and regional coordination (roles, nature of support, mechanisms for incorporation); process adoption (perceived benefits and costs, working atmosphere, engagement, acquisitions); and effects (operation, working practices, cooperation with partners). Results on the dimension regarding patient and parent involvement during the pilot phase PHARE-M training year are reported in this article.

P&PI analysis as part of PHARE-M effectiveness assessment after 3 years

Since the introduction of PHARE-M in France in 2011, questions were raised by the stakeholders about the effectiveness of this quality improvement program. The first evaluation concluded that effectiveness could not be assessed at the end of the first year, neither on patient outcomes nor on results of changes in the care process, but should be assessed after three years on the basis of the program's measurable effects.

A research project was drawn up by the Centre of Expertise of Nantes-Roscoff to analyze the performance of the PHARE-M program after three years (2015) at the 14 CF centres involved in the pilot phase of the program. This research project was funded by the French ministry of Health (Decision of the Call for project PRePS – Dec 2012). The aims and protocol of the broader project from which the results are drawn are described in the OJRD supplement [159]. In brief, the protocol combines a quasi-experimental evaluation of the effectiveness of the program on patient outcomes evolution over three years with a process evaluation [160]. Following a realistic approach, the latter was designed to understand what works, for whom and under which circumstances (context) [161]. To understand which dimensions of the context were critical for the effectiveness of the programme, a questionnaire was designed assembling existing validated tools when they existed and developing new tools when necessary.

- Development of the questionnaire

The questionnaire was prepared by a panel of experts (professionals and parents/patients), tested with 3 multidisciplinary teams (N=29 respondents including 1 parent and 2 patients) and reviewed by experts in Sorbonne Paris Cité University. The final questionnaire was composed of 7 chapters covering the various aspects of the organization of care and the PHARE-M effectiveness at the centres: quality of the

care process, organizational culture, patient centredness, leadership, mastering of the QI process and tools, quality team functioning and patient/parent involvement.

- Studied population

Every professional in the 14 centres, including professionals belonging to the quality teams and the patients and parents involved.

- Variables

The items in five chapters were based on existing instruments validated in previous research [162; 163]. The items characterizing the chapter about quality of care were developed for this research following the 5 dimensions of the Chronic Care Model [164]: existing goals for improvement; multidisciplinary care; self-management support; support in decision making (guidelines); electronic patient records and resources in the patient community. The items of the questionnaire analyzing patient and parent involvement were developed according to the framework proposed by Carman [165] and adapted by Pomey [135] : 1) patient and parent information/activation 2) patient and parent empowerment and 3) patient and parent contribution to the QI work.

- Data collection

The questionnaire was self-administrated during 14 on site investigations conducted by a clinical research assistant. The respondents had no opportunity to discuss their answers amongst themselves. Each topic is covered by a list of assertions requiring a response on a 5 degrees Lickert scale from « completely agree », to « fully disagree » with a neutral response « don’t know/no opinion ».

- Data analysis

The responses were managed using SAS and XL and were analyzed, according to the purpose, grouping different categories of respondents: professionals in the quality teams, patients and parents. During restitutions to the centre teams, reports by centres were produced to share the results and discuss new improvement goals for the care process.

To answer the questions from the point of view of the patients/parents and the professionals, the analysis of the responses on all items of the questionnaire was made for the two groups of respondents: the patient/parent group (N=12) and the

professional group in the quality teams pooled for all teams and all disciplines (N=64). We first identified the items that achieved a « strong consensus » in the patient/parent group considering unanimous or nearly unanimous responses (unanimity less one vote or unanimity less two votes; >80%) as either positive (grouping « agree » and « completely agree »), negative (grouping « disagree » and « fully disagree ») or neutral (« don't know » or « no opinion »). We then identified the items that achieved a strong consensus in the professional group (> 80% responses with either positive, negative or neutral answers). We define dissensus or consensus between the patient/parent group and the professional group using Fisher's exact test [166] (Results available on request).

The results highlighted the following categories: 1) items achieving a consensual position between the two groups of respondents (consensual positions were found always in the same sense in the 2 groups, positive (+), negative (-) or neutral (N)); 2) items achieving consensual position in the patient group only; 3) items achieving consensus in the professional group only; and 4) items achieving no consensus (NC) in either of the two groups.

Presentation of consensus/dissensus between the Patient/Parent and the Professional groups

Items category	Consensus amongst P&P	No consensus (NC) amongst P&P
Consensus amongst Professionals	1) (+,+) or (-,-) or (N,N)	3) (NC,+) or (NC,-) or (NC,N)
No consensus (NC) amongst Professionals	2) (+,NC) or (-,NC) or (N,NC)	4) (NC,NC)

Due to the small sample of patients and parents (N=12) and their affiliation to 12 different centres, variations in their responses regarding local culture, organization, leadership and the performance of the QIP achieving no consensus are mainly to be attributed to "centre effects". We did not set out to compare the responses of the patient/parent to the responses of the professionals by center.

Regulatory authorizations were granted from the Ethics Committee of the Brest University Hospital and by CNIL (DR2015040).

Results:

Results from the observations and interviews conducted as part of the QIP transposition process to France

The opinions and concerns regarding the participation of parents and patients involved in the QTs during the program training year are summarized in Table II. The following themes emerged:

- The place of the patient/parent in the healthcare system

Patient and parent involvement disrupted assigned places, led to readjustments and reinterpretations, and highlighted resilient patient and parent profiles.

- Reasons and barriers expressed by parents for participating

They stressed contributing their testimonial on their experience and sticking to merely conveying their feelings and day-to-day experiences. They were careful not to appear to teach professionals their profession.

- Reasons and barriers expressed by patients for participating:
    - **Wariness/caution** towards the care team and the medical world.
    - **Consent and curiosity** to get to know a CF setting, to better get to know the teams that they visited as their care providers.
    - **Engagement under tension** between on one hand, the desire to understand, be curious, gain autonomy and confidence, remove obstacles, and, on the other hand, the difficulty of pushing oneself to talk in front of others about one's experiences with the care of a disease that one would like to keep at a distance.
- Healthcare providers' vision of patients/parents involved in the quality teams:

Their vision of patients/parents was confronted with real patients and parents. The presence of a patient on the team called into question healthcare providers' preconceived notions and desire. Some healthcare providers recognized that they granted themselves the authority to have a particular vision of patients and parents and to talk about them, about what they believe to be patients' experience and feelings, given their in-depth knowledge of the « ill human being ». The presence and intervention of a real patient or parent in the quality team challenged their representation and some raised the question of the representativeness/validity of the speech of the patient or parent involved.

The patient or parent participation on the QTs and their presence at the PHARE-M Face-to-Face training sessions as well as at many local meetings was perceived as an opportunity for the healthcare providers to reflect on their conceptions of the patients/parents as both patients and healthcare system users. Curiosity about the teams' functioning and comparison between the various center organizations and their outcomes led patients to overcome their initial barriers and grant their consent to participate.

Results from the *PHARE-M effectiveness assessment after 3 years*

Volunteer patients and parents were recruited by all care teams after information given on the QI program and on the importance of their involvement to improve care at their centre [167]. Over the 3 years, three of them stopped their participation. One parent wanted to stop because of health worsening of her child and was replaced by another parent who happened to be a quality engineer in pharmaceuticals. One CFC stopped the program when the physician leader retired. The 3rd CFC chose to work with the parent group (as historically) and collect feedback on change actions at annual patient group meeting.

During on site investigations 140 people from the 14 CFCs completed the questionnaire, either as QT participants or as multidisciplinary team members outside the QTs. The QT respondents totaled **76 people** (54% of all respondents): **12 patients and parents** (6 patients and 6 parents) and **64 professionals**, including 56 healthcare providers and 8 non-healthcare providers (quality engineers and others). Two CFCs were unable to contact the patient or parent to ask them to complete the questionnaire. Forty-six (82%) professionals in the QTs belonged to the CF multidisciplinary "core" team (physician, nurse, physiotherapist). Psychologists and dieticians were heavily engaged in the QTs (9 people).

Quality of care at the centre

Table III presents the items that achieved consensus or dissensus among the patients/parents and the professional groups on items related to Quality of care and organizational features at the centres after three years of joint QI work.

All the items that achieved a strong positive consensus among the patients and parents also achieved a strong positive consensus among the professionals on the QTs. They were related to the following domains of the chronic care model: 1)

GOALS: the existence of improvement goals at the CFC and indicators to monitor them, 2) SELF-MANAGEMENT SUPPORT : the existence of a therapeutic education program and professionals trained to deliver it 3) MULTIDISCIPLINARY CARE: an adequate multidisciplinary team, stable over time and possessing expertise in CF care, as well as KEY PROCESSES OF CARE: an optimized clinic visit process allowing the patient to see all members of the core team and any referral professionals from various disciplines when necessary as well as an optimized process of answering telephone or email messages from patients and families 4) INFORMATION SYSTEM: the existence of an electronic patient record (EPR) system at the centre.

Items detailing patient therapeutic education in practice, as well as items regarding certain information contained in the patient record achieved no consensus neither in the patient/parent group nor in the professional group.

The patients and parents granted unanimous neutral response (“Don’t know”) to items regarding the use of the EPR by the team during the staff meetings and the existence of a procedure to inform professionals on updates to guidelines when the professionals showed no consensus on these items.

Three items achieved a strong positive consensus among the professionals only. They were related to the following domains of the chronic care model: 3) MULTIDISCIPLINARY CARE: the systematic review of the records of the patients who came to the CFC; 5) DECISION SUPPORT: the availability of care guidelines to all professionals and 6) COMMUNITY NETWORK: the organization of a network of professionals in the patient community for managing care at home.

Organizational features at the centre

Unanimity was achieved for items related to PATIENT CENTREDNESS, taking patient needs and requests into account and analyzing causes of complaints to prevent problems from recurring. However, no consensus was achieved with respect to using data from the patients themselves to improve services. The same results were observed for the responses of the professionals with a rate of agreement of more than 90% on the first items, and a lower rate of agreement (< 70%) on using data from the patients themselves.

A consensus was achieved both in the patient/parent and in the professional group in perceiving LEADERSHIP as driving the organization to meet patient needs and ensure safety of care. Other aspects of leadership related to the multidisciplinary team management were mostly answered by patients/parents with “Don’t know”. The responses of the professionals by centres, displayed along the 5 axes of “radar” graphics, also show different types of leadership across the centres.

Table III: Consensus and dissensus between the P&P and the Professional groups on Quality of care and Organizational features at the centres

Categories: Quality of care, Patient centredness, Leadership	Consensus amongst P&P	No consensus amongst P&P
Consensus amongst Professionals	**Quality of Care:** **(++)** Existence of improvement goals at the CFC and indicators to monitor them **(++)** Existence of a therapeutic education program and professionals trained to deliver it **(++)** Adequate multidisciplinary team, stable over time and possessing expertise in CF care **(++)** Optimized clinic visit process allowing the patient to see all members of the core team and any referral professionals from various disciplines when necessary **(++)** Optimized process of answering phone or email messages from patients and families **(++)** Existence of an electronic patient record system at the centre **Patient Centredness:** (**++)** Taking patient needs and requests into account **(++)** Analyzing causes of complaints to prevent problems from recurring **Leadership:** **(++)** Driving the organization to meet patient needs and ensure safety of care	**Quality of Care:** **(NC,+)** Periodic review of the records of the patients who came to the CFC, during the multidisciplinary staff meetings **(NC,+)** Availability of care guidelines to all professionals **(NC,+)** Organization of care providers in the patient community
No consensus amongst Professionals	**Quality of Care** **(N,NC)** Use of the EPR by the team during the staff meetings **(N,NC)** Existence of a procedure to inform professionals on updates to guidelines	**Quality of Care:** **(NC,NC)** Patient therapeutic education meeting patients’ needs **(NC,NC)** Biology or Imaging Information contained in the EPR **Patient centredness:** Using data from the patients themselves to improve services

PHARE-M performance and QT effectiveness

Table IV presents the items that achieved consensus or dissensus among the patients/parents and the professional groups on items related to the program's performance and the QTs' effectiveness.

The perceived performance of the PHARE-M was expressed with items focusing on the experience of the respondents as members of the QTs. A strong positive consensus was achieved amongst both patients/parents and professionals regarding their satisfaction as a member of the QT and their wish to remain on a similar team working on QI. Moreover, their perception of the usefulness of the work of the team in improving care and meeting the organization's needs was unanimously positive. All stated that an ongoing quality improvement process had to be maintained to continuously improve care at the centre.

The performance of PHARE-M as a "training-action" program on this QI approach was appreciated by the respondents with items characterizing their mastery of the quality methods and tools. There was a strong positive consensus in the two groups that the PHARE-M led to a clear vision of the area on which to focus the efforts for improvement at the centre, provided a guide for organizing QI work, and enabled the team to change its way of working and analyze data to ensure that these changes represented an improvement. Both groups agreed that a specific data collection had to be established for the QI work. The others topics related to the availability of data at their centre, by the end of the program, to allow to analyze and identify problems as well as to follow the implementation of changes achieved no consensus neither in the patient/parent group nor in the professional group.

Regarding the techniques to lead changes, no consensus was achieved in both groups on PDSA cycles monitoring to implement changes through tests and evaluations before extension. The support for changes implementation from the other departments in hospital achieved no consensus among the two groups.

376 *Table IV: Consensus and dissensus between the P&P and the Professional groups*
377 *on PHARE-M perceived performance and QT effectiveness*

Categories: PHARE-M performance QT effectiveness	Consensus amongst P&P	No consensus amongst P&P
Consensus amongst Professionals	**Experience on the QT:** **(++)** Satisfied with my experience as a member of the QT **(++)** Wish to remain on a similar team working on QI **QI work done by the QT:** **(++)** Usefulness of the work done by the quality team in improving care **(++)** QI work meets the organization's needs **(++)** An ongoing quality improvement process has to be maintained to continuously improve care at the centre **Mastery of PHARE-M method and tools:** **(++)** A clear vision of the area to focus the improvement efforts on **(++)** A guide for organizing the QI work **(++)** Ability to implement changes **(++)** Ability to analyze data to ensure changes were improvements **(++)** Need to set up a specific data collection for QI work	
No consensus amongst Professionals		**Mastery of PHARE-M method and tools:** **(NC,NC)** Ability of the QT to analyze variations in processes over a period of time **(NC,NC)** Availability in routine of data to analyze and identify problems **(NC,NC)** Availability of routine data collection to follow the implementation of the new processes of care **Change Management (PDSA cycles):** **(NC,NC)** Ability to conduct tests of changes with PDSA cycles and learn from the results **(NC,NC)** Support from the other hospital departments to conduct changes

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QT functioning

Table V presents the items that achieved consensus or dissensus among the patients/parents and the professional groups on items related to the QT's functioning. Those items address successively QTs process strategies, decision-making in the QTs, normative management, and internal or external collaborations [163].

A strong positive consensus was achieved on the items describing **QT process strategies**: the leader's behavior reflecting the importance he/she placed on the quality team functioning well, the team receiving all information required to plan and organize its work and, the availability of enough resources and skills on the team to work properly. The process of **shared decision making** on the team was rated as highly positive with attention being paid to the contributions of each member of the team, most team members participating in decision-making, and ease for all members in suggesting ideas for change. The **normative regulation** on the QTs was rated high regarding the agreement on and achievement of the objectives of the QI project. Though consensus was achieved on the professionals group on all members focusing on achieving the same goals, there was no consensus among the patient/parent group on this item. Last, internal **collaborations** in the QTs were rated high in the two groups but no consensus was achieved on external cooperations with the other departments of the hospital.

Table V: Consensus and dissensus between the P&P and the Professional groups on QT functioning

Categories: QT functioning	Consensus amongst P&P	No consensus amongst P&P
Consensus amongst Professionals	**Process strategies:** (**++**) Leader's behavior reflecting the importance he/she placed on the quality team functioning well **(++)** Members of the team came from different backgrounds, experiences and skills **(++)** Availability of enough resources and skills on the team to work properly **(++)** Team receiving all information required to plan and organize its work **Decision Making**: **(++)** Attention being paid to the contributions of each member of the team **(++)** Most team members participating in decision-making	**Process strategies:** **(NC+)** The leader also asked the opinions of the other members of the team **Decision Making**: **(NC+)** We appreciated and built with our differences **Normative:** **(NC+)** The team members were all focused on achieving the same goals.

	(++) Ease for all members in suggesting ideas for change **Normative:** **(++)** Team members agreed on the project's objectives **(++)** The achievement of the objectives guided the activities of the members of the team. **Internal/external collaborations:** **(++)** The people I've worked with are comfortable suggesting changes and improvements	
No consensus amongst Professionals		**Normative:** **(NC,NC)** The team members did what was expected of them. **Internal/external collaborations:** **(NC,NC)** There was a lot of cooperation between the departments of the hospital.

Patients and Parents involvement in the PHARE-M

Table VI presents the items that achieved consensus or dissensus among the patients/parents and the professional groups on items related to Patient and Parent Involvement in the PHARE-M.

The first series of items concerned the selection and activation of the patient/parent recruited. There was a consensus that the presence of a patient or parent on the quality team was “a given and an asset” despite a possible lack of education or their personal problems. A strong consensus was found to recruit a patient or parent well informed regarding the QI program goals and the need for a good relationship between the team and the patient/parent involved. The development of coping skills (*knowing how to manage emotions and stress; solving problems, making decisions, and making choices; knowing how to communicate and being at ease in relationships with others; and knowing how to put oneself in the place of others)* was by consensus a requirement for the patients and parents to be recruited to the QT. These items also achieved a strong consensus among the professionals, who had a higher rate of agreement on the “required qualities” for the patient or parent to join the team. Those qualities were not explicitly stated in the questionnaire.

Three items achieved a consensus among the patients and parents regarding their empowerment for participation: the reimbursement of their travel fees, their high motivation to improve care for all – achieving a weaker consensus to improve care for themselves, and the fact that their role on the QT was conveyed to the other patients or parents followed up at the centre. Only 8 out of 12 patients/parents

agreed on the need to be knowledgeable about the disease and its management beyond the requirements of their own care – while professionals had no consensus on that need. The professionals had a higher rate of agreement on the importance of the patients and parents taking a step back and drawing general lessons from their own experience. No consensus was achieved in both groups on the need for the patient or parent involved to understand the general functioning of the hospital. Finally, the patients and parents unanimously indicated that the organization of the PHARE-M throughout France promoted their membership on QTs.

Regarding their contribution to the QI work, the two groups agreed that patients and parents could make significant contribution to the work of the quality team and that their ideas and proposals were generally taken into account. Both groups agreed that patients and parents had to participate in the local QT meetings – rather than in the national meetings, to make these contributions. No consensus was achieved in both groups on the assertion that certain decisions made by the quality teams were inspired by the patient/parent.

Table VI: Consensus and dissensus between the P&P and the Professional groups on Patient and Parent Involvement

Categories: P&PI	Consensus amongst P&P	No consensus amongst P&P
Consensus amongst Professionals	**Activation/Recruitment:** (**++**) The presence of a patient or parent on the quality team is "a given and an asset" **(++)** Importance of the information provided to the patient or parent regarding the QI program goals **(++)** Need for a good relationship between the care team and the patient/parent involved **Empowerment:** **(++)** P&P role on the QT has to be conveyed to the other patients or parents followed up at the centre **(++)** The patient or parent is motivated to improve care for all **(++)** The organization of the PHARE-M throughout France created good conditions for their membership on QTs **Contribution:** **(++)** The patient or parent participates in and contributes significantly to the work of the QT. **(++)** Their ideas and proposals were generally taken into account **(++)** The patient or parent's regular participation at team meetings at the	**Activation/Recruitment:** **(NC,+)** The patients and parents are informed regularly (annually or more often) by the team about general subjects concerning cystic fibrosis care and research. **(NC,+)** P&P must have "required qualities" to join the team **Empowerment:** **(NC,+)** P&P have taken a step back and drawn general lessons from their own experience (**NC,+)** The patient or parent is also motivated to improve his or her own management by participating in the program.

	CFC is indispensable.	
No consensus amongst Professionals	**Activation/Recruitment:** **(+NC)** Patients/parents should have developed copying skills (with the disease) **Empowerment:** **(+NC)** Reimbursement of P&P travel fees	**Activation/Recruitment:** **(NC,NC)** The patients and parents are rather familiar with general cystic fibrosis information: research, progress made, and Registry data **Empowerment:** **(NC,NC)** The participation of a patient or parent should be facilitated by the reimbursement of other expenses: child-care, lost working hours, etc. **(NC,NC)** P&P need to be knowledgeable about the disease and its management beyond the requirements of their own care **(NC,NC)** The participating patient or parent does not represent all patients **(NC,NC)** It would be necessary to include several patients or parents to ensure that more different points of view are represented **(NC,NC)** P&P need to understand the general functioning of the hospital **Contribution:** **(NC,NC)** The participation of a patient or parent on the team at French national training and information meetings is indispensable. **(NC,NC)** The patient or parent participated and contributed as much as the professionals during the French national meetings **(NC,NC)** The atmosphere of work at the QT meetings is better and more productive when the P&P is present. **(NC,NC)** The pace of work is slower when the patient or parent is present at the QT meetings. **(NC,NC)** Certain decisions made by the QT are inspired by the patient/parent.

Discussion

Following the results of the investigations conducted with the care providers and patients/parents, we review the highlights on the instrumentality of the method to involve patients and parents in PHARE-M QIP. We then discuss the initial questions raised about this partnership during the PHARE-M program in France and propose a list of success factors which seem essential to long term patient/parent involvement in QI work in Table VII.

Highlights on the method to involve patients and parents in PHARE-M

PHARE-M quality improvement program was innovative in France in 2012 as it intends to install a culture of quality improvement in the CF care teams, focusing on patient outcomes improvement and process of care redesign. Patients and parents were involved on a long time period with the care teams at their centre to work together on quality improvement of care.

- Conditions for patient and parent recruitment

The essential selection criteria underlined by both patients/parents and professionals were a good relationship with the team, a desire to improve care for all patients and a willingness to take a step back and draw general lessons from their experience with the disease. Training on the general functioning of the hospital or the management of the disease have not been offered at recruitment and didn't appear to be a pre-requisite for participating. The professionals contributed their in-depth knowledge of the disease and its treatments to the discussions. This was made easier by the stability, expertise and experience of the team members. Extensive information on the program provided to the other patients or parents of patients followed up at the centre as well as to the hospital administration was indispensable to legitimize the participation of the patients and parents. Nevertheless, three parents stopped their participation at the end of the first year for reasons related either to the physician at the centre or to a worsening in the patient's health status. This illustrates the impact of the medical leadership on patients/parents' long-term involvement and confirms that a stable health condition on the part of the patient is a prerequisite to engage or stay in such a program [146].

- Participation at the quality improvement national training meetings

The participation of patients/parents in the national training meetings about the QI method and tools was an integral part of the program. The reimbursement of their

travel fees appeared to be mandatory to enable them to participate at these training meetings. Such participation gave all team members an equal opportunity to be trained in the quality improvement method. Given that none of the « students » had any prior knowledge of this particular quality approach, despite their different professional expertise and background, they all engaged in discussions effectively. The transparency of the outcomes from all centres involved at these meetings was another aspect of the method [154]. It provided results from the patient registry report by centre comparing patient health outcomes to identify potential best practices at some centres. Although this transparency was novel within the French CF care network, it was well accepted by the professionals and well received by the patients and parents, as it led to the choice of a theme for improvement at the centre. Condition for effective partnership between professionals and patients in QI work involved transparency of the results and the commitment to improve them [152]. Given that the goals were clear and shared from that time forward, the patients, parents and professionals were equally committed to achieving them during the program [168]. Moreover, the collaborative aspect of the program created a community of centres willing to continue sharing their work on quality improvement and their results as part of an open process of « benchmarking of practices » [169].

- Contributions made by patients and parents

The contributions made by patients and parents obviously depended on their frequent participation in the QT meetings at their centre. The experience of the patients and parents was brought to the discussions using questionnaires during the clinic visits or phone calls as well as patient shadowing during clinic visits and observation of multidisciplinary staff meetings. The joint work on these processes resulted after three years in the shared opinion of having implemented optimized processes. The patients and parents sometimes also contributed their own expertise (quality, IT, communication etc…) by « specific tasks » assigned to them depending on their wishes, availability and own expertise. Some examples were cited in the comments: a multi-purpose notebook was created to communicate with the care team about events at home, treatments prescribed and educational material ; internet surveys were developed and the results were analyzed for the QT ; a dashboard of indicators in the form of a smiley face was develop for the children to assess their care at the end of the visit; a « gazette » about the QI program was issued by

parents and adolescents; a bulletin board was created to display information about the QI project in the CFC. These contributions seem to have accelerated the QI work of the team and facilitated communication with the other parents/patients. Most often, it was ultimately difficult to attribute certain changes in the centre organization and process of care specifically to any specific team member – patient, parent or professional.

Questions raised by this partnership during PHARE-M in France

The following questions were raised by the stakeholders of the PHARE-M program, including the professionals' and the patients/parents' representatives, on the feasibility, efficiency and utility of this partnership during the program.

- **How were perceived the conditions in place to allow the participation of patients and parents in the program?**

The patients/parents as well as the professionals agreed that the organization of the PHARE-M throughout France created good conditions for their membership on QTs. All the respondents were satisfied with their experience, mostly favorable to further participation on a similar quality team and agreed with the necessity of an ongoing quality improvement process to continuously improve care at the centre. These opinions reinforce the French national PHARE-M team's belief that the program enhances the involvement of patients/parents along with their care teams to improve care at their centre. It also indicates that the participation in the program does not cause deleterious effects to the patients/parents involved, which could have come from the vision of the "defects" seen in the management of care at their centre.

Some items remained not consensual: they may be addressed through further experimentations during the next sessions of the program. They concern "the participation of a patient/parent should be facilitated by the reimbursement of other expenses such as child-care, lost working hours..."; "the necessity to include several patients or parents to ensure that more points of view are represented" and, "the need for patients/parents to understand the general functioning of the hospital". At the beginning of the program, questions about « representativeness » of the patients/parents involved were evoked. Should those involved be individuals recruited by the care teams according to the mentioned criteria or national patient organization or local patient group representatives, when they exist? Is the experience of patients/parents involved sufficient to inform QI work? Should the experience of other patients and parents be captured to complement their own?

These questions raise matters of legitimacy, democracy and responsibility. In the frame of our QI project, the legitimacy of the patient and parent involved appeared to be granted by the care team and not by a patient organization or patient group. It happened in some settings that the parent was a member of the CF local patient group but their involvement was decided upon by the care team and not requested by the patient group. Their position in the quality team did not change the rules for communication between the quality team and the patient group. It was clear that the patient or parent involved spoke to their own experience and not to that of a group of patients/parents. These questions are important and should be clarified at the meso- and macro-system level to facilitate and foster patient involvement in the quality improvement work with their care team, as it has been done for patient representation in hospital committees. Financial aspects related to the participation of the patient/parent in meetings with the care team, in particular travel fees or other allowances, could be part of this clarification.

- **How did the quality team's professionals perceive this participation and what were the feelings of the participating patients and parents?**

At the introduction of the program, barriers from professionals as well as from patients and parents had to be overcome. In the interviews, the switching of roles in parents (I come as a parent to the consultation, and in the quality group I commit myself as a user/ a designer of the process) and in patients (I come as a patient to the consultation, and I commit myself in the quality group as a user/improver) creates a tension between those positions of the patients/parents. The potential for tension arose when they didn't feel satisfied with their experience of the care delivered by the team or with the quality of communication with certain members of the team, and when they had not coped with a previous painful circumstance such as the diagnosis of CF for their child or the management of a complication of the disease. The attenuation of this tension is critical to gradually increase the involvement of parents and patients during the QIP. This attenuation was observed in the results of the investigations after three years, which lets us hypothetize that the QIP might have acted as a process of resilience for patients, parents and professionals.

A shift in the representation of care by professionals and patients/parents was observed in the course of the program towards a co-produced service which co-production is based on a mutual understanding of roles and competences, mutual

participation in communication and actions and respective responsibilities in delivering care. French teams that had previously developed a culture of patient therapeutic education and were used to partnering with patients/parents for their own care, were more favorable to patient and parent involvement in care QI work than the teams that had not. This observation, and whether the other teams have overcome their initial reluctance, will have to be further analyzed in the results by centre, as there was a high consensus after three years that “the presence of a patient or parent on the quality team is a given and an asset”. Our experience confirms that the more the professionals and the patients collaborated to plan and develop services, the more this collaboration was accepted among both the professionals and the patients [170].

Upstream conditions could be created to support the participation of patients/parents in the health system, especially in quality of care improvement programs along with their care team. In Canada, a framework for interprofessional education and collaborative practice was developed to address the needs in terms of skills and behaviors for professionals engaged in collaborative practice with healthcare practitioners, patients, families and communities [171]. Six domains were identified: interprofessional communication; patient and family centered care; role clarification; team functioning; collaborative leadership; and interprofessional conflict resolution. Several assumptions underpin this framework one of them being that interprofessional practice is not innate but requires a consistent culture of learning and practice. Further reflection would be needed to refine such a framework to the French system of health continuing education and thus foster the necessary shift towards patient involvement in quality of care improvement programs [172].

- **Is the quality of care at the centre appreciated the same way by patients and professionals after three years of joint work?**

All agreed that the care team was patient centred and eager to meet patient needs and insure safety of care. After three years of joint work, the awareness of the patients and parents on care organization and processes at their centre was high – similar to that of the professionals – concerning matters relevant to them: multidisciplinary care, patient education, the clinic visit process… But their awareness on some aspects of the organization such as the information system (patient electronic record) and the management of care guidelines, remained low.

Even so, these aspects are not to remain fatally out of their attention for quality of care improvement: the impact of educating parent in care guidelines on clinician adhering to them has been demonstrated in a pediatric CF program [173] and patient-led training in medical education has had an impact on the application of safety guidelines by clinical teams [174]. In Sweden, patient electronic records have been opened to allow patients access to their health record and provide input such as the schedule of the next visit, results on health outcomes followed at home and various mailings [175]. When these matters are explicitly shared with them as part of their care, patients and parents will probably be able to contribute to improve these fields by reporting their experience and needs.

- **How effective were perceived the quality teams in organizing the QI work and mastering the QI method and tools to which they had been trained?**

The work of the teams was fostered by leadership intending to achieve high quality of team functioning as well as by a shared decision-making process and clear shared goals, and its efficacy was supported by a good command of the quality tools including the ability to measure the results – despite a more difficult appropriation of PDSA cycles as a change management tool. The absence of consensus on items regarding availability of data in routine to follow and standardize the new processes and lack of support from other departments in the hospital raise doubts about the sustainability of continuous improvement of care at the CF centre after the 3 years. In the centres where the risk is high, a new session of the PHARE-M QIP is proposed on a new theme of improvement to sustain changes over time. The recognition of the PHARE-M program as a Professional Continuous Development program by the hospital continuing education department and the associated credits facilitates the CF teams' participation.

- **Was the participation of all QT members in the discussions and in decision making effective?**

All members felt that they could participate in decision-making, that attention was paid to their contributions and were at ease in suggesting ideas for change. The goals were clear and shared, which probably channelled the discussions amongst the members of the QTs who came from different backgrounds, experiences and skills. Normative characteristics were not dominant except the emphasis on the

goals. The patients / parents' contribution was highly appreciated but changes in the organization or process of care were not specifically attributable to them.

Reflections for further experimentations and research on involving patients' views in quality of care improvement programs

Our experience of patient/parent involvement in the PHARE-M QIP raise matters in relation to the nature and extent of the patient experience incorporated in the QI work. In 2005, Bate et al defined the concept of experience-based design (EBD) as a new way of co-designing health services with the patient in a context where they are no longer a « passive recipient of a product or service » but are « integral to the improvement and innovation process » [176]. Like other design sciences – such as architecture, healthcare is associated with the three aspects of functionality (*how well it does the job and fit its purpose - performance*), safety (*how safe and reliable it is - engineering*) and usability (*how the user interaction with the product or service is experienced*). According to Bate, *EBD is a user-focused design process with the goal of making user experience accessible to the designers, to allow them to conceive of designing experiences rather than designing services*. Which consequences such a vision has on QI work in healthcare? First, patients are incorporated for their experience of care, not necessarily for any prior expertise they may offer. Second, words are used to translate events (adverse or positive events) into experiences which may then be presented in the form of storytelling, sometimes played by actors. Third, experience amounts to more than views, complaints or satisfaction; it features *almost everything that is required to understand strengths and weaknesses and what needs to be redesigned in the care process*. For all these reasons, the acquisition and use of patient experiences in care improvement is a specialized activity which needs to be learned and practiced. It represents one valuable way to incorporate the patient experiences into care improvement. [177].

To address the question of patients' experience incorporated into QI work, specific « patient experience surveys » have been drawn up in some countries [178 ;179]. These surveys intend to collect information on the care pathway and on the characteristics of the care delivered to the patient in the previous months. They are designed to reflect the care that the patient should have received according to the standards of care for the disease. If they are administrated in ways that insure a good response rate from patients and parents, they enable the preparation of a center

report of Patient Reported Outcomes in terms of quality of care [180]. They may provide information about the variability of care across geographic or socioeconomic dimensions and avenues for quality of care improvement. These instruments help fill the gap between individual experiences of care and the general features of the care delivered to most patients.

We cannot conclude without comparing the commitment of patients and parents who accept or sometimes claim to be involved in QI programs to the activism defined by Rabeharisoa [181]. This commitment actually takes up the main features characterizing patient activism:

1) Include and shape the experiential knowledge of patients and parents;
2) Articulate it with credential knowledge in clinical, organizational and quality fields;
3) Reframe what is at stake, that is co-redesign the process of care;
4) Defend the cause: “the best possible care here and now for all patients”; and
5) Organize a network of expertise with credentialed experts in quality, patient therapeutic education, and academic instances.

Limitations of the study

Our research has some limitations. First, the sample of centres as well as patients/parents, all of which volunteered to engage in the PHARE-M QIP sessions and test the program before its roll-out throughout France, may not reflect general opinion at all CF centres in France from 2011 to 2015. Second, the appearance of numerous publications and mediated interventions in favor of taking patients' voices into account in healthcare services has triggered a beginning of a cultural shift in the last years in France. A movement called « Démocratie en Santé » emerged in France in 2015 building on this trend. In the latest PHARE-M sessions, it becomes more obvious to professionals as well as to patients and parents that the latter should be systematically involved in the QI work at the centre, and sometimes more than one at a centre. Their recruitment becomes also easier. It is hoped that arrangements will be made to facilitate patient participation in quality improvement of care, which will in turn have to be evaluated.

Table I: Number of Patients at the CFC engaged in PHARE-M by year

CF Program	Year PHARE-M	# Patients Data 2014	Pilot PHASE 2011-2013
PEDIATRIC			
Angers	2013	122	122
Bordeaux	2016	148	
Clermont-Fd	2013	103	103
Créteil	2015	109	
Dunkerque	2015	71	
Grenoble	2013	122	122
Lille	2015	181	
Lyon	2012	290	290
Nancy	2016	113	
Nantes	2012	104	104
Paris R Debré	2012	168	168
Rennes	2013	131	131
Roscoff	2012	75	75
Tours	2016	116	
Vannes-Lorient	2013	81	81
Versailles	2012	65	65
ADULT			
Lyon	2012	313	313
Nantes	2013	203	203
Rennes	2013	101	101
Montpellier	2015	197	
Reims	2012	131	131
Roscoff	2013	75	75
TOTAL Patients in PHARE-M Group		3019	**2084**
% Patients recorded in Registry		47%	**33%**

707 **Table II -** Opinions, concerns, and illustrative quotes regarding P&PI

Opinion	Concern	Quote
Patients/parents involvement in the Quality Teams		
The place of the patient/parent in the health system	This involvement upset assigned places, led to readjustments and reinterpretations, and highlighted resilient P&P profiles.	**Physician**: "Certain physicians are not ready to accept that there is a patient at the medical staff meeting, or a meeting like the ones that we have, who gets up and disagrees, who bursts in as a consultant who gives his or her opinion." **Parent1**: "I can see that parents who are often negative or react badly to certain situations are parents who are suffering. Sometimes I feel that I stand out from other people, because I am very optimistic by nature and I have a fighting spirit. This may be why I always go a little bit beyond."
Reason for participation by **Parents**	They affirmed contributing their testimonial on their experience and sticking to merely conveying their feelings and day-to-day experiences.	**Parent2**: "I do not aim to teach anyone in a medical setting their profession — one day a physician told me that I was not going to teach him his profession. In participating, I contribute my testimonial as a parent, and that is all. More than anything else, I want to contribute my positive energy and fighting spirit." **Parent3**: "My motivation in participating in the meeting with the pediatric team is being able to give my position as a parent. So I am going to tell them my feelings regarding some of their actions. Sometimes, when I tell them my feelings, they are surprised and tell me that they had not seen things in that way."

Reasons for **Patient** involvement from their perspective	**Wariness**: patients were waried of a medicalized world. **Consent and curiosity**: to get to know a setting, to better get to know the teams that they visited as their care providers. **Engagement under tension between**: *on the one hand*, the desire to understand, be curious, gain autonomy and confidence, and remove obstacles, and, *on the other hand*, the difficulty of pushing oneself to talk in front of others about one's experiences with an invasive disease that one would like to keep at a distance.	**Patient1**: "The idea of meeting with the physicians stressed me out a bit. I wondered what I was going to do, what I should say, how it was going to go." **Patient2**: "The differences that there could be between different hospitals were quite astonishing. For example, the outcomes in FEV1% were quite impressive compared to the outcomes we had. You saw that there were distinctly better figures than what we had, indeed... So that was a bit striking to me. It was also interesting to see how other hospitals functioned and provided care, and what could be done to improve quality for patients, basically." **Patient3**: "I gave my opinion on the feasibility of things. It is all well and good to say, 'We have to do X drainages, X treatments, X thingies, etc.,' but in the end, there is real life which is different from hospital life."
Projection of healthcare providers on patients in QT	The presence of a patient on the team questions healthcare providers' professional ideas and desire. It is tempting for healthcare providers to authorize themselves to have a particular conception of patients and parents and then to talk about them, about what they believe to be their experience, in the name of healthcare providers' experience and in-depth knowledge of the person — his or her journey and record.	**Nurse**: "It would also be necessary to critique healthcare providers. Healthcare providers need to create the patient's needs. That is what they do and they are proud of it. Nevertheless, it assumes having a patient who is completely ideal, compliant, etc. Such a patient does not exist. We do not know such a patient. We have never seen one before. These healthcare providers' pushes always make me very afraid, because I do not lose sight of the fact that they are about the ideal of healthcare providers." **Nurse**: "Sometimes, saying that people do not know their disease suits us well in the end, because we will be able to have an effect on them, to explain and re-explain to them. These people understand very well and live with their disease on a day-to-day basis better than us. I do not think that we have the slightest idea of what they are really going through. They know very well what this disease is about, that the final outcome is death. When these patients relax their efforts, we should respect this and not necessarily go and add things."

Table VII: Success factors sustaining long term patient and parent involvement in QI projects

Factors related to patients and parents: • Good relationship with the care team • Coping with the disease, its complications and the effects of treatments • Stable health condition of the patient or the child of the parent • Stable socio economical family situation • Motivation to improve care for all (beyond improving care for oneself) • Possibility of involving more than one patient or parent in the team to insure the presence of one of them at each meeting and to bring diverse experiences to the discussions (for instance parents of children of various ranges of age or transplanted and non transplanted patients…) • Ability to give time to the project, participating to the trainings and local meetings, and availability of communication tools (internet) at home
Factors related to the care team: • Mature relationship with the patient/parent: readiness to a partnership for care, being at ease with shared decision making and/or patient education • Leadership wishing to involve patients/parents on a long-term basis, « playing the rule » of transparency and effectively taking the responsibility for the project and for the change actions implemented • One professional being the correspondent of the patient/parent for the QI project solving practical issues • Awareness to the guidelines and consensus for care and ability to discuss/share them with the patient/parent • Attention paid to psychosocial difficulties encountered by the patient potentially contradictory with their involvement
Factors related to the QI method • Present the involvement of a patient/parent as a pre-requisite to engage in QI work, based on literature and a « safe » framework to recruit them • Take the financial charge of patient and parent involvement at the program level (thanks to an agreement with the patient organizations if possible) • Offer an appropriate set of communication tools towards the patients/parents followed at the center, including the patient group if any, as well as towards the hospital administration • Provide the same training on the quality methods and tools to the professionals and the

patients/parents involved

- Install resources for the QI work at the centre and manage the regular participation of the patient/parent or his update on the project
- Secure the framework with ethical rules allowing full participation of all members, recalling roles and responsibilities
- Start from where the teams are in terms of patient outcomes, professionals, processes and patterns
- Challenge the teams so that they fix their problems and choose a shared realistic goal to be achieved at the deadline of the project
- Offer new perspectives, facilitate benchmarking with other practices, provide access to guidelines and consensus for care to the whole team
- Provide an on-site Coaching to support the team in analyzing their processes of care from the point of view of the patient/parent (shadowing a patient) and reinsuring the place of the patient/parent involved
- Proceed by PDSA cycles, measuring the results of the test and adjusting if necessary, and share the results with the whole team
- Consider that the results achieved are attributable to the whole quality team and beyond, to the multidisciplinary team who implement the new process of care, and not to one member in particular, be it a patient/parent or a professional

VIII- DISCUSSION

Les points de discussion sur cette expérience sont multiples, sur l'intervention elle-même, sa normalisation dans le contexte du système de santé français, sa transposabilité à d'autres prises en charge de maladies chroniques et/ou rares, ou sur l'enseignement et la recherche sur ce type de démarche qualité collaborative. Nous avons retenu uniquement ***trois dimensions*** concernant l'apport du partenariat patient (et parent) à une démarche qualité des soins.

VIII-1. Démarche qualité et pratique collaborative en équipe pluridisciplinaire

Le lien entre pratique collaborative et démarche qualité des soins a déjà été montré par divers travaux de recherche internationaux et notre expérience en témoigne dans le contexte français d'une Filière de soins maladies rares, caractérisée par une collaboration ancienne avec les associations de patients ainsi qu'une forte collaboration internationale dans la recherche et les standards de soins. Ce contexte a créé les conditions requises pour l'implémentation de cette démarche qualité collaborative : 1) l'existence d'un programme adapté à la prise en charge de la pathologie, développé aux USA 2) le portage de la mise au point du programme dérivé français par le centre de référence de Nantes-Roscoff et 2) le financement initial des modalités pratiques de formation des équipes des centres par l'association. Un développement ex-nihilo d'une telle démarche aurait sans doute été impossible par la filière, faute de ressources de diverse nature. Seul un centre de référence maladie rare pouvait porter une action nationale, transversale à tous les centres de soin, grâce à une mission d'expertise reconnue par les institutions et des ressources dédiées. L'absence de soutien financier associatif aurait de même rendu impossible la participation des centres pilotes au programme. Si l'initiative d'associer les patients et parents dans les équipes de pilotage de la qualité des centres est le résultat d'une conviction et d'une culture portée par l'équipe du centre de référence, elle s'est produite « à bas bruit », par analogie avec ce qui était pratiqué dans le groupe ETP national porté par le CRMR, y compris pour le financement des frais des patients et parents par l'association.

L'étude portant sur cette démarche qualité montre, après trois années, un ***fonctionnement en équipe pluridisciplinaire,*** une prise de conscience de l'importance d'une ***démarche qualité continue*** pour améliorer les soins et la ***volonté des participants de continuer à s'y impliquer***. Elle témoigne des acquis des équipes caractéristiques des équipes dites ***performantes*** (cf. Fig.4) : une vision et un engagement sur des objectifs d'amélioration partagés, une cohésion d'équipe face au défi des changements organisationnels, un enrichissement à partir des différences entre ses membres, un environnement de travail sécure permettant de communiquer aisément ses idées, une écoute mutuelle et une prise en compte des idées des autres membres, une entraide (au détriment quelquefois du strict respect des tâches attribuées) en vue d'atteindre l'objectif, et un sentiment d'utilité et de performance de l'équipe. La poursuite du travail (au-delà de la thèse) devrait permettre de caractériser les contextes dans lesquels le fonctionnement interdisciplinaire a été associé à une plus grande efficacité de l'équipe en termes d'appropriation de la démarche et d'amélioration des soins.

Figure 4 : Dix caractéristiques des équipes « FAB » (FABulous)

VIII-2. Démarche qualité : progrès organisationnels et évolution culturelle

La démarche collaborative PHARE-M incluant la participation des patients/parents a été le vecteur de ***progrès organisationnels*** en même temps que d'une ***acculturation progressive aux concepts de la qualité*** dans les organisations des CRCM. Cette dynamique illustre des principes généraux développés par la psychologie au travail parmi lesquels : le lien entre la reconnaissance de ses pairs ou de l'équipe et le sentiment d'utilité ; le lien entre le sentiment d'utilité et la satisfaction au travail ; le lien entre l'accomplissement de l'action et la confiance en soi ; le lien entre la cohérence de l'action avec les valeurs (du soin) et le sens donné au travail ; etc. Nous proposons une lecture des résultats de l'étude qui témoigne de l'imbrication de ces deux dimensions au travers des éléments principaux rapportés.

Alors qu'au démarrage du programme PHARE-M, la démarche qualité était perçue par les professionnels comme une obligation administrative laborieuse qui prélève du temps sur les soins – vision partagée par les représentants de l'association de parents qui avaient cette crainte alors que les ressources des équipes étaient déjà contraintes -, le premier résultat a été la ***satisfaction au travail*** exprimée par les professionnels, qui disent avoir appris à ***construire ensemble des solutions*** à des problèmes, dont ils avaient parfois déjà conscience sans avoir réussi à les surmonter, et ***conduire les changements d'organisation*** nécessaires à leur résolution.

Les nouveaux processus définis et mis en place ont été rapidement ***généralisés*** à l'ensemble des professionnels du centre (pour des raisons évidentes de simplicité d'organisation) et, sous condition de ressource, à l'ensemble des patients dont l'état de santé le nécessitait (au-delà de la population cible initiale) en réponse à une valeur forte partagée par les équipes (et les patient/parents) d'équité de prise en charge des patients qui ont des besoins ou des difficultés similaires. Lorsque les

ressources professionnelles étaient jugées insuffisantes pour généraliser le processus à tous les patients qui le nécessitaient (par exemple, manque de temps de diététicienne pour généraliser la consultation diététique au cours de la consultation pluridisciplinaire de tous les patients ayant un BMI dégradé) deux choix ont pu être observés: maintenir un processus amélioré pour la population ciblée (une tranche d'âge priorisée par exemple) au cours du programme PHARE-M et négocier une augmentation de ressource avec l'hôpital pour l'étendre ensuite à tous les patients entrant dans le critère de patient à risque (généralisation sous condition de ressource); attendre pour mettre en place le processus défini que la ressource supplémentaire soit obtenue, soit auprès de l'hôpital, soit auprès d'un financement complémentaire de l'association Vaincre la Mucoviscidose dans le cadre des appels à subvention annuels. Dans ce dernier cas un sentiment de frustration et de « décrochage » de l'équipe par rapport à d'autres a été exprimé. Des différences entre centres sont nettement apparues sur le choix de l'option, à associer dans la suite de l'étude réaliste avec d'autres facteurs contextuels (leadership).

La ***méthode*** et ***l'accompagnement*** proposé par le programme PHARE-M ont permis aux équipes d'avoir le sentiment de ***l'efficacité*** du temps consacré à la démarche, garantie de la participation des professionnels et des patients et parents aux réunions sur la durée des trois ans. La présence du patient ou parent a crédibilisé les ***actions prioritaires*** par rapport à ***l'objectif d'amélioration clinique*** choisi par tous au regard des indicateurs de résultats de santé du centre. Le partage des résultats des indicateurs de santé avec tous les professionnels et les patients et parents en a été la condition première.

Cette conscience du « ***pouvoir agir*** » sur l'environnement de travail pour servir des objectifs cliniques qui motivent les soignants et les patients et parents, et la ***reconnaissance*** des efforts et des résultats obtenus, dans la communauté PHARE-M et quelquefois dans la communauté internationale, ont contribué à la satisfaction partagée. Concernant les professionnels, les publications préparées, malgré l'effort supplémentaire demandé, ont permis de développer un sentiment de ***fierté*** de participer à l'innovation et de communiquer sur leurs valeurs de soignants.

Une clarification s'est produite au fil de la démarche qualité entre l'effort de ***standardisation des processus*** et la nécessité d'une ***médecine personnalisée***, les deux principes paraissant de prime abord s'exclure mutuellement. Il est apparu que la standardisation des processus de soin et la réduction des variations dans leur exécution favorisaient 1) l'équité d'accès à des soins de qualité pour tous les patients, quelles que soient les conditions de service et 2) la prise en compte des besoins spécifiques du patient par les intervenants professionnels à l'intérieur du cadre aménagé par les processus, sans occasionner de désorganisation « en cascade » (pour les autres patients).

L'exemple de la réorganisation des consultations pluridisciplinaires pour garantir la succession optimale des interventions en réduisant les temps d'attente et en favorisant la transmission des informations entre les professionnels a permis d'en faire la démonstration. La volonté de standardisation des consultations pour tous les patients quelles que soient leur date de venue ou la période de l'année a posé la question de la disponibilité des ressources pluridisciplinaires (ou des locaux et équipements) à ces différents moments. La standardisation ne signifiant pas unicité de l'offre, des processus de consultation adaptés aux besoins des patients en fonction de critères de santé ou de situations personnelles ont été prévus, décrits et planifiés au cours de la programmation des consultations à venir. Des aléas peuvent

survenir le jour même, provenant du patient ou des professionnels, mais leur gestion par l'infirmière coordinatrice et si besoin une réunion d'équipe de quelques minutes en début de journée permet dans la majorité des cas de s'adapter sans désorganisation majeure de la consultation. De même, la structuration des staffs pluridisciplinaires a permis de planifier les situations des patients à examiner dans le temps imparti (qui ne permet pas toujours de revoir toutes les situations de tous les patients venus la semaine précédente). Une classification par critère de priorité a permis d'éviter un « tri subi » par le critère du temps écoulé. Le temps passé à l'examen de chaque situation a été mieux géré. La préparation de chaque professionnel sur les éléments à partager a permis d'augmenter le nombre de dossiers examinés…

La démarche qualité a produit un effet plus inattendu mais prévisible dans la culture française : ***apprendre de ses erreurs***, accepter l'imperfection pour s'améliorer, ne pas attendre d'avoir la certitude de « faire parfait » pour agir. La méthode PDSA théorise ce processus d'apprentissage par le test, qui admet la valeur des imperfections et libère ainsi l'action, dans un cadre sécurisé permettant d'en contrôler les effets.

La participation du patient ou parent a représenté un ***facteur de résilience pour l'équipe*** du fait de la dynamique qualité fondée sur : 1) la transparence des informations sur des processus ou des indicateurs insatisfaisants, 2) le choix consensuel de l'objectif d'amélioration et 3) la co-construction des actions pour atteindre l'objectif. Comme supposé (59) la dynamique d'amélioration enclenchée a permis de dépasser l'insatisfaction et la frustration initiales sur les résultats ou les processus et de mobiliser les énergies sur l'action plutôt que sur la recherche des « coupables » ou des « excuses ».

Au cours de la démarche, le regard des équipes sur la ***comparaison de leurs résultats de santé*** avec ceux des autres centres a été révélateur de l'humilité des professionnels et de la bienveillance des patients et parents. A aucun moment la comparaison des résultats de santé des patients n'a été l'occasion d'un jugement de « bon » ou « mauvais » centre, chacun ayant à son actif des points forts et des points à améliorer. La communauté PHARE-M a décidé, à l'issue du programme, de partager ses indicateurs de résultats cliniques dans le Registre en toute ***transparence.*** Le suivi de ses indicateurs par l'équipe est devenu un moyen de s'améliorer continuellement par rapport à soi-même et de s'informer sur les pratiques des centres qui ont les meilleurs résultats en vue de s'en inspirer sans complexe.

Deux points difficiles pour la qualité des soins sont apparus à travers les résultats de l'enquête, dont la résolution relève respectivement du méso-système (hôpital) ou du macro-système (Filière Muco-CFTR) :

- Le premier concerne l'utilisation d'un dossier électronique performant au sein de l'établissement utilisable notamment pour les réunions de staff pluridisciplinaire, et pour le suivi au long cours du patient
- La mise à disposition EN FRANÇAIS des recommandations de soin internationales et de leurs mises à jour, dans tous les centres, accessibles à tous les professionnels impliqués dans la prise en charge de cette maladie rare, et aux patients/parents.

En effet, la faiblesse ou l'inadéquation d'un dossier électronique patient pénalise la qualité des soins délivrés à des patients atteints d'une maladie, chronique et/ou rare, suivis pendant toute leur vie dans différents établissements et par différents

spécialistes, en France et, selon leur trajectoire de vie, à l'étranger. La continuité, la réactivité et la pertinence des soins peuvent en être affectées. La méconnaissance par les patients du contenu et de l'utilisation du dossier électronique, observée dans les résultats de l'enquête, reflète l'absence d'accès à leurs données patients : cette situation n'est pas inéluctable, d'autres pays ont ouvert l'accès à leurs dossiers aux patients (182).

L'absence de traduction française systématique des recommandations internationales publiées en anglais rend illusoire leur prise de connaissance généralisée par les soignants (et les patients) et donc leur application – et même leur discussion en équipe. Malgré la participation aux congrès internationaux de quelques cliniciens actifs dans les sociétés savantes et déjà parmi les mieux informés, cette absence de traduction en français génère potentiellement un retard à leur prise en compte dans tous les centres et une impossibilité pour les patients de s'informer sur les traitements qui leur seraient applicables, et de contribuer ainsi à l'adhésion à ces recommandations (183).

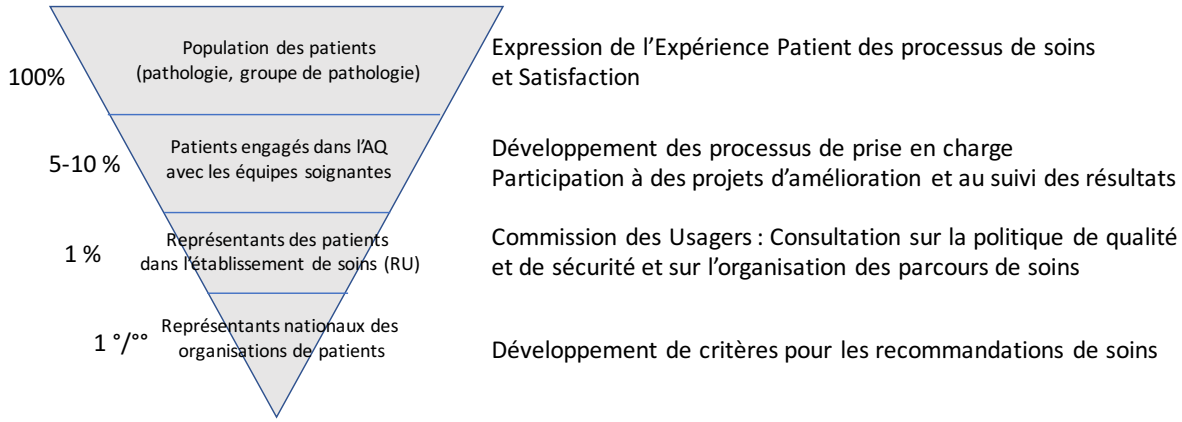
VIII-3. La révolution de la place des patients/parents dans l'amélioration de la qualité des soins

Dans de nombreux domaines de la santé, l'engagement des patients/parents se développe : des patients/parents experts sont recensés, formés et interviennent dans des formations aux étudiants en médecine ou dans d'autres disciplines (IFSI, école de kinésithérapeutes…), les patients/parents sont interrogés sur les priorités à donner aux orientations de la recherche, certains s'investissent dans les projets de recherche en tant que chercheurs, leurs avis ou leurs témoignages sont recueillis en matière de sécurité et de réorganisation des soins (184). Cette évolution semble irréversible et devrait s'accélérer encore avec les usages des nouvelles technologies de l'information dans le suivi des patients à domicile et la gestion des relations entre le patient et l'équipe de soin.

Cette évolution est internationale, même si elle s'inscrit dans la culture du pays et du système de santé qui en est le produit. Dans les pays francophones, le mouvement d'émancipation et d'autonomisation des patients atteints de maladie chronique initié par l'éducation thérapeutique se prolonge dans une dynamique de démocratie citoyenne en santé dont le potentiel dépasse les interventions traditionnelles des associations de patients au niveau du macro-système de soins. Les relais des associations dans tous les établissements ou instances en région sont limités et la portée de leurs actions locales dépendante de quelques individus fortement engagés. Mais le partenariat patient dans l'amélioration des soins ne peut être limité par un manque de couverture de la représentation associative dans tous les centres, alors que les patients/parents suivis dans le centre peuvent s'associer à une dynamique collaborative.

La proposition de représenter l'engagement des patients aux différents niveaux du système de santé semble confirmée par l'expérience du programme qualité dans la mucoviscidose (Fig.2).

Fig.2 (*rappel*) : Niveaux d'intervention des patients pour l'amélioration de la qualité des soins



Au niveau national, dans le cadre de la formalisation de la Filière Muco-CFTR (185), des ***instances mixtes*** regroupant les représentants de l'association et des représentants des centres de référence ont été constituées : le ***Conseil Médical*** est consulté sur les questions relatives à l'organisation des soins, prépare les révisions du PNDS ainsi que les programmes des rencontres scientifiques nationales ; le ***Conseil National,*** composé des membres des bureaux du CA de l'association et du CA de la Société Française, prépare les orientations communes à discuter dans les réunions institutionnelles où ses représentants sont conviés. Les informations sont diffusées d'une part par les réseaux associatifs auprès des adhérents et du public, et d'autre part par la société savante auprès des professionnels des CRCM.

A l'inverse, il existe peu (ou pas) de représentants des patients atteints de mucoviscidose parmi les RU. Dans les établissements hospitaliers où sont hébergés les CRCM, la présence des représentants associatifs est généralement limitée à une venue par an, à l'occasion des remises du chèque de subvention accordé par Vaincre la mucoviscidose. Il est même fréquent que le représentant associatif ne connaisse pas le CRCM, si lui-même ou son enfant n'est pas suivi ici. L'expérience patient du suivi dans le CRCM n'est pas partagée dans l'association, en dehors d'événements indésirables survenus et rapportés auprès de la direction médicale. Il semble irréaliste qu'un représentant de chaque pathologie soit membre de la CDU de chaque établissement.

La démarche qualité collaborative structurée pour la filière et appelée à se déployer dans chaque CRCM est ***le moyen d'associer localement patients/parents et soignants dans l'amélioration des soins***, dans le cadre d'une dynamique nationale structurée diffusant les recommandations nationales et internationales. Cette démarche et le partenariat patient dans l'amélioration de la qualité des soins semblent donc étroitement liés et permettre d'intégrer l'expérience patient de la prise en charge au CRCM et à domicile dans la réflexion sur le microsystème clinique.

Même si leur contribution a été jugée maximale dans les réunions locales des équipes, les concepteurs du PHARE-M ont maintenu au cours des sessions suivantes du programme, la ***participation des patients et parents aux réunions de formation nationales à la démarche***. L'objectif est de donner à tous les participants, soignants et patients/parents, une compréhension globale de la démarche et une identique maîtrise des outils, de leur permettre d'initier leur collaboration pendant les séances de formation, avant de prolonger et approfondir

ensemble les travaux dans leur CRCM. Ce parti pris de « loger tous les participants à la même enseigne » est apparu comme une garantie de bonne intégration des patients et parents dans l'équipe en tant que partenaires des soignants dans les discussions et non pas en tant que simples témoins de leur propre expérience. De fait, il ressort de l'auto-évaluation de leurs compétences, que l'acquisition des notions a été perçue au même niveau chez tous les participants - les mêmes difficultés ayant été relevées par exemple sur la maîtrise des cycles PDSA.

Il résulte de cette expérience une ***communauté de patients et parents intervenants dans cette démarche qualité*** capables de contribuer à l'amélioration de l'organisation des soins au sein des équipes professionnelles. L'animation de cette communauté en vue de partager leur expérience et de la diffuser pour « activer » d'autres patients/parents, de continuer à exprimer leur point de vue sans craindre une marge de dissensus avec l'équipe dans laquelle ils sont intégrés, en cultivant « *l'art de l'interstice* », est une préoccupation partagée avec l'animation de patients intervenants dans les équipes d'éducation (186).

Au-delà du modèle relationnel patient-soignant actuel, un débat s'ouvre dans notre pays sur une vision du ***patient partenaire de ses soins***, porté par les réflexions d'associations en lien avec des expériences de ce partenariat patient à l'étranger, notamment au Québec (187). Au-delà du patient « éduqué », apparaît la figure du patient « partenaire » considéré comme un soignant à l'égal des soignants professionnels, partant du constat que plus de 98% du temps consacré aux soins est passé en auto-soin et à peine 2% de ce temps est passé à des soins administrés par des soignants professionnels en établissement ou en ville. Le dernier stade du modèle de Montréal est ainsi discuté autour du concept d'empowerment (pouvoir d'agir) du patient, nécessitant un changement profond de la relation patient-soignant.

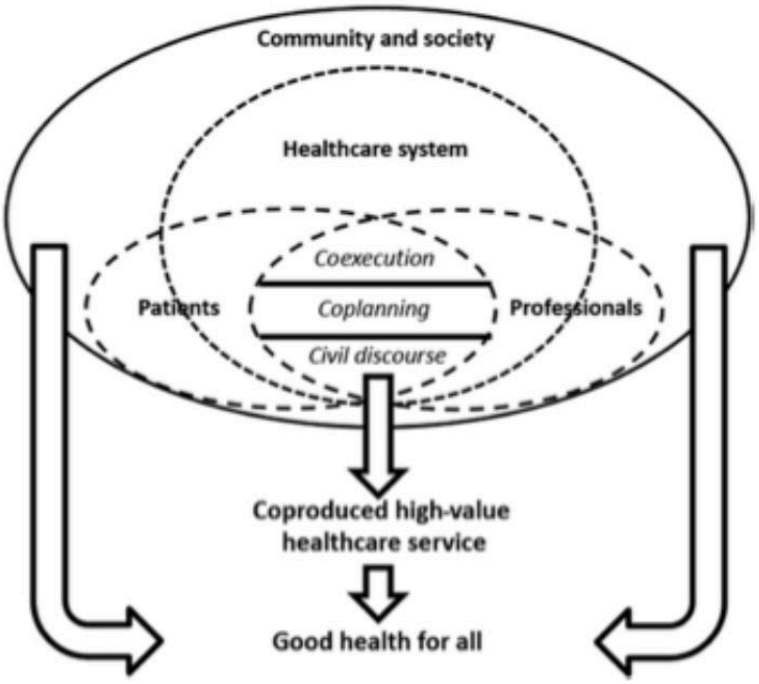
Quelques questions concrètes parmi les principales interrogations que pose ce changement radical de relation sont les suivantes : le patient doit-il participer au staff pluridisciplinaire pour les discussions qui le concernent (s'il le souhaite) ? La prescription de traitements doit-elle être établie avec le patient pour les seules thérapeutiques qu'il a décidé de suivre ? A l'aune de quelles prescriptions l'observance doit-elle être évaluée ? La notion d'observance ne doit-elle pas disparaître au profit du suivi du degré d'application par le patient de ses propres décisions de soins ? Quelle information et quelle éducation du patient lui permettent de prendre des décisions éclairées ? Quel accompagnement du patient par les équipes à l'occasion de décision d'arrêt de certains soins ? Quel partage de responsabilité sur ces décisions et leurs conséquences ?

L'ambition des promoteurs de ce modèle est une diffusion la plus large possible de la figure de patient partenaire de ses soins, tout en respectant les situations dans lesquelles le patient souhaite rester ou revenir à un stade de plus grande dépendance à l'égard des soignants, lorsque l'aggravation de sa santé ou des difficultés de vie personnelle l'y incitent.

Cette révolution du patient pleinement partenaire de ses soins et le changement relationnel avec les professionnels qu'elle entraîne rejaillit sur l'organisation du système de soin avec la mise en œuvre de ***nouveaux processus collaboratifs*** et de ***nouveaux indicateurs de process et de résul***tats, qui restent pour la plupart à définir. Une réflexion s'est engagée autour du « Collaborative Chronic Care Model », dans le prolongement du modèle de Wagner, pour conceptualiser un modèle de co-production des services de santé (cf. Figure 4), explorant les questions de co-

responsabilité et d'évaluation des résultats dans cette nouvelle vision du système de soin (188).

Figure 5 : Conceptual framework of healthcare service coproduction



Concrètement, la mise en œuvre d'une nouvelle relation soignant-patient partenaire de ses soins ne peut passer uniquement par la formation des soignants, même lorsque des patients interviennent dans cette formation pour partager leur expérience et affirmer ainsi leurs savoirs acquis au cours de leur vie avec la maladie.

Il s'agit bien de mettre en œuvre, dans l'organisation du système de soin (et avec des modalités de financement adaptées), les « espaces » permettant de telles expérimentations et d'en évaluer la faisabilité, l'utilité et les effets, en pratique. Ces processus collaboratifs ne pourront pas être définis sans ces patients partenaires, dans une démarche elle-même collaborative d'adaptation de l'organisation des soins à ce nouveau modèle de soin.

IX- PERSPECTIVES POUR L'INTERVENTION, LA FORMATION ET LA RECHERCHE SUR LES DEMARCHES QUALITE COLLABORATIVES

IX-1. Intervention PHARE-M

A mi-parcours de la formation de la Filière Mucoviscidose, avec 23 CRCM formés sur 45 fin 2017, de nouvelles réflexions doivent être menées au niveau national, entre les instances de la Filière (centre de référence et centres constitutifs) et l'association Vaincre la Mucoviscidose pour maintenir la dynamique d'amélioration continue de la qualité des soins pour les équipes déjà formées et motiver les équipes restant non formées à s'inscrire. En effet, si certaines équipes formées ont intégré la démarche dans leur fonctionnement et démarrent localement d'autres projets sur l'amélioration d'autres indicateurs de santé en utilisant les outils de la méthode, d'autres équipes considèrent le programme PHARE-M comme ayant permis ponctuellement d'améliorer leurs processus. Ces dernières équipes, bien que satisfaites de leurs avancées avec PHARE-M, n'ont pas standardisé en routine l'examen de leurs indicateurs de santé et le suivi de leurs processus. Des actions faisant office de rappels, l'animation d'une communauté et la valorisation par les instances de la filière paraissent indispensables pour que l'investissement dans le transfert de compétences continue à produire des effets.

Les actions prioritaires incluent :

- Faire état des réalisations, résultats obtenus et communications dans les congrès internationaux par les équipes françaises, à l'occasion de réunions nationales (Journées Francophones et Journées Scientifiques de la Mucoviscidose)
- Poursuivre la standardisation du programme dans les procédures d'accompagnement de l'association Vaincre la Mucoviscidose aux CRCM, à l'occasion des demandes de subvention de postes de soignants ou de projets par les CRCM, en incitant à une mise en cohérence de ces demandes ponctuelles avec une démarche qualité à long terme (incitation financière)
- Standardiser sous le format DPC des cycles post-PHARE de maintien des connaissances et de suivi des actions de changement, au-delà de l'année de formation initiale
- Proposer des « sessions PHARE-M avancées » à des équipes déjà formées mais qui souhaiteraient participer à une nième session sur un thème d'amélioration différent ou spécifique (prise en charge du diabète de la mucoviscidose, de la fin de vie ou des transitions) ou à l'occasion du remplacement du médecin leader ou de l'arrivée de nouveaux professionnels paramédicaux
- Renforcer l'animation de la « communauté PHARE-M » à travers d'une part les professionnels référents dans les CRCM et d'autre part les patients et parents investis dans le programme ou les instances associatives
- Evaluer les résultats de la démarche à l'échéance de 7 et/ou 10 ans (2019 et 2022) dans la Filière Mucoviscidose en comparaison avec les autres pays où elle est déployée (USA ; Canada ; Angleterre).

Si 7 CRCM adultes (sur 24 formés) ont déjà participé à la démarche qualité PHARE-M depuis 2012, en intégrant des patients adultes dans les équipes de pilotage, des difficultés particulières ont été identifiées dans le contexte de la médecine adulte. Ces difficultés sont de différentes natures : 1) la croissance régulière et forte de la file active d'adultes (+5% par an) sans que la disponibilité des ressources soignantes suive le même rythme de croissance 2) la grande disparité d'états de santé, du jeune

adulte dont la fonction respiratoire est « normale » (VEMS>80%) à l'adulte en attente de transplantation pulmonaire avec une fonction respiratoire sévèrement dégradée (VEMS% < 30%) et diverses comorbidités (diabète, hémoptysies, pneumothorax…) : la priorisation par l'urgence se fait au détriment des patients « en bonne santé » 3) une organisation de la médecine adulte « par organe » : en l'occurrence le patient atteint de mucoviscidose est suivi par le CRCM au sein du service pneumologie mais doit articuler ce suivi avec de multiples médecins d'autres spécialités 4) une transition pédiatrie – adultes souvent difficile pour des patients jeunes, éduqués mais déstabilisés par la transition et les changements de vie personnelle qui se produisent en même temps (autour de 18 ans) tels que l'entrée à l'université ou dans le monde du travail, un déménagement avec les formalités administratives associées par le changement de Maison Départementale du Handicap, et l'apprentissage de l'autonomie dans la vie quotidienne : il s'en suit une perte de vue plus ou moins longue ou un espacement du suivi au CRCM comme en ville et un décrochage des indicateurs de santé. Les CRCM adultes expriment des résistances aussi bien à la mise en pratique de l'ETP qu'à l'inscription dans le programme qualité, qui sont davantage culturelles (médecine de spécialité peu formée à la pluridisciplinarité et centrée sur l'intervention médicale) qu'organisationnelles. Une fois l'équipe engagée, la démarche qualité se déroule sans spécificité notable, sauf en ce qui concerne l'importance donnée aux processus de transmission entre services internes de **l'hôpital et avec les urgences. Les CRCM adultes ayant participé au programme** étaient de taille « moyenne » (entre 100 et 200 patients, avec un maximum à 280 patients). Une interrogation concerne les très gros CRCM (plus de 400 patients) dans lesquels le nombre de pneumologues prenant en charge des patients est élevé (5 à 7 médecins) et pour lesquels l'homogénéisation des processus peut être plus difficilement acquise.

IX-2. Intervention(s) PHARE-X

L'expérience acquise dans le contexte de la prise en charge de la mucoviscidose est transposable à d'autres prises en charge pluridisciplinaires pour d'autres pathologies chroniques et/ou rares, ainsi qu'en témoignent les expériences américaines pilotées par le Dartmouth Institute. Des conditions favorables ont été identifiées dans le contexte de la mucoviscidose pour faciliter cette transposition avec la participation des patients et parents (Tableau III), en lien avec les représentants associatifs selon les différents niveaux de la pyramide de participation. Des programmes de type PHARE-X pourraient notamment faire l'objet de développements dans d'autres Filières maladies rares, dans le cadre d'actions transversales visant l'amélioration de la qualité de la prise en charge en associant notamment l'optimisation des processus organisationnels et la mise en œuvre de l'éducation thérapeutique.

Tableau V : Facteurs de succès de la participation des patients et parents dans une démarche qualité des soins (*extraite de l'article V – Traduction française Tableau VI page 172*)

Factors related to patients and parents: • Good relationship with the care team • Coping with the disease, its complications and the effects of treatments • Stable health condition of the patient or the child of the parent • Stable socio economical family situation • Motivation to improve care for all (beyond improving care for oneself) • Possibility of involving more than one patient or parent in the team to insure the presence of one of them at each meeting and to bring diverse experiences to the discussions (for instance parents of children of various ranges of age or transplanted and non transplanted patients…) • Ability to give time to the project, participating to the trainings and local meetings, and availability of communication tools (internet) at home
Factors related to the care team: • Mature relationship with the patient/parent: readiness to a partnership for care, being at ease with shared decision making and/or patient education • Leadership wishing to involve patients/parents on a long-term basis, « playing the rule » of transparency and effectively taking the responsibility for the project and for the implementation of change actions • One professional being the correspondent of the patient/parent for the QI project solving practical issues • Awareness to the guidelines and consensus for care and ability to discuss/share them with the patient/parent • Attention paid to psychosocial difficulties encountered by the patient potentially contradictory with their involvement
Factors related to the QI method • Present the involvement of a patient/parent as a pre-requisite to engage in QI work, based on literature and a « safe » framework to recruit them • Take the financial charge of patient and parent involvement at the program level (thanks to an agreement with the patient organizations if possible) • Offer an appropriate set of communication tools towards the patients/parents followed at the center, including the patient group if any, as well as towards the hospital administration • Provide the same training on the quality methods and tools to the professionals and the patients/parents involved • Install resources for the QI work at the centre and manage the regular participation of the patient/parent or his update on the project • Secure the framework with ethical rules allowing full participation of all members, recalling roles and responsibilities • Start from where the teams are in terms of patient outcomes, professionals, processes and patterns • Challenge the teams so that they fix their problems and choose a shared realistic goal to be achieved at the deadline of the project • Offer new perspectives, facilitate benchmarking with other practices, provide access to guidelines and consensus for care to the whole team • Provide an on-site Coaching to support the team in analyzing their processes of care from the point of view of the patient/parent (shadowing a patient) and reinsuring the place of the patient/parent involved • Proceed by PDSA cycles, measuring the results of the test and adjusting if necessary, and share the results with the whole team • Consider that the results achieved are attributable to the whole quality team and beyond, to the multidisciplinary team who implement the new process of care, and not to one member in particular, be it a patient/parent or a professional

IX-3. Intégration des démarches qualité collaboratives dans la formation des soignants

L'amélioration continue de la qualité des soins fait partie intégrante du métier du soignant, quelle que soit la discipline, médicale ou paramédicale, concernée. Ce constat est d'autant plus prégnant lorsque la prise en charge de la pathologie chronique doit répondre aux caractéristiques du ***Chronic Care Model***. De plus, l'évolution vers un nouveau modèle de soins partenaire pour des patients mieux informés, éduqués et empouvoirés – encore accentuée par l'émergence de la e-santé, rend incontournable leur participation à la réflexion et à l'organisation des processus de prise en charge (189).

Cette pratique soignante pluridisciplinaire et collaborative n'est pas innée. Elle répond à un cadre d'enseignement élaboré, notamment au Canada, et dont la mise en œuvre à la fois en formation initiale et en formation continue (DIU) inclut la participation de patients partenaires ou experts. Ce cadre est orienté sur les compétences de « management » : fonctionnement d'équipe ; clarification des rôles et responsabilités de chacun ; leadership collaboratif et réparti ; gestion des conflits éventuels au sein du groupe et développement de points de consensus. Impliquer des « patients-ressource des démarches qualité collaboratives » dans la formation des soignants à ces pratiques collaboratives, comme se sont développées récemment des expériences d'implication de patients « experts de la vie avec la maladie », constituerait un facteur facilitant la mise en place de ce type de démarche qualité de terrain avec les équipes soignantes et les patients volontaires. La construction d'un *DIU pluridisciplinaire mucoviscidose* destiné aux nouveaux soignants et aux patients et parents désireux de devenir « patient expert », incluant un volet sur le partenariat soignants-patients en plus d'un volet sur la clinique et d'un volet sur l'éducation thérapeutique, constitue une initiative en formation continue pour consolider et développer la pratique pluridisciplinaire collaborative dans la Filière. Ce DIU est complémentaire du DPC PHARE-M en préparant les acteurs au travail collaboratif de la démarche qualité dans le CRCM.

Une mise en cohérence est nécessaire entre d'une part cette démarche qualité initiée par les équipes soignantes en partenariat avec leurs patients et d'autre part le niveau de l'établissement de santé et au-delà, le niveau du territoire de santé ou de la filière nationale de soins. La contribution de la démarche qualité PHARE-M à la certification, à travers l'élaboration de plans d'actions pluriannuels, l'évaluation des pratiques professionnelles, la réalisation de patients-traceurs et un partenariat avec les départements qualité, est un exemple concret de cette mise en cohérence au niveau de l'établissement – qui n'est toutefois pas encore relayée jusqu'au niveau de la CDU. A fortiori, le prolongement de cette démarche sur le parcours de soins du patient chronique en dehors de l'hôpital, nécessite un appui dans les territoires et au niveau national, au sein de structures mixtes associant professionnels et représentants des patients. Le cadre du DPC peut permettre d'engager les soignants libéraux aux côtés des soignants hospitaliers dans un programme qualité tel que PHARE-M. A défaut d'un élargissement aux parcours de soins, ces démarches risquent de rester des expériences centrées sur l'établissement de soins, ne répondant que partiellement aux exigences du *CCM* et aux besoins des patients.

IX-4. Vers une contribution française à la recherche internationale sur les démarches qualité des soins intégrant la participation des patients et sur leur apport dans l'introduction des innovations dans les organisations de soin

De nombreuses expériences de démarches qualité sous le format des Learning and Leadership Collaboratives sont menées dans les pays d'Amérique du Nord ou d'Europe, notamment en Suède et en Angleterre, ainsi qu'en Asie du Sud-Est. Une communauté internationale est active sur ces sujets, à la fois autour des « microsystèmes cliniques » (Festival annuel des microsystèmes cliniques de Jongköping, Suède) et des forums « Quality and Safety in Healthcare » organisés par BMJ Quality Safety et le IHI. Des recherches sont menées pour identifier les facteurs de succès et les éléments de contexte favorisant l'efficacité de ces démarches et évaluer leur performance. Elles ont permis de concevoir des méthodes d'évaluation des interventions complexes, complémentaires des méthodes quantitatives classiques.

Participer à la recherche internationale sur ces questions, et en particulier sur le thème de la participation des patients à ces démarches qualité collaboratives permettrait une contribution à la réflexion internationale et motiverait les équipes soignantes, notamment médicales, à s'impliquer dans l'amélioration de la qualité des soins en vue de valoriser leurs résultats.

Par ailleurs, la transformation émergente du système de soins, notamment avec l'introduction des nouvelles technologies de la e-santé et l'empouvoirement accru des patients, laisse entrevoir des évolutions importantes dans les processus de prise en charge. ***Innover, c'est introduire de manière structurée et collective, un changement qu'il soit majeur ou mineur dans ses fonctions***. Innover dans le système de santé peut procéder d'une démarche incrémentale (évolution progressive du modèle de soins) ou de rupture technologique et/ou organisationnelle (e-santé). La mise en place de l'innovation dans le système de soins implique au premier chef les équipes soignantes et les patients en ce qui concerne le changement des pratiques, des outils et des habitudes. L'intégration de l'innovation dans une organisation de soins, qu'elle soit technologique ou clinique – et plus vraisemblablement les deux à la fois, relève ***d'une démarche structurée*** qui doit permettre une réelle modification des pratiques quotidiennes des acteurs pour être un succès.

La vision organisationnelle associée à la conduite du changement qui est le propre des démarches qualité permet de structurer l'introduction de l'innovation, tandis que leur dimension pluridisciplinaire et collaborative, incluant les patients, permet de s'approprier l'innovation en ***co-construisant ses usages*** dans le cadre d'une prise en charge adaptée aux besoins des patients et aux valeurs du soignant. Les deux étapes clés de préparation du terrain (acteurs et organisations) et d'appropriation de l'innovation, sont facilitées dans le cadre d'une démarche qualité focalisée sur le changement des pratiques et qui intègre ***un accompagnement par le porteur de l'innovation*** technologique, médicale ou culturelle. Le développement de la mise en pratique de l'éducation thérapeutique du patient dans les équipes pluridisciplinaires des CRCM au cours du programme PHARE-M a été soutenu par l'étroite collaboration avec les porteurs de la démarche ETP mucoviscidose nationale et l'offre d'outils éducatifs aboutis et adaptés. La recherche sur ***la synergie entre les démarches qualité et l'introduction de l'innovation dans les organisations de soins*** constitue encore un champ important à explorer.

Tableau VI : Facteurs de succès de la participation des patients et parents dans une démarche qualité des soins (*Traduction française)*

Facteurs dépendants des patients et parents : • Avoir développé une bonne relation avec l'équipe soignante • Avoir surmonté l'épreuve du diagnostic de la maladie ou de ses complications récentes, et le poids de la gestion des traitements • Se trouver dans un moment relativement stable du développement de la maladie ou de celle de son enfant • Se trouver dans une situation économique et familiale relativement stable (pas dans une situation d''urgence) • Être motivé par l'amélioration de la prise en charge pour tous, au-delà de l'amélioration de sa propre prise en charge • Recourir à plusieurs patients ou parents dans l'équipe de pilotage pour s'assurer de la présence permanente d'au moins un représentant sur l'ensemble des réflexions, ou pour apporter des points de vue plus divers sur la problématique traitée (parents d'enfants d'âges différents ou patients de conditions différentes : âge, complications, statut vis-à-vis de la transplantation…) • Capacité du patient ou parent à consacrer du temps au projet, en participant à des réunions locales et aux réunions de formation nationales, maîtrise des outils de communication via internet au domicile
Facteurs dépendants des professionnels de l'équipe soignante : • Avoir développé une relation mature avec le patient ou parent : une attitude partenaire pour les soins individuels, la mise en œuvre du processus de prise de décision partagée, et l'accent mis sur éducation thérapeutique du patient ou parent • Avoir développé un mode de leadership encourageant la participation de chaque membre de l'équipe, y compris le patient ou parent, en jouant la transparence sur les résultats de l'organisation, prenant la responsabilité du projet et des actions de changement retenues et mises en œuvre • Un professionnel paramédical est le correspondant privilégié du patient ou parent pour le programme qualité et l'aide à résoudre ses difficultés de participation de toute nature • Les professionnels sont bien informés des recommandations de soins, et sont prêts à les partager de façon ouverte et objective avec le patient ou parent. • Les professionnels sont vigilants à l'impact psychologique de la participation du patient ou parent sur lui-même ou sur d'autres membres de l'équipe.
Facteurs dépendants de la démarche qualité : • Indiquer que la participation des patients/parents est un prérequis pour l'inscription d'une équipe, grâce à un cadre pour leur recrutement et des conditions adaptées à leur participation et informer l'administration de l'hôpital de cette participation • Prendre en charge les frais de mission du patient ou parent au même titre que ceux des professionnels participants • Informer les patients ou parents suivis par le centre du rôle du patient ou parent recruté et les tenir régulièrement informés de l'avancement du programme • Former les patients ou parents recrutés à la démarche qualité comme tous les membres professionnels des équipes de pilotage

- Attribuer des ressources pour que l'équipe de pilotage puisse se réunir et travailler dans le centre, et pour associer le patient ou parent, physiquement autant que possible, via internet si besoin
- Définir des règles d'éthique de collaboration permettant l'entière participation de chaque membre, en rappelant les rôles et responsabilités de chacun
- Prendre en compte la situation de départ de chaque équipe en termes d'indicateurs de santé des patients, de disponibilités des professionnels, de processus, d'habitudes de travail et de culture
- Motiver les équipes afin qu'elles identifient elles-mêmes leurs problèmes et qu'elles choisissent un objectif d'amélioration atteignable à la fin de la durée de leur projet.
- Proposer de nouvelles perspectives, organiser un benchmarking de pratiques dans un centre ayant de bons résultats ou connus pour une pratique innovante, donner accès aux recommandations et publications scientifiques à toute l'équipe y compris le patient ou parent.
- Accompagner sur site les équipes dans la démarche qualité pour soutenir le travail de l'équipe, analyser les processus de soin (shadowing a patient) et réaffirmer la place du patient ou parent participant
- Renforcer l'appropriation de la méthode des cycles PDSA par l'équipe à travers une revue fréquente de son utilisation sur site, en assurant le suivi des résultats du test par toute l'équipe et en facilitant l'expression d'ajustements si nécessaire
- Considérer que les résultat obtenus sont le fruit du travail en commun de toute l'équipe et au-delà de l'équipe pluridisciplinaire du centre, et non pas à un membre en particulier, afin de développer la solidarité et la cohésion de l'équipe

X- CONCLUSION, LIMITES & OPPORTUNITES

Conclusion

Notre expérience témoigne de la ***faisabilité et de l'utilité de ce type de démarche qualité collaborative dans notre système de soins français***, permettant d'intégrer des particularités de notre modèle de soin pour les maladies chroniques – comme les programmes d'éducation thérapeutique formalisés ou les traitements à domicile liés à la maladie (et remboursés au titre de l'ALD). De plus, si les systèmes de soins de chaque pays présentent indubitablement des spécificités et ont développé une culture d'organisation inscrite dans la culture du pays et adaptée à son système de santé, les similarités de pathologies chroniques qui ne connaissent pas de frontières, la diffusion des traitements issus de la recherche internationale et des standards de soin homogènes entre les pays « développés » plaident pour la participation des équipes françaises à la ***communauté internationale d'échanges*** sur l'organisation des soins et les ***démarches qualité collaboratives***.

Ces démarches proposent une méthode et un cadre propice pour ***un engagement de longue durée de patients*** atteints de maladie chronique ***volontaires,*** aux côtés de leurs équipes soignantes, pour contribuer à la mise en place d'une prise en charge centrée sur des objectifs cliniques et les besoins des patients (190). Démarche qualité collaborative et fonctionnement en équipe performante sont ***indissociables*** et source d'une satisfaction au travail des professionnels et d'une meilleure compréhension mutuelle entre les professionnels et les patients et parents. La culture du travail en équipe s'apprend et un ***cadre d'enseignement*** a été proposé, pour préparer les professionnels et les patients volontaires à l'exercice de pratiques collaboratives. L'articulation de ces démarches avec la ***certification*** des établissements est possible, souhaitable, et réalisable. Au-delà de l'établissement hospitalier, un champ reste à explorer sur l'expérimentation de programmes d'amélioration de la qualité des ***parcours de soins***, par des équipes pluridisciplinaires de plusieurs établissements ou en réseau de soins, et avec des patients, articulé avec les instances territoriales de santé et les représentants des patients. La ***synergie entre les démarches qualité collaboratives et l'introduction de l'innovation organisationnelle en santé*** constitue un champ pour l'expérimentation et la recherche. L'évolution vers un patient pleinement partenaire de ses soins associée à une évolution profonde de la relation patient – soignant encouragera la participation de patients-intervenants dans la co-conception et la mise en œuvre d'une organisation des soins adaptée. Une démarche qualité développée dans le microsystème clinique peut en être le support.

Limites & Opportunités

L'évaluation du programme dans le cadre de cette recherche a bénéficié d'un environnement favorable du fait de l'innovation que constituait cette démarche dans les CRCM entre 2011 et 2013. Une évaluation à 5 ou 10 ans des effets attribuables au programme serait confrontée à des difficultés méthodologiques nouvelles, du fait des interventions synergiques menées dans la filière pour développer l'éducation thérapeutique et former les soignants et des patients experts au modèle de soin mucoviscidose (DIU). Il n'est pas au demeurant certain qu'une telle évaluation soit une priorité.

James Moses, dans son billet publié sur IHI Open School, souligne la priorité de l'évaluation continue pour soutenir l'amélioration de la qualité : « *Improvement, as we know, is about cycles of testing – not for proof of effectiveness – but, in essence,*

cycles of testing to learn what's going to improve. And so, your measurement is not about "pre" and "post". It's about continually measuring your metric of interest that you want to move, and coming up with not just one intervention, but multiple interventions, based on learning from prior cycles so that you can actually get to the point of realizing sustained improvement through a series of interventions that were informed by testing in the actual system that you want to improve. I think that a lot of times the sustained improvement realized in quality improvement helps to be justified and validated through good research assessment of its effectiveness. But I don't think that we should have improvement prioritize proof of effectiveness over sustained improvement. »

La question de la prise en compte, dans les démarches d'amélioration de la qualité des soins, de la réalité de ***l'expérience des soins*** (191 ;192) vécue par les patients s'est posée depuis quelques années, dans la communauté mucoviscidose et dans d'autres pathologies chroniques, notamment le cancer (193 ;194 ;195). Cette expérience est à distinguer de la satisfaction du patient en ce qu'elle décrit le parcours de soin réalisé par le patient et caractérise les modalités du suivi aux différentes étapes du parcours. Elle est ainsi tantôt à l'origine de la définition de parcours cibles tantôt outil de l'évaluation des parcours réels au regard des recommandations cliniques.

L'amélioration de l'expérience patient est un champ émergent de réflexions qui rejoint les thèmes au cœur des démarches qualité collaboratives. L'enrichissement des démarches qualité par les résultats d'enquêtes portant sur l'expérience patient du parcours de soins est une opportunité pour maintenir une amélioration continue de la qualité des soins, l'amélioration de cette expérience patient pouvant constituer une mesure, complémentaire de l'évaluation de l'implémentation du Chronic Care Model, de l'impact d'une démarche qualité des soins. Expérience patient et démarche qualité continue contribuent au changement de paradigme de gouvernance du système de soins, qui vise à placer véritablement l'usager au cœur du système de santé.

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RESUME

Titre : Partenariat patient dans une démarche d'amélioration de la qualité des soins : l'expérience du programme qualité en mucoviscidose

Contexte : Un programme d'amélioration de la qualité des soins est implémenté depuis 2011 en France dans la filière mucoviscidose en adaptant la démarche qualité collaborative développée aux USA par la Cystic Fibrosis Foundation et le Dartmouth Institute pour les centres spécialisés américains.
Objectif : Evaluer l'apport de la participation des patients et parents d'enfants malades, aux côtés des professionnels soignants, dans les équipes qualité des CRCM formés au programme qualité
Méthode : Design mixte de recherche associant un volet quantitatif sur l'évolution des indicateurs de santé des patients et un volet qualitatif selon une étude réaliste à travers une enquête par questionnaire et focus group auprès des patients, parents et professionnels impliqués dans le programme qualité.
Résultats : Les résultats témoignent des bonnes conditions créées par le programme pour la participation des patients et parents, de l'appropriation de cette démarche par les professionnels et les patients/parents, de son utilité perçue pour améliorer la qualité des soins et de l'évolution de la représentation de la place de l'usager dans l'amélioration de l'organisation et des processus jusqu'à la considérer comme une évidence et un atout.
Discussion : La démarche qualité développe la pratique collaborative interdisciplinaire et avec les patients/parents. Les progrès organisationnels observés sont concomitants du développement d'une culture de la qualité. L'implication des patients/parents dans une démarche qualité au sein du microsystème clinique constitue une évolution majeure pour l'amélioration du système de soin.

Summary

Title: Patient and parent involvement in a Quality Improvement Program in Cystic Fibrosis (CF) care in France

Background: A quality improvement program (QIP) has been implemented since 2011 in the CF care network in France adapting the Learning and Leadership Collaborative program developed in the US by the CF Foundation and the Dartmouth Institute for the American CF Centre network.
Objective: Assess the contribution of patients and parents of children with CF engaged in the CF center quality improvement teams, besides their care team, to improve care in their center.
Method: Mixed design research including a quantitative study focusing on patient outcomes evolution and a qualitative study according to a realist approach using a questionnaire and focus groups to patients, parents and professionals engaged in the QIP.
Results: Participants attested of the good conditions implemented by the QIP to allow patient and parent engagement, a consensus about the appropriation of the quality method and tools, the usefulness of the program to improve the quality of care; in the end, patient and parent engagement in the QIP was found to be a given and an asset.
Discussion: The QIP has developed collaborative practice in multidisciplinary teams and with patients and parents. Organizational improvements were concurrent with a cultural shift towards a culture of quality improvement. Patient and parent engagement in a QIP within the clinical microsystem is a major development for the improvement of the health care system.

Mots clés : amélioration des soins, qualité des soins, démarche qualité, collaboration, engagement des patients, mucoviscidose, maladie rare, expérience patient

Keywords : healthcare, quality improvement, learning and leadership collaborative, cooperative behavior, patient engagement, cystic fibrosis, rare disease, patient experience

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G.Rault, P.Lombrail

Strategies for care quality improvement in Cystic Fibrosis

Cystic fibrosis is a "model" of international collaboration for therapeutic research, social science research, development of international guidelines and care management all at once, because of its characteristics: it is a genetic disease which is progressive, chronic and multisystemic, with a prevailing impairment of the respiratory function, and also a "rare disease", albeit the most common of "rare diseases" in Caucasian populations.

Globally, the 1980s were marked by the first successful pulmonary transplant on cystic fibrosis patients and the discovery of the *CFTR* gene. "Resignation" gave way to hope, based on the acceleration of research efforts shown by the simultaneous increase of articles on this disease.

In France, a greater interest for this disease from medical teams, a better care management by multidisciplinary teams in specialized health centres and the creation of the National Cystic Fibrosis Observatory (1992) marked this turning point. In the early 2000s, the national application of systematic neonatal CF screening led to a structuring characterised by the recognition by the health authorities of Cystic Fibrosis Centres (CFCs) (2002) meeting the criteria of CF care specifications. In the frame of the National Plan for Rare Diseases, 2 expertise centres for CF (CF-CERD) were certified in 2006 and the CFTR care sector was identified (2014).

The implementation of the PHARE-M care quality improvement program (*'A hospital-based program for improvement of results and expertise in cystic fibrosis care')* is the logical, yet pioneer, extension of the care sector structuring for this rare disease. The PHARE-M puts forward a major development to bring interdisciplinarity at the center of the teams' practice and to strengthen the partnership with patients and parents to improve patient care at their CFC.

Indeed, this quality approach targets the clinical microsystem, which includes the CFC professional team, patients and their relatives, and professionals in the city involved in care, because the health results and the patient's quality of life depend on the functioning of the overall system [1]:

- in a systemic vision of the care production process (the care manufacture): "*a system is perfectly designed to produce the results it produces*"

- and the assertion of the interdependency of the various links: "*no one is solely responsible for the results, whatever they are*"

This cultural evolution is supported by a collaborative dynamic and requires an ethics of cooperation that enables exchanges between CFCs on their results and on the "potential best practices" identified through benchmarking. It is perpetuated through the implementation of measuring tools that allow to follow the results of the actions undertaken and the facilitation of a community that exchanges on continuous quality improvement. It is the subject of research on prevention and healthcare services, a token of continuous improvement of care quality founded on "evidence-based" data.

What was the genesis of the PHARE-M program in cystic fibrosis* in France?

The PHARE-M program rely on the success of the American experience hailed by an article in the Thorax journal in August 2011[2]. The triggering event that occurred ten years earlier was the publication by the US Institute of Medicine of the article entitled "Crossing the quality chasm: a new health system for the 21st century" [3].

Immediately following this publication, the American Cystic Fibrosis Foundation (US CFF) called upon the services of experts from the Institute for Healthcare Improvement (IHI, Harvard) and The Dartmouth Institute Microsystem Academy (TDIMA). It then observed a great disparity of survival results from one center to the next, based on the indicators found in the US Cystic Fibrosis Registry; it organised a benchmarking visit of the 10 "best" centers to identify the key success factors; it decided to release with full transparency the results indicators for the various centers; and it made the decision to establish a Care Quality Improvement Program in the United States.

From 2002 to 2013, the CFF organized, with experts from the TDIMA, annual collaborative sessions under the program and gradually tailored this latter to the specificities of cystic fibrosis care management in the USA [4]. The special May 2014 issue of the BMJ Quality & Safety journal entitled "Ten years of improvement: innovation in cystic fibrosis care" [5] recounts in detail that experience and the results achieved.

From 2008 onwards, close ties developed between the Nantes-Roscoff CF-CERD, the 'Vaincre la Mucoviscidose' association and the US CFF[6]. In September 2011, the CF-CERD launched the PHARE-M program with a pilot phase, involving 7 CFCs representing about 1,000 patients out of nearly 6,000 patients present in the French Cystic Fibrosis Registry in 2011 [7].

* A hospital-based program for improvement of results and expertise in cystic fibrosis care

What can be found in this supplement?

Beyond the origins of the PHARE-M program, the purpose of this special issue is to report on the quality approach implemented since 2012 at the CFCs involved under the PHARE-M program, its standardization in the landscape of continuing hospital training and the results observed in 2015 after three years of ongoing work. These articles therefore contribute to introducing this intervention in various clinical microsystems and concern different sectors of cystic fibrosis care, nutritional care in pediatrics [8], psychosocial care for teenagers [9], as well as the preparation for pulmonary transplant in adults [10].

In December 2012, the ministry selected and funded the PHARE-M Performance research program, which seeks to assess the impact of the PHARE-M on the evolution of patient health indicators and includes a realistic analysis "to understand what works, for whom and under which circumstances"[11]. The description of the research program protocol [12] and the results of the quality controls of data transferred to the Registry conducted for that purpose [13] enable to understand the assessment methods of the PHARE-M quality program performance and identify their limitations. The conclusion seeks to emphasize the contributions of patients and parents to this collaborative program for the improvement of care quality side by side with the teams at their CFC [14].

Despite the difficulties related to the transposition and adoption of such an approach in different cultural and healthcare systems, we can state that this strategy has had a profound impact on the network of CFCs trained in France, with a great satisfaction within the healthcare teams, an improvement of their interdisciplinary practice, the development of patient therapeutic education, and a strengthened collaboration between patients, parents and healthcare staff in improving care, all of the above supported by a constant research endeavour.

Trans-Atlantic Collaboration: Applying Lessons Learned from the US CF Foundation Quality Improvement Initiative

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Abstract

Background:

The Cystic Fibrosis Center of Expertise for Rare Diseases (CF CERD) of Nantes-Roscoff in partnership with the French CF Society and the French CF Association (Vaincre la Mucoviscidose) sought to adapt the U.S. Cystic Fibrosis Foundation's (US CFF) national initiative, *Accelerating the Rate of Improvement in CF Care*, to improve the quality and length of life for individuals with CF. To launch the Program to Improve Results and Expertise in CF (le Programme d'Amélioration des Résultats et de l'Expertise en Mucoviscidose - PHARE-M), French leaders pursued mentorship and guidance from leaders at the US CFF, the Dartmouth Institute, and clinical care teams at CF centers across the U.S.

Methods:

The following activities enabled the Nantes-Roscoff CF CERD team members and a parent involved with the French CF Association board, quality engineer by training, to gain the leadership and quality improvement knowledge and skills necessary to implement the PHARE-M program: 1) regularly attending national meetings, tracking publications, leveraging existing partnerships; 2) completing two sabbaticals to visit U.S. CF centers; 3) enrolling in academic and professional training courses; and, 4) inviting US CFF and Dartmouth Institute leaders to France to meet key opinion leaders and frontline teams.

Results:

The national CF CERD drafted a call to action to CF centers in two French regions to engage in a pilot phase project, introducing the PHARE-M. The Nantes-Roscoff CF CERD team dedicated a national coordinator, the parent associated with the French CF

Association board, to execute the strategic plan. They adapted and applied the Clinical Microsystems approach and lessons from frontline U.S. CF care teams in preparing a curriculum and adapting material for the French CF teams. They engaged all stakeholders--clinical care teams, individuals with CF and families--in improvement efforts.

Conclusions:

The Nantes-Roscoff CF CERD team adapted critical success factors of the U.S. initiative and continues to partner with U.S. leaders. They are currently seeking alignment with European colleagues to standardize and improve the quality of care for individuals with CF and their families across Europe.

Background

Inter-professional healthcare teams at 124 centers, each accredited by the US CFF, deliver care to approximately 28,000 individuals with CF in the US. With patient consent, medical outcomes and data about the processes of care are captured and reported by way of the US CFF's Patient Registry.[1] Variation in center-level pulmonary and nutrition medical outcomes, first reported in 1999, prompted the US CFF to launch a national improvement initiative, *Accelerating the Rate of Improvement in CF Care,* in 2002.[1,2] The aim of this ongoing initiative is to improve the quality and length of life for individuals with CF through the delivery of exemplary care at all centers. Goals such as individuals with CF and families (i) are full partners with their team of healthcare professionals, (ii) will have normal growth and nutrition, (iii) will receive appropriate therapies to maintain lung function and prevent exacerbations, (iv) are informed to prevent acquisition of respiratory pathogens, (v) screened for complications to enable aggressive management, (vi) supported in making decisions regarding transplantation and advance care, and (vii) will have access to treatments regardless of race, age and ability to pay, further define the initiative's aim.[3]

The initiative encompasses several key elements: a web-based patient registry facilitates data capture and reporting; a quality improvement learning collaborative to teach leadership skills and improvement methods; a benchmarking initiative to identify and enable best practice; discipline-specific mentoring programs to connect healthcare professionals new to CF care with more experienced peers; public reporting of center-level data from the patient registry; publication of evidence-based clinical care practice guidelines; and, a framework for partnering with patients and families to improve care.[1-7]

Progress on each of the initiative elements coupled with the remarkable advances in basic science and therapeutic discovery led the US CFF to report a 10 year (31.3 years to 41.1 years) increased survival for individuals with CF between 2002 and 2012.[2] The US CFF also reported improvement in median values for pulmonary and nutrition outcomes across all centers: median forced expiratory volume in 1 second (FEV_1) percent predicted for individuals with CF aged 6 to 17 years in 2002 was 88.3 and increased to 94.3 in 2012; and body mass index (BMI) percentile for individuals with CF aged 6 to 17 years was 40.8 in 2002 and increased to 51.3 in 2012.[2] Survival and center

level results continue to improve as seen in the 2014 US CFF Center Directors Report (see Figures 1-3).

Over the course of executing the initiative, the US CFF regularly reports updates through national meetings and publications (www.cff.org) and routinely invites community members to participate via a number of opportunities ranging from accessing on-line material in the form of the CF Action Guide (www.clinicalmicrosystems.org) to formal invitations to join a collaborative, serve on a committee, or enroll in a specific program.

In 2004, the French Ministry of Health launched the first national plan for rare diseases aimed at not only invigorating research on rare diseases, but also recognizing the national CF CERD to lead cross-cutting activities. Two CF CERDs were thus certified in 2006, one at the Hospices Civils de Lyon and one bi-site at Nantes and Roscoff including the cardiothoracic transplant unit in Nantes and the rehabilitation center in Roscoff. The Nantes-Roscoff CF CERD action plan featured the following priorities: information and communication system, therapeutic patient education, clinical research (in humanities and social sciences and in transplantation), and a program for care quality improvement (Pougheon Bertrand, Article 2). Between 2008 and 2011, leaders from the Nantes-Roscoff CF CERD and the French CF Association (Vaincre la Mucoviscidose) approached the US CFF and The Dartmouth Institute Microsystems Academy to serve as mentors to enable the French CF community to adapt the U.S. initiative (Pougheon Bertrand, Article 2). This report outlines the specific lessons from the U.S. experience applied in the French CF care system.

Methods

The Nantes-Roscoff CF CERD leader was first introduced to the improvement activities taking place in the U.S. by attending sessions at the annual North American CF Conference (NACFC) and an invitation to participate in the Newborn Screening Special Interest Group. To learn more and to foster a partnership with U.S. leads, he organized sabbaticals to the U.S. for both professionals and the parent involved in the French CF Association board. He also invited U.S. leaders to participate in national strategic meetings, regional conferences, and to visit CF centers in France.

Plenary sessions delivered by US CFF leaders, improvement experts, and parents between 2003 and 2007 provided visibility to the US CFF's initiative and emerging results: *"Accelerating the Improvement of CF Clinical Care"* presented Bruce Marshall, MD

and Gerald O'Connor, PhD, ScD (2003); *"Care Providers and People with CF: Together We Can Make Great Things Happen!"* presented by Paul Batalden, MD, Jim Acton, MD, and Honor Page (2004), and *"Improving Patient Outcomes Using the Tools We Have Now"* presented by Michael Boyle, MD (2007) (www.cff.org). Symposia, workshops, and poster sessions at the NACFC showcased data transparency and public reporting and center-level improvement activities.[8-19]

An invitation extended to international newborn screening leaders to participate in annual special interest group meetings at the NACFC forged personal contacts between U.S. and French physicians. Leveraging these relationships, the leader of the Nantes-Roscoff CF CERD approached US CFF leaders in 2008 to organize a 6-month sabbatical to conduct site visits to US CFF centers engaged in improvement activities, to enroll in the Dartmouth Institute Clinical Microsystems course, and to learn more about Dartmouth's role in supporting national efforts, specifically organizing the Learning and Leadership Collaborative and CF Quality Coaching Program.[20]

In 2011, the French CF Association supported a health care professional (physiotherapist) and the parent affiliated with the association board to return to the U.S. for two months. The physiotherapist and the parent conducted benchmarking site visits to high performing centers and enrolled in the Dartmouth Institute Clinical Microsystems course and the Dartmouth Institute Microsystems Academy Coaching Program.[21]

Between 2008 and 2011, the French CF Association hosted meetings at the Annual French CF Conference with US CFF and Dartmouth Institute leaders to develop and deploy a national improvement initiative. Invited as speakers and advisors, U.S. leaders met with French executive leaders, presented the U.S. activities and results at regional meetings of CF healthcare professionals and patients and families, and met one-on-one with center-level improvement teams during site visits (see Table 1).

Results

Leadership for Improvement

Attending the NACFC sessions and forging relationships with the US CFF and the Dartmouth Institute, French leaders were able to build consensus for a national care improvement strategy. US CFF and Dartmouth leaders shared their insights and lessons of moving from commissioning analyses from the registry to understand variation, to

convening a committee of respected CF clinicians and external improvement experts to draft a strategic plan, and finally operationalizing each of the initiative's key elements.

The Nantes-Roscoff CF CERD team applied these lessons by approaching leaders in the French CF Association and the French CF Society to issue a call to action to provide exemplary care for all individuals with CF. The national CERD formed a standing committee, including the physician lead of the national Patient Therapeutic Education program who was closely aligned with the national CF CERD, to launch the Program to Improve Results and Expertise in CF (*le Programme* Hospitalier d'*Amélioration des Résultats et de l'Expertise en Mucoviscidose - PHARE-M*) (Article 2, Pougheon Bertrand). This committee commissioned registry analyses and drafted a national charter to steer reporting from the Patient Registry mandating linkages between patient outcomes and quality improvement goals to inform and activate the CF community. They hired the parent engaged in these activities as an improvement coordinator to oversee and manage execution of the initiative. This committee continues to direct PHARE-M and members meet regularly with the US CFF and the Dartmouth Institute to seek advice and to learn of new activities being undertaken in the U.S. such as the deployment of a national Patient and Family Experience of Care Survey and development of a dashboard to enable the coproduction of CF care.[22-24]

Direct Observation of Frontline Improvement Teams

On two occasions French leaders seized the opportunity offered by the US CFF to visit U.S. CF centers participating in the quality improvement learning collaborative or noted as high performing centers based on medical outcomes reported from the US CFF registry. Organized with the help of the US CFF and the Dartmouth Institute these visits provided the French leaders with the opportunity to make direct observations of clinical care and interview team members about their experience participating in the national initiative. The Nantes-Roscoff CF CERD leader visited 6 centers in 2008 (Denver, Salt Lake City, Seattle, Chicago, Akron and Madison) and 2 members of the standing committee visited 4 centers in 2011 (Burlington, Akron, Minneapolis and Chicago). During each visit, they shadowed clinicians, participated in team meetings, and reviewed center-level data. These visits gave them an appreciation for the role of local leaders in creating the conditions for personnel to acquire the knowledge and skills for improvement, using data to inform improvement, and the role of external coaching to facilitate learning and achieving center goals.

Two site visits were convened in conjunction with a Learning and Leadership Collaborative session, facilitated by the Dartmouth Institute Microsystems Academy. These opportunities enabled the French team to participate in the didactic learning sessions of the collaborative and the special sessions convened for CF Quality Coaches, facilitating the connection between the Clinical Microsystems model for improvement and its application in a CF care center.

Immersion in Clinical Microsystems

During both sabbaticals to the U.S., members of the French team enrolled in the 10-week Clinical Microsystems course at the Dartmouth Institute. As students they studied the theory of clinical microsystems and participated in the practicum of working with a clinical team to improve care. This experience exposed the team teaching and didactic methods to apply in preparing material for CF teams in France and time to translate and adapt CF specific improvement examples. The team also took advantage of participating in the Dartmouth Institute Microsystem Academy Coaching Program to learn how to create action plans and timelines and offer encouragement to frontline teams.

This deep immersion into the theory and practice of improvement not only facilitated the adaption of material to the French care center context, but also expedited the development of the team's knowledge and skills to lead and teach improvement in the French CF community. The French leadership team continues to participate in the Dartmouth Institutes learning community, most recently participating in a workshop on Standards for Quality Improvement Reporting Excellence (SQUIRE2.0).

Communicating New Ideas and Adapting the U.S. Initiative

The committee convened to spearhead PHARE-M invited US CFF and Dartmouth Institute leaders to France on 4 separate occasions to communicate and spread highlights and lessons from the U.S. initiative. The Senior Vice-President of Clinical Affairs was invited to present at the national French CF Annual Conference in Marne-la-Vallée (March 2008). He communicated lessons from the U.S. initiative and met one-on-one with key opinion leaders at the French CF Association and their board. In March 2012, the Co-Director of the Dartmouth Institute Microsystems Academy was invited to serve as an advisor to the PHARE-M face-to-face meeting in Marseille to provide guidance and expertise on the poster presentation of the 7 Centre teams involved in the first collaborative. The French team continues to rely the Co-Director's expert guidance.

The US CFF's quality improvement program manager was invited by the PHASE-M committee to participate in a 3-day planning retreat at the headquarters of the French CF Association in 2010. This meeting was convened to review center data, define roles, and draft a work plan for the coming year. In 2011, the program manager returned to France to meet one-on-one with teams at two CF centers forming improvement teams to participate in PHARE-M, to speak at the Vaincre la Mucoviscidose Annual Conference in Reims and meet with individuals with CF and families.

Following the sabbaticals and visits from US CFF and Dartmouth Institute leaders, the French team completed the initial adaption of the U.S. initiative. They articulated a vision for improving care; adapted patient centered goals supported by data from their patient registry; published an improvement guide; and engaged CF center teams, including individuals with CF and families, in a learning quality improvement collaborative (Pougheon Bertrand, Article 2).

Discussion

Success Factors and Future Considerations

The Nantes-Roscoff CF CERD, specifically the PHARE-M committee, adapted the five critical success factors of the U.S. initiative to launch a national program: issuing a strategic plan with a call to action, committing as an organization to a culture of improvement, investing in the capacity of professionals to engage in improvement, partnering with individuals with CF and families, integrating improvement into the system of CF care.[2] Within the context of the French health care system, French leaders successfully navigated and partnered with governing bodies to enact appropriate policies and secure resources to embark on improving care for individuals with CF. They prioritized hiring and investing to develop staff to serve as national leaders and coordinators to execute the improvement initiative and regularly convened with U.S. leaders to seek input and advice. They engaged care center teams, individuals with CF and their families in their efforts to improve care, tackling the continuum of CF care including transition from pediatrics to adult care and lung transplantation (cite articles in the French supplement) and enhancing patient education activities. They continue to spread these improvement activities across their network of care centers.

While the French leaders did adapt the critical success factors of the U.S. initiative there remain elements that could be deployed to continue to enrich and accelerate improvement efforts. Pursuing a plan to standardize registry data capture and

reporting to facilitate care management, inform center-level improvement activities, and engage in international comparisons may accelerate timeliness of data for improvement and lead to data transparency and benchmarking opportunities both in France and other European countries with similar health care systems.[25] The French CF leaders could consider establishing discipline-specific mentoring programs to engage professionals new to CF care in both learning more about CF and promoting quality improvement.[7] It may also be worth exploring deployment of a national survey to capture first-hand the patient and family care experience to supplement process and outcomes registry data and to more deeply engage individuals with CF and families in improvement.[22]

Conclusions

The Nantes-Roscoff CF CERD team, with the financial support of the French CF Association Vaincre la Mucoviscidose, successfully adapted the US CFF's initiative to accelerate improvement in CF care by establishing a partnership with U.S. leaders to communicate and exchange strategies and lessons learned; intentionally studying and adapting the Clinical Microsystems approach to quality improvement; and learning directly from the experience of frontline teams in the U.S. They continue to partner with U.S. leaders and are seeking to collaborate with European colleagues to continue to improve care for individuals with CF and their families across Europe.

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Figure 1: Median Predicted Survival Age as reported in the 2014 US CFF Patient Registry Report

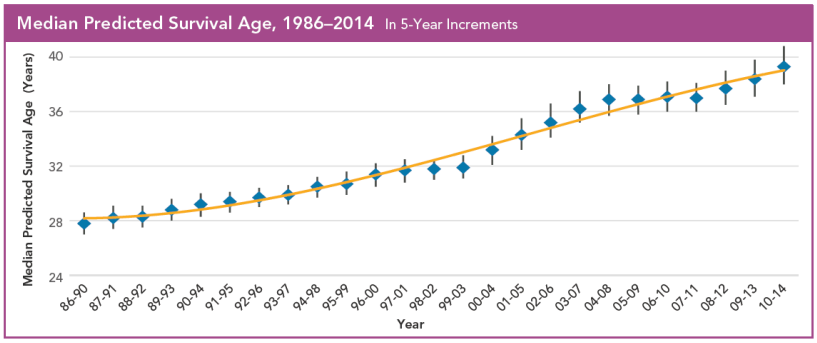

Figure 2: 2004 vs. 2014 US CFF Accredited Center-Level Pulmonary and Nutrition Outcomes for Individuals with CF 6 to 17 Years of Age.

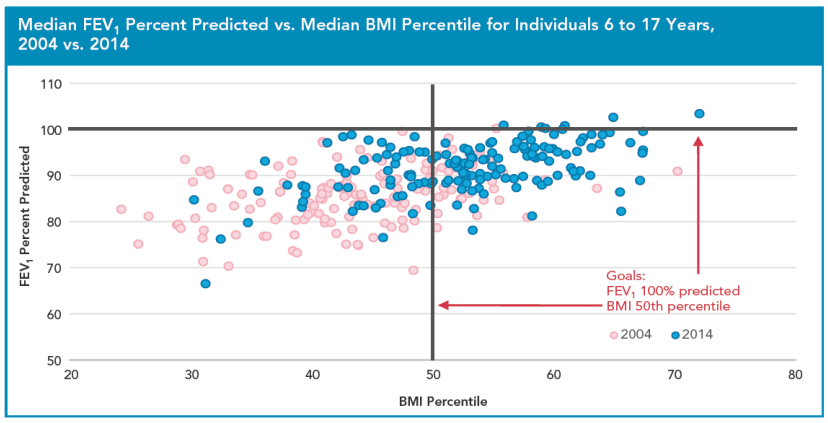

Figure 3: 2004 vs. 2014 US CFF Accredited Center-Level Pulmonary and Nutrition Outcomes for Individuals with CF 18 to 30 Years of Age.

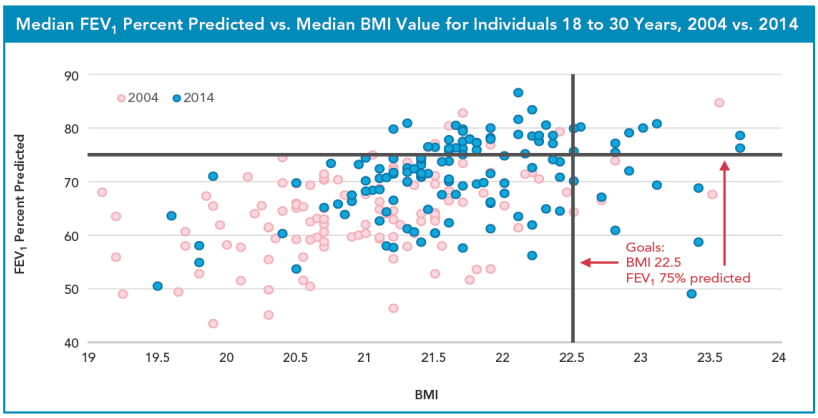

Table 1: US CF Foundation, The Dartmouth Institute, and French CF Leadership Partnership Timeline

Date	Event	Purpose
October 2006	NACFC	French CF leaders attend QI sessions.
June 2007	European CF Society Conference	US CFF and French CF leaders meet to organize a sabbatical for the Nantes-Roscoff CF CERD leader.
February-June 2008	Nantes-Roscoff CF CERD Leader U.S. Sabbatical	Participate in strategic meetings at the US CFF; site visit 5 US CF centers; attend QI training at Dartmouth and US CFF QI learning collaborative.
March 2008	National French CF Meeting	US CFF leader invited to present *Accelerating the Rate of Improvement in CF Care Initiative*.
September 2010	National French CF Meeting	Dartmouth leader presents US CFF Initiative progress; agrees to collaborate to launch PHARE-M.
October 2010	NACFC	Dartmouth and French leaders agree to support a French QI team in formal QI training at Dartmouth, adaptation of material, and participation in a US CFF QI learning collaborative.
March 2011	National French CF Meeting	Dartmouth leader presents Initiative progress; site visits 2 French CF centers; reviews PHARE-M progress.
April-September 2011	French QI team U.S. Sabbatical	Site visit 4 US CF Centers; attend QI training at Dartmouth and US CFF QI learning collaborative; complete adaptation of US QI material.
September 2011-June 2012	PHARE-M Pilot	7 French CF Centers participate; Dartmouth leader attends the collaborative kick-off.
September 2012–June 2013	PHARE-M 2	8 French CF centers participate.
May 2013	National Canadian CF Meeting	Dartmouth and French QI leaders meet to share progress; French team site visits 3 Canadian CF Centers.
September 2013–December 2014	PHARE-M Standardization	French QI curriculum receives national recognition as a professional development program.
January–December	PHARE-M 3	4 French CF centers participate.

2015		
July 2014– June 2015	PHARE-M Research Project	14 French CF centers participate in an evaluation and audit.

Introduction of a Collaborative Quality Improvement Program in the French Cystic Fibrosis Network: the PHARE-M initiative

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Abstract

Introduction

An agreement, signed in 2007 by the 49 French Cystic Fibrosis Centers, included a commitment to participate, within the next five years, in a care quality assessment and improvement program (QIP).

Objective

To roll out in the French Cystic Fibrosis (CF) care network a QIP adapted from the US program for Accelerating Improvement in Cystic Fibrosis Care developed by The Dartmouth Institute Microsystem Academy (TDIMA) and customized by the US CF Foundation between 2002 and 2013.

Method

The French national team at the Nantes-Roscoff CF Center of Expertise was trained at TDIMA and visited US CF centers involved in US Learning and Leadership Collaboratives (LLCs). It introduced the PHARE-M† in France by transposing the Action Guide and material. A PHARE-M LLC1 including seven centers, underwent two external assessments. Adjustments were made, then a PHARE-M LLC2 was rolled out at seven more centers in two regions. On-site coaching was strengthened. The teams' satisfaction was assessed and further adjustments were made. In 2014, the program sought recognition as a continuing education program for healthcare professionals.

Results

Ninety-six trainees including 14 patients/parents from the 14 CFCs volunteered to participate, test and adapt the program during LLC1 and LLC2 sessions. Comparison of patient outcomes collected in the Registry report by CF center, reflection on potential best practices, selection by each team of an improvement theme, implementation of improvement actions, and exchanges between teams fostered the adhesion of the teams. The program strengthened quality of care, interdisciplinary functioning and collaboration with patients/parents at the centers. The satisfaction expressed by the teams increased over time. A post-PHARE-M cycle maintains the focus on continuous quality improvement (CQI). In

† Programme Hospitalier d'Amélioration des Résultats et de l'Expertise en Mucoviscidose – A hospital-based program for improvement of results and expertise in cystic fibrosis care

2015, PHARE-M was recognized as a continuing professional development program in healthcare.

Conclusion

The PHARE-M is a complex intervention in multidisciplinary teams working in a variety of hospital settings. A confluence of factors motivated teams to engage in the program. Involving Patient/Parent in quality improvement (QI) work and developing patient therapeutic education for self-management appeared to be complementary approaches to improve care. Incorporating the program into hospital continuing education insures its sustainability. Transparency of Patient Registry indicators per center published in a brief lapse of time is required to effectively support CQI. The impact of the PHARE-M on patient outcomes after three years is the subject of a research program funded by the French Ministry of Health whose results will be available in 2017.

Keywords: cystic fibrosis, quality improvement program, clinical microsystem, learning and leadership collaborative, rare disease, patient registry

Introduction

The follow-up of cystic fibrosis (CF) patients in specialized care centers has been shown as an independent factor for patients better outcomes and longer survival in patients [15; 16]. In the 21st century Quality Improvement Programs (QIPs) have emerged as new strategies to reduce variability of care and facilitate the implementation of best practices across centers. Following the publication in 2001 of the report entitled *Crossing the Quality Chasm* [17], the US CF Foundation (US CFF) launched a benchmarking study to analyze the differences in patient outcomes across the CF care network. This study highlighted differences in median survival between the 10 best centers and all other centers. The decision was made to design and implement Learning and Leadership Collaboratives (LLCs) with an overarching goal of delivering the best possible care to all patients and improving clinical outcomes [18]. This program was developed by the Dartmouth Institute Microsystem Academy (TDIMA) [19], then adapted, tested and implemented into the CF network starting in 2002 [20].

The cystic fibrosis care center network in France was formalized in 2002, following generalization of systematic newborn screening for CF, to deliver specialized CF care from the diagnosis to adulthood [21]. In 2006, the French National Authority Health published a CF Diagnosis and Treatment Protocol for CF [22]. The French National CF Observatory, modelled on the CF American Patient Registry questionnaire, was established in 1992. Its objective has evolved into taking a comprehensive census of the population [23]. It is now known as the French CF Registry [24] and was certified by the French National Committee of Rare-Diseases Registries in 2007. It is fed into the European CF Registry and contributes to European epidemiologic studies [25]. Within the framework of the first French National Plan for Rare Diseases, the French Ministry of Health designated two CF Centers of Expertise in 2006 to carry out national action plans across the CF care network. The Nantes-Roscoff Center of Expertise action plan featured the following priorities: health information and communication systems, therapeutic patient education, clinical research in the social sciences and transplantation, and a care QIP. An agreement prepared in 2007 and signed by the heads of all CF centers included a commitment to "*participate, within the next five years, in a care quality assessment and improvement program to be offered by the Centers of Expertise in collaboration with the French CF Society, the French Ministry of Health and patient organizations*."

Since 2006, communications at the North American CF Conference and the European CF Conference have reported successful experiences on the part of centers engaged in the US CF LLCs. At a conference in France in 2008 by the French CF patient organization Vaincre la Mucoviscidose and the French CF Society, results of the US LLCs on CF care and

patient outcomes were presented to an assembly of clinicians, care providers, patients and parents. A working group including representatives of the patient organization and of the Nantes-Roscoff EC was formed to reflect on a method for developing and implementing a QIP in France inspired from the US CF QIP. With the support of the CF Foundation, a training for the lead physician of the Nantes-Roscoff EC at The Dartmouth Institute as well as visits to centers engaged in the US CF QIP were organized in 2008. These confirmed the interest of transposing this program to France in order to benefit from this experience and reduce the time taken to develop a QIP in France [[26]]. A team including a parent (an engineer by training) and a physiotherapist was formed at the Nantes-Roscoff Center of Expertise. A presentation by the US QIP coordinator at the Vaincre la Mucoviscidose General Assembly (Reims 2011) was made to inform the French CF community of the importance and feasibility of such a QIP in CF care in France. Both the physiotherapist and the parent went to TDIMA for training and to US centers engaged in LLCs to observe the results achieved following the implementation of a QIP. This was made possible by a grant from the patient organization. Under the supervision of experts from Dartmouth and the CFF, the French team began the translation of the CF Action Guide and educational tools, registered on the Dartmouth CF network's collaborative website, and reflected on the resources needed to implement the program in France. When the program started in France in 2011, some differences between the two countries, such as certain characteristics of the French healthcare system and unique features of the French CF care model and the French cultural context, questioned the success of transposition of the program, the adherence by stakeholders and the achievement of results on the level reported by the United States.

The aim of this article is to report and reflect on the experience of introducing the PHARE-M† QIP in France, between 2011 and 2015, through two annual LLCs leading to the standardization of the final program as a continuing professional development training program on the French hospital continuing education website. We present the factors that gained the teams' adherence, the synergies at work and the adaptations that led to the adoption of the program in the French CF network. Based on our experience, we discuss the elements that we believe to be essential in transposing this CF LLC QIP to the context of another country, since the European CF Society have paved the way for care quality improvement initiatives across the CF care center network in Europe.

Method

This QIP, designed according to the systematic approach described by Nelson, Batalden, and Godfrey [27], is focused on the clinical microsystem, which includes the multidisciplinary care team, patients and their family. The LLC QI format has been adopted by the CF Foundation in 2002 *to support the CF centers' work to reduce the variation in patient outcomes across the US network*. *This adoption included adaptations to the specificities of the care center network, such as local culture, patient population and multidisciplinary staff and the healthcare system in which it existed,* as described by Godfrey and Oliver [20]. The French program is derived from the 2011 US LLC program and benefitted from the experience with and customization of the program in the US CF care network.

French national team responsible for transposing of the US CF LLC

A French national team was formed comprising the lead physician at the Nantes-Roscoff Center of Expertise, his assistant, a parent of an adolescent with CF (an engineer by training), a physiotherapist and the head of information and communication system projects. The physician, physiotherapist and parent had been trained in a quality course at TDIMA, and had visited several CF centers involved in the CF LLCs for years [26]. The physician in charge of the French national therapeutic patient education program (TPE) and director of the pediatric CF center in Nantes, was closely associated with the team and led its testing at her center. This team is hereinafter referred to as the "national team". Due to its composition, the national team included two main features unique to French CF model of care: 1) the CF therapeutic patient education program, validated in 2005 by the French health authorities and structured according to developmental stages in children and needs in terms of management of complication in adults (http://etp.centre-reference-muco-nantes.fr), and 2) respiratory physiotherapy care, delivered to patients at home according to the French National Diagnosis and Treatment Protocol and reimbursed by the French national health insurance system. The national team also strongly emphasized the involvement of patients and parents in the QIP at each center. A recruitment procedure was put in place to identify in the patient caseload at each center individuals with CF or parents of children with CF who were motivated, available, at ease in their relationships with professionals, capable of self-expression in a group, able to communicate via Internet with the team. The patient or parent was enlisted as a full member of the local quality improvement team and their travel expenses were reimbursed by the patient organization Vaincre la Mucoviscidose.

Transposition of the US CF LLC into a first version of the PHARE-M LLC

Training materials were provided free of charge by the US CFF and access to TDIMA's electronic resources was authorized. Resources were developed before the program started in France (September 2011). They included:

- the translation of training materials, including the Action Guide for Accelerating Improvement in Cystic Fibrosis Care [28] under a Dartmouth Director supervision;
- the drafting of a French national report entitled "Registry, a Tool for Quality Improvement" (RTQI), to inform patients and parents and present the usefulness of the French CF Registry to assess improvement on patient outcomes; "The 10 Goals of the PHARE-M" (see Box 1); and an itemization of each goal with the respective roles in a for care improvement partnership to be played by the patients, their family and the healthcare providers;
- the creation of a website dedicated to the PHARE-M [29] containing tools, training materials and updates and serving as a messaging tool dedicated to the teams engaged in the PHARE-M; and
- the selection of a web conference tool for remote training meetings.

Box 1: The 10 Goals of the PHARE-M

1 Parents and patients are full partners of the healthcare team. Each patient/family has a right to clear and understandable information.

2 Each patient, regardless of his or her geographical, social, and cultural circumstances, enjoys effective multidisciplinary care.

3 Each patient/family has a right to therapeutic education to aid in acquiring or strengthening the skills required to best manage life with cystic fibrosis.

4 Patients grow normally and have a normal nutritional status.

5 Respiratory infections and exacerbations thereof are detected as early as possible, and appropriate treatments are started without delay.

6 Physical and sports activities are encouraged from an early age and adapted to each patient throughout his or her life.

7 Suitable measures are put in place and hygiene advice is given to prevent cross-contamination.

8 Complications, including diabetes, are diagnosed and treated early.

9 All patients who progress to a state of severe respiratory failure are informed of their therapeutic alternatives, then either supported in their decision to undergo transplantation or accompanied at the end of life.

10 Post-transplant care aims at sustainable improvement in quality of life and in physical, psychological, and social health.

The Pilot PHARE-M LLC1 (September 2011 – June 2012)

The PHARE-M LLC1 enrolled 7 volunteer centres, including four CF centers from the two national French national Centers of Expertise of Nantes-Roscoff and Lyon, thanks to close professional networking. A multidisciplinary "quality improvement team" was formed at each center included a physician leader, four to five professionals and a parent or a patient. Vaincre la Mucoviscidose agreed to reimburse the travel fees of the teams – including those of the patients/parents – and give each center a grant covering a 0,20 FTEs for a nurse for one year, corresponding to the extra time required for data analysis and teamwork management.

Four Face-to-face LLC meetings were organized. At these meetings, theoretical presentations of the method illustrated with examples drawn from the American teams were alternated with practical exercises by the French center teams. Each team analyzed its patient outcomes and selected a theme for improvement for a target patient population. Patient data was available for each center from the 2009 Patient Registry report by center; however, some indicators presented weaknesses such as body mass index (BMI) being expressed for children as an absolute value and not as a percentile or Z-score. This forced the teams to collect specific data from their patient electronic records. The teams were offered Action Guide tools (satisfaction surveys, activity analysis grids, communication tools, etc.) and took advantage of the opportunity to adapt them to their setting. International experiences published in the literature were presented [30;31] and the teams were reminded of CF care guidelines [32]. Each team identified actions to redesign its processes, in line with its theme for improvement, to be tested according to successive PDSA cycles. The teams' satisfaction and suggestions were recorded at each meeting and an overall score was displayed on the PHARE-M website.

Close collaboration with the TDIMA and the CFF was sustained over the course of LLC1 through:

- the participation of members of the national team, as well as physicians at several pilot centers, in the adult LLC session at the North American conference in Anaheim (October 2011);
- the participation of the Director of TDIMA Clinical Microsystem Group in the third face-to-face meeting to supervise the poster session meeting (PHARE-M LLC1, Marseille, March 2012);

- the trainings for the physiotherapist and the parent on the national team in the TDIMA's "eCoach the Coaches" course at the same time as the PHARE-M LLC1.

Assessments of the pilot PHARE-M LLC1

The PHARE-M being an innovative approach to QI in France, some key stakeholders were dubious as to its applicability in the French CF care network. The head of the Nantes-Roscoff Center of Expertise asked a Nantes Mines Engineering School sociological researcher to perform a first assessment of the program to analyse the factors for its success and barriers to its adoption, and the patient organization asked a consulting a firm to perform a second assessment to inform its decision as to whether to continue to fund the program.

The first assessment took place during LLC1. The assessor participated as an observer during two web meetings and the third Face-to-Face meeting. The assessment included familiarization with PHARE-M documents, interviews with a panel of professionals and patients/parents on the quality improvement teams, an interview with the members of the national team, an interview with the Director of TDIMA, and a visit to one site. All interviews and focus groups were recorded and fully transcribed. The data was exploited (coding, categorization), processed (analysis, validity) and interpreted according to the standard thematic content analysis protocol (Miles & Huberman, 2003 [33]). This was followed by manual grouping and counting within an analysis framework with the following dimensions: process applicability (terminology, formalization, tools, distance web meetings); incorporation of patients and parents (roles, time spent, barriers); national/regional coordination (roles, nature of support, incorporation mechanisms); process adoption (perceived benefits and costs, working atmosphere, engagement, acquisitions); and impact (operation, working practices, cooperation with the stakeholders). The report was submitted in July 2012 for consideration to adjust the PHARE-M LLC2.

The second assessment was contracted at the end of LLC1 to evaluate the effectiveness of this QI method in France, and to perform a comparative analysis between aims and outcomes achieved (efficiency) and between actions performed and expenses (efficacy). The study methodology included: familiarization with the PHARE-M documents and the literature on CF (French National Diagnosis and Treatment Protocol, French National Registry, etc.); investigations into four engaged CFC sites (Versailles, Lyon pediatric, Reims, and Roscoff) with professionals and patients/parents; telephone interviews with the members of the national team and patients/parents. The report was submitted during the October 2012 meeting of the board of directors of the patient organization, and the decision as to whether to continue funding was voted on in December 2012.

Main adjustments in the PHARE-M LLC2

Following these two assessments, the national team made adjustments to the program, thus further customizing the second version of the PHARE-M (see Box 2). The patient organization continued to fund the travel fees of the teams and the extra-time worked by a referent professional on the team at each center. No funding was allocated to the national team for intensive coaching of the teams at each center.

Box 2: Main adaptations in the PHARE-M LLC2

1 Drafting of a second version of the Action Guide illustrated with examples from the French teams in LLC1 instead of examples borrowed from the American teams;

2 Reduction of certain theoretical presentations in the training materials in favor of more exercises during face-to-face meetings;

3 Updated and revised version of the RTQI with was more systematically offered to patients/parents and professionals, either in its entirety or as separate chapters focusing on the goal chosen by the team at the center;

4 Formalization of the "PHARE-M referent" role on each quality improvement team, for a non-physician professional subsidized by the patient organization;

5 Incentive to enlist a quality engineer from the hospital quality department on the quality improvement team at the center, this professional sometimes becoming the PHARE-M referent;

6 One on-site coaching of the team at each center, offered during a visit by the program coordinator and focusing on mapping the clinic process with the “Shadowing a Patient” method [34]; and

7 Simplification of the PHARE-M website by withdrawing the PHARE-M specific messaging tool for the teams engaged in the PHARE-M as they did not use it in addition to their existing messaging tool.

Inter-regional rollout of the PHARE-M LLC2 (September 2012 – June 2013)

A second PHARE-M LLC session was planned to enroll the centers in the two French inter-regions of Rhône-Alpes-Auvergne and Grand-Ouest belonging to the regional care network of the two CF Centers of Expertise of Nantes-Roscoff and Lyon that could not have been included in the first session.

The teams' satisfaction and suggestions were recorded at every face-to-face meeting and web conference during LLC2. They led to two more adjustments to the training material:

- rearrangement of the content of the third and fourth face-to-face sessions by moving up the benchmarking visit and delaying the poster at the end of the LLC session; and

- strengthening of the link with TPE, underlying the importance of programming time for educational sessions during the clinic visit, focusing on the improvement goal and particular needs of the patient.

The teams also requested that a "post-PHARE-M cycle" be established to maintain a focus on quality improvement and have CFCs continue to exchange experiences after the LLC until they achieved their goal for improvement (two to three years after the training year). This was discussed with the patient organization for purposes of obtaining additional funding to organize an annual CQI meeting at a CF center for benchmarking and sustaining QI work.

Standardization and sustainability of the PHARE-M

The growing difficulty of enlisting new CFCs and the risk of jeopardizing patient organization funding led the national team to conceive of different avenues for perpetuating the PHARE-M and its rollout throughout the CF network.

First, a research project was drawn up in an attempt to respond to the recurrent request for evidence of the PHARE-M's positive impact on patient outcomes. The PHARE-M Performance project was submitted at a call for projects by the French Ministry of Health in February 2012. The project was selected by the Ministry on 5 December 2012 and funded for a three-year study. Its protocol combined a quasi-experimental evaluation of the effectiveness of the program to change patient outcomes over the course of three years with a process evaluation [35]. Following a realistic approach, the latter was designed to understand what works, for whom and under which circumstances (context) [36]. The success of the PHARE-M performance project at this call for projects was seen as a means to give credibility and recognition to the PHARE-M as well as funding to the national team for further interventional research.

Second, systematic efforts were made to incorporate the PHARE-M's into hospital accreditation process. The announcement of certain professional practice evaluation (EPP) actions for improvement and the participation of a hospital quality engineer on the quality improvement team at several centers were actively sought to improve the acceptability of the program in hospitals alongside more traditional certification methods.

Finally, continuing professional development in the field of hospital continuing education, which started in 2013 [37;38;39], offered an opportunity to standardize the PHARE-M into a hospital continuing education program without modifying its content or curriculum except to have it take place during a calendar year (January through December). Recognition by the hospital continuing education authority of the PHARE-M as a CPD program was sought as it was key to further roll-out.

Results

Results of PHARE-M LLC1 & LLC2

Seven centers volunteered to test and propose improvements to the program in the PHARE-M LLC1: four pediatric centers (Lyon, Nantes, Paris Robert Debré, and Versailles), one adult CFC (Lyon), and two pediatric teams at mixed centers (Reims and Roscoff) following up a total of about 1,200 patients out of the 6,500 patients in the Registry in 2011. Seven more centers from the two French inter-regions of Rhône-Alpes-Auvergne and Grand-Ouest engaged in the PHARE-M LLC2: three pediatric centers (Angers, Grenoble, and Rennes), two adult centers (Nantes and Rennes), and two mixed centers (Clermont-Ferrand and Morbihan), to which the adult team at the Roscoff center was added, following up about 800 more patients.

Ninety-six trainees from the 14 CFCs participated in the two annual PHARE-M sessions. More than half of the participants (54%) belonged to the multidisciplinary "core" team and 15% were patients or parents of patients. Healthcare providers on the quality improvement teams represented a total of 75 people, patients/parents represented 15 people, and non-healthcare professionals represented six people. Psychologists and dieticians were particularly strongly enlisted to the quality improvement teams (9/75 (12%) and 7/75 (9.3%) respectively).

Among those 14 centers (out of 45 CF care centers in France), three elected a theme for improvement related to adult care, one chose a theme related to transition to transplantation, one chose a theme related to transition to adult care, and nine chose a theme related to either respiratory or nutritional pediatric care. Four of them worked closely with the Quality Department at their hospital. Companion articles in this supplement present the changes in processes and clinical outcomes achieved in some centers between 2012 and 2015 and the links developed between the program and the general quality process at the hospital [40;41;42]. They show that working in QI has allowed these teams to achieve their goals and even exceed them on various themes of improvement such as FEV1 for adolescents, BMI for children 2 to 12 y.o. or time on the lung transplant waiting list. The statistical analysis of the PHARE-M Performance research project, which will assess the effectiveness of the program to change patient outcomes at centers involved in LLC1 & 2, will be performed on the Registry data from 2011 to year 2015 and results will be available by the end of 2017.

The assessment of the teams' satisfaction showed an increase between LLC1 and LLC2, as expressed at each training meeting and for the LLC overall, reflected in the median of all the participants' scores on a scale from 0 to 10, where 10 represented maximum satisfaction (median score = 7.48) and the LLC2 (median score = 8.16).

The final PHARE-M curriculum is presented in Box 3.

Box 3: PHARE-M Curriculum

Phase	Activity: 44 hours, 32h face-to-face meetings, 8h web conf. **ESE:** expertise and sharing of experience face-to-face meeting **Web Conf.:** remote conference organized via internet **PDSA:** plan-do-study-act
Phase 1: Organization of the quality improvement teams at the centres	**Information meeting on the PHARE-M**
	Organization of the quality improvement teams at the CFCs and enrollment in continuing education
	Web conf.: progress report on the preparatory phase
Phase 2: Analysis of the clinical microsystem	**ESE1: Presentation of the methodology and analysis tools (5Ps) and initialization of the analyses in practice**
	Analysis of the clinical microsystem by the quality improvement team at the CFC
	Web conf.: progress report on the analyses at the CFCs
Phase 3: Planning of the actions for improvement in the clinical microsystem	**ESE2: Presentation of the results of the analyses, selection of the themes for improvement and quantified objectives, examination of the ideas for change and foreshadowing of the actions for improvements (PDSA cycles)**
	Organization of the actions and preparation of the PDSA
	Web conf.: progress report on the definition of the PDSA cycles
Phase 4: Implementation of the actions for improvement according to the PDSA cycles and measurement of the outcomes	**ESE3: Benchmarking visit, incorporation of best practices into the actions for improvement, and review of the schedules for implementation of the PDSA cycles**
	Implementation of the first PDSA cycles and operational measurement indicators
	Web conf.: progress report on the implementation of PDSA cycles
	ESE4: Presentation of the teams' posters and presentations

At the teams' request, two post-PHARE-M cycles were offered in 2014, one pediatric and the other adult, consisting of one meeting per year at a CFC, including a benchmarking visit, an account of the progress and outcomes of the teams' actions, exchanges between the teams, and reminders fundamental aspects of the QIP.

Thirteen teams prepared their poster at the end of the PHARE-M session, and these posters were presented at the 1st CF Francophone Conference (2014). Three posters and their updates after three years were presented at the European CF conference (2012, 2014 and 2015) and the North American CF conference (2012). Videos featuring best practice recommendations concerning respiratory physiotherapy, physical and sports activities were prepared.

Improvement of the patient Registry

The French Registry contains one value in a given year for patient health outcomes and long-term treatments, while patient data are recorded at each clinic visit in the electronic patient record within the hospital information system. The Registry Committee establishes rules to select the clinic visit in a given year from which the FEV1, height and weight values are taken to be transmitted to the Registry.

In 2011-2012, the histograms presenting the median values of the centers remained anonymous in the Patient Registry report by center. The transparency brought in the PHARE-M meetings opened up discussions between the teams, leading them either to focus on the themes of improvement when the centers presented unsatisfactory results compared to national median values, or to question the measurement processes at the center. An on-site quality audit of the data transmitted to the Registry was organized in 2014-2015 pointed to variability in the measurement processes and in the application of the selection rule [43]. Avenues for improvement have been identified to support quality improvement of the data transmitted to the Registry by the centers.

To respond to the requests were made to the Registry team, the body mass index (BMI) for children was presented in Z-score value for LLC2. The lag between the year to which the data refer and the time of publication of the report (approximately two years in 2011) led the teams to supplement the Registry data with more recent data pulled directly from their patient records. The 2015 Patient Registry report has been issued by the end of 2016 and then provide more actual data for the PHARE-M LLC5.

Sociological assessment of PHARE-M introduction

The assessment pointed to themes related to cultural acceptance of the PHARE-M at the time of its introduction:

1) the progressive adherence by the teams at the centers to the different steps of the program, taking into account initial feelings of resistance towards administrative hospital quality processes and the associated system of formalization. Putting patient outcomes at the different centers into perspective sparked interest in the process and clarified its purposes. The rapid consensus reached on the priority theme for improvement and the preparation of the poster were unifying;

2) the successful organization of the PHARE-M project, i.e. at national level (program coordinator and program management) and at local level (quality improvement team). However, on the local level, the specific difficulty and required skills of the “referent” position suggested that the role of the “referent” should not be taken by the physician in the quality

improvement team and that the functioning of the physician leader/referent tandem is essential for the dynamic of the team.

3) the innovation consisting of patient or parent participation on the quality improvement teams, alongside their care providers, and their presence at the national face-to-face meetings as well as several local meetings was well perceived [44].

4) the gains for the functioning of the center teams were identified:

- a "collective enlisting of the team" for a unifying, energizing project for which the team learns to work together on what can be improved, thereby creating a "professional dynamic" in which professionals give new meanings to collective and profession-specific work practices;
- "reflexivity" on practices and relationships with patients/parents;
- a "calling into question" of care processes in front of other teams and transparency of outcomes, which may be sustained in a spirit of humility and desire to improve
- a "chance to speak" for all participants, which was possible in the melting pot of the face-to-face meetings;
- "rationale work" around the tools and processes, which objectivized and formalized practices and established a discourse to patients and parents;
- "dissemination" among the teams regarding quality management and tools;
- a "small-gains approach," which allowed pragmatic actions to be implemented with often limited resources and outcomes to be measured to consolidate practices.

The assessment for the patient organization funding recommendations

The consultant highlighted factors related to the feasibility and satisfaction regarding the PHARE-M training year:

- the 5P diagnosis phase faced challenges of feasibility within the training year with respect to 1) analysis of patient data, as Registry indicators were published with a two-year lag and BMI was expressed as an absolute value and not as a Z-score, and 2) analysis of patient satisfaction, as it took longer than expected for patients and parents to return their responses to the questionnaire*;*
- acceptance of the method was overall good, with the teams affirming that they were able to use the tools effectively and will be able to continue to do so beyond the training;
- team satisfaction was high concerning the consensus choice of a theme for improvement, the ability to comment on how they dealt with their work at sometimes difficult times (departures and reduced team), and the enlisting of the team around a joint project to improve patients' outcomes; and
- implementing the actions at the centers met with several difficulties: the building of a consensus on the choice of priority and feasible actions, for example, therapeutic patient

education, which does not always build a consensus on the teams; the availability of the resources to perform certain actions, for example, dieticians who cannot always be enlisted to abide by reconfigured care processes; cultural differences between teams that acted as obstacles to disseminating potential best practices.

Finally, the consultant assessed the effectiveness of the program (see Box 4) and concluded that PHARE-M mainly impacted care quality by allowing centers to use existing resources and innovative actions to comply with CF care recommendations, and that such an impact on quality of care should improve other aims, including the partnership with families and patients, provided that the patient organization support is strengthened.

Box 4: Training's effectiveness after one year assessed according to four criteria

1) sustainable care improvement: **high,** due to adoption of perpetuated tools or practices;

2) improvement in patient health outcomes: **weak after one year**, except in a limited sample of patients included in the new process of care related to improvement actions;

3) development of professional expertise: **average**, especially when there was a slow start; and

4) development of a partnership with patients/parents and care providers: **limited** to the patients involved in the new process of care.

Clinic visit process redesign

During the on-site coaching visits, the clinic visit process was analyzed at most centers by the program coach coordinator according to patient shadowing and process mapping. Multidisciplinary team (MDT) staff meetings, at which patients' situations and treatment plans were determined, were also analyzed. Observation of the multidisciplinary consultation process enabled identification of seven key steps of an "optimal" process (Figure 1) and description of the tasks corresponding to each step (Table I).

Implementation of the process first of all depends on the configuration of spaces. It also incorporates a therapeutic patient education session into the visit. It is linked to multidisciplinary staff meeting at which team members exchange information and hold discussions to ensure that the patient receives genuinely interdisciplinary care and that essential organizational aims are achieved: i) anticipating the consultations scheduled for the following week and having the professionals confirm their planning for these visits by specifying their aims for the patient; ii) drawing conclusions on the situation of the patients seen in the past week and establishing actions to be coordinated before the next visit by the professional in charge of monitoring them; and iii) preparing the visit report and scheduling the next visit.

Most coaching visits pointed out difficulties in sticking to this optimal process. At several centers, there was not enough time to review the situation of all patients seen the past week; as a solution to this problem patients having had an Annual Review or patients with specific needs were prioritized. It was sometimes difficult to get the entire MDT to meet at the same time. Patient records could not always be displayed during the staff meeting. Time was wasted on sharing data rather than making decisions. Effective meeting skills were developed and actions were taken according to a Professional Practice Evaluation process in order to improve the clinic visit process and the staff meeting.

PHARE-M standardization into a CPD program

The PHARE-M was approved as a multidisciplinary CPD program in 2014, and the 2015 PHARE-M LLC3 could be offered as a CPD program (see Box 4).

Box 4: Features of the PHARE-M CPD program

1 The PHARE-M as a CPD program received the approval of the Medical and Paramedical Independent Scientific Committees and will be re-evaluated prior to the extension of this approval (2021); formalized evaluation of each PHARE-M annual session is the responsibility of the hospital continuous education authority.

2 The training center at the Roscoff Foundation runs the PHARE-M CPD program, and the teams' registration fees provide the national team resources to continue to assess, improve and up-date the program and its website.

3 An annual request for application from the director of the Roscoff Center of Expertise, sent in May, invites and reminds the centers to register for the PHARE-M on a volunteer basis; an information meeting is organized in October to present the program and provide documentation to hospital continuing education directorates and quality departments.

4 The professionals on the team at the centers take administrative steps at their hospital to apply for the multidisciplinary PHARE-M CPD program to register for the next year and earn further CPD credits; the professionals on the CF team who are registered must include a lead physician lead and four to five multidisciplinary professionals.

5 The professionals on the teams at the centers are authorized to be absent from their posts for CPD training meetings, both face-to-face and web meetings, and another professional should replace them in their absence.

6 The professionals on the teams at the centers are reimbursed for their travel fees by hospital continuing education.

7 The patient organization is asked to reimburse the travel fees of the patients/parents and for the professionals unable to register to the PHARE-M CPD program.

8 The patient organization is continuing to fund 0.20 FTEs for the extra-time required for a PHARE-M referent on each team during the training year.

Discussion–Conclusion

The PHARE-M represented a "complex intervention" in clinical microsystems embedded in hospital systems marked by their diversity, their constant evolution, and the current economic pressure on the health care system. The various aspects of the program, essentially putting patient outcomes at the heart of quality improvement efforts and involving patients and parents on the quality improvement teams, led to a rapid consensus on the priority theme for improvement and identification of improvements on the process of care. Barriers linked to cultural differences between the United States and France were overcome by “Frenchifying” the Action Guide and the training material. This went beyond translating them into French, and involved searching for synergies with the quality departments. The PHARE-M contributed to the hospital certification process, and thanks to hospital continuing education reform, it was recognized as a multidisciplinary CPD program.

Limitations of the program roll-out

The pace of the roll-out of the PHARE-M throughout France could be accelerated by identifying sources of leverages. This would require professionals and patient organization representatives to pool their efforts (Box 5).

Factors for success in replicating the US CF LLC program

Developing an understanding of the initial model of improvement…

The 2011 Dartmouth and CF LLC model included involving patient and family on CFC improvement teams, using standardized evidence and practice-based ideas for change, preparing regular CF center progress reports, coaching teams, actively using the Patient Registry and applied measurement, and getting to know patients and families through observation and inquiry skills [20]. The following actions laid the foundations for an in-depth understanding of the method and its effects and dynamics: training the physician leader, the physiotherapist and the parent engineer on the national team at the Dartmouth Institute, giving them the opportunity to closely observe US CFCs with a long history of engagement in LLCs, increasing their awareness and energizing them through participation in several US LLC face-to-face meetings at the annual North American CF Conference, and training the parent to the “Coach the coaches” course. The method cannot be learned in its entirety from books, and the practical experiences of the US centers were enlightening. The supervision of the translation by the Dartmouth Institute and the CFF ensured that the training material

initially conformed to the improvement model. The humility of the national team, who recongnized its inability to understand the whole QI approach in depth through training and visits to centers alone, led them to stick to the US Action Guide and training materials during the French LLC1.

… And then adapting the model to the French context

Inevitably, the first LLC had to face the cultural gap between the US and France. This would have led to a great deal of conflict had the national team not anticipated cultural shock and asked the teams to help adapt the program to the French context. Opening up this opportunity decreased the tensions which arose as much from the program as they did from existing frustrations towards the hospital system: burdensome administrative quality procedures, economic pressure on the teams, inadequate facilities, and insufficient resources in every discipline in the CF team compared to standards of care were some of the issues that made the teams uncomfortable with the program.

The modifications made to LLC2 consisted mainly of replacing examples from US teams with examples from French pilot teams in the Action Guide and simplifying some of the theoretical presentations that the pilot teams had rejected, such as the reminders of QI in industry (e.g., process optimization steps) and statistical measurement techniques (e.g., control limits). On-site coaching was intensified and focused on patient shadowing and process mapping, which appeared to be more relevant and usable for the teams. After three years, as the teams engaged in LLC1 and LLC2 were invited to report their results, measurement became a new priority. This topic was addressed in post PHARE-M cycles while writing for publication was envisaged and SQUIRE guidelines were presented.

Performativity of the process initiated with the PHARE-M

All processes pertaining to care quality are evaluated and judged by the professionals with respect to their performativity[‡], that is to say, their contribution by acts that bring about the reality uttered by this process. "*When the players started to prepare and produce their data and their poster, to exchange and compare experiences, the performative capacity of the PHARE-M was perceived and legitimized. The performativity of the action guide was revealed and rationalized in the eyes of the participants on the teams after a few months, when the results that they had presented and debated highlighted the method's organizing nature".* The salience of the outcomes that are put in perspective, the feeling of having

[‡] The notion of "performativity," borrowed from linguistic pragmatics, shows that the medical and healthcare sciences in particular, in the case examined here, and the sciences in general, are not limited to representing the world: they also make it, cause it, and form it, at least to a certain extent and under certain conditions. In linguistics, an utterance is said to be performative when it establishes that of which it speaks. Extended and adapted to the sciences, this insight allows the classification of situations in which the subject of a methodological work is not merely observed or described, but modified or even called into being.

reinvested in care tasks, and the perception of producing and thinking differently most precisely characterize the program's performance. The medical and healthcare population generally had a negative conception of the quality engineering movement. Its culture is the very opposite of the medical, clinical, and healthcare culture which, from the outset, conceives of quality as something incorporated into individual practice, not something existing outside of individual practice or tied to an organization. PHARE-M partially reconciled these two visions.

On-site coaching

The recommendation concerning the strengthening of on-site coaching was verified to be operative during LLC2, with the establishment of visits by the coach coordinator, which at once allowed process mapping to be performed and organizational problems to be addressed. Team coaching was underlined as the most effective measure to develop the capability for improvement of the multidisciplinary teams at the centers [20]. However, this undertaking is costly and could not be offered to the centers during LLC1, as no specific funding had been obtained from the patient organization. Following the assessment, some funding was offered for LLC2 through a specific grant from the Foundation ildys. This grant acted as an investment in the future development of the PHARE-M as a CPD program supported by the training center at the foundation: on-site coaching could be offered, but not at the level achieved in the US. To compensate for the lack of on-site coaching, it was decided to develop the skills of one member of each CF team, referred to as the PHARE-M referent, and to search for synergy with the hospital quality department.

Synergy between therapeutic patient education and patient/parent involvement in QI

Therapeutic patient education in cystic fibrosis has been developed in French CF care, especially at pediatric centers, as it was recognized by law in 2005 as a right for persons suffering from chronic diseases. In practice, it establishes a lasting alliance between the healthcare team and the patient/parent with a view to developing the latter's autonomy and adaptation skills, adjusting them regularly as their needs evolve, and working to remove obstacles to establishing treatments [45]. On the PHARE-M side, the national team fostered patient and parent involvement as a pre-requisite for participation in the program, integrating them as members in the quality improvement team at their center as members so that they would contribute the user's point of view to QI and potentially co-design care processes [46,47]. This convergence between the two dimensions of patient involvement, in self-care and in the process of care redesign, was innovative in 2011 in France, based on the experience of the national team experience rather than on science.

More specifically, the national team fostered links between care improvement actions and educational interventions during the care process. The participation of the patients/parents on the quality improvement teams made it possible to ensure that their preferences and experiences were taken into account when new processes were proposed or care was intensified (nutritional care). Furthermore, within the framework of the PHARE-M, therapeutic education actions were strengthened as sources of leverage to improve home care and thus improve patient outcomes. Prioritizing certain health aims led to priority education actions. Reorganizing multidisciplinary clinic visits allowed an educational session to be incorporated into the course of the visit. Sharing of educational tools among the teams participating in the PHARE-M was boosted. A tool to identify and react to pulmonary exacerbations (REACT) was developed by the national TPE working group after the teams identified the variability in the practices of diagnosing and treating pulmonary exacerbations. Despite fears of therapeutic education competing for space in the teams' tight schedules, the PHARE-M strengthened the practice of PTE and the use of educational tools.

Prospects for the roll-out of PHARE-M and a CQI process in CF care in France

As of early 2017, the PHARE-M has been implemented at 23 centers (out of 45) and LLC6 is ongoing with adult teams. The teams' satisfaction is still increasing, with a median score of 9.1 for LLC5, which was a pediatric program. The outcomes of the centers will be made transparent among the professionals and the patient organization board only in the next few months. Public transparency will take more time.

The research program is aimed at assessing the impact of the PHARE-M on patient outcomes after three years, though it may be difficult to establish a causal link to the PHARE-M, given the evolving context in which centers operates and CF treatments are provided, and the bias inherent to recruiting centers that volunteer to participate. The realistic assessment will conduct an in depth examination of "how and why" a stronger impact of the PHARE-M may have been observed at certain centers engaged in PHARE-M [[48]]. Presenting the results of the research program in 2017 and publishing on PHARE-M initiative will definitely increase the visibility of PHARE-M and raise awareness in France on this quality improvement approach.

Six years after the PHARE-M was launched in the CF network in France, half the centers have been trained, and the various stakeholders – professionals, patient organization representatives and hospital quality department members in some hospitals – perceive the strength of this LLC QI approach and wish to participate in it and contribute to rolling it out further. Interest in this approach is growing outside of CF care, for example among hospital quality professionals willing to test patient shadowing in other chronic care departments.

Beyond these short-term contributions, the need for overall reflection to adapt the method to another model of care (translated in a disease specific Action Guide) requires a dedicated task force at an appropriate level of the health system. Experience with the QIP in CF may inspire its application to the care of other chronic diseases, and this article may contribute to its dissemination.

Aknowledgements: We would like to thank Vaincre la Mucoviscidose and the Foundation ildys for their financial support to the PHARE-M program.

Box 5 Next steps to accelerate the pace of the roll-out of the PHARE-M in France

1 Develop the French CF Registry

- Reduce the time taken to produce annual Registry reports;
- Achieve public transparency of the results by center;
- Advance towards an encounter-based national CF database which produces annual Registry reports as well as ongoing (quarterly) results for the monitoring of the QIPs at the centers

2 Strengthen the motivation of the teams to enroll in PHARE-M program

- Report the PHARE-M experience, results and satisfaction during professional conferences and patient organization assemblies;
- Get the CF community leadership, professionals and the patient organization more involved in continuous quality improvement;
- Continue to obtain funding from the patient organization for the extra-time needed for the PHARE-M referent at each center during the training year;
- Validate continuing professional development credits through the PHARE-M;
- Maintain a focus on continuous quality improvement with financial support for post-PHARE-M cycles until other funding is available (see below);
- Develop a convergence between the roll-out of the PHARE-M and other actions to increase the availability of professional resources, access to CF care guidelines translated in French, and tutoring by discipline within the network;

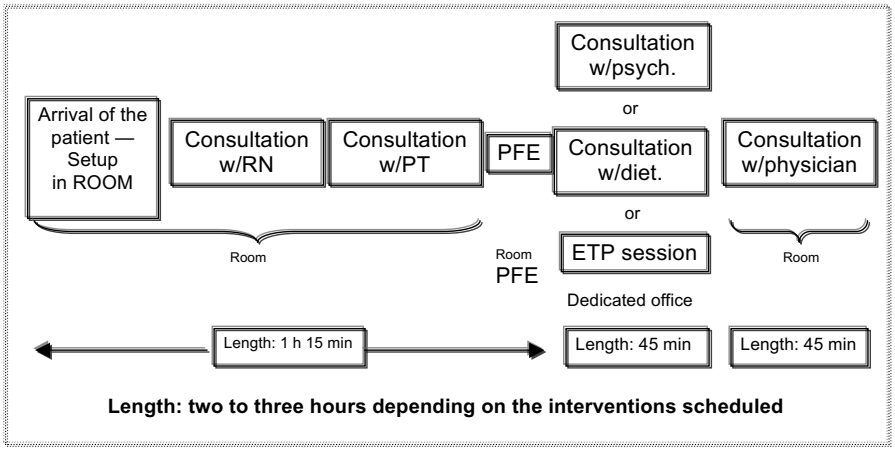
3 Consolidate and develop expertise and resources for the PHARE-M

- Organize a community of PHARE-M referents from the centers for advanced training on measurement, effective meeting skills, quality tools (fishbone diagrams, PDSAs, patient shadowing);
- Develop a culture of publishing QI initiatives according to SQUIRE standards
- Improve and adapt the PHARE-M website to show the various aspects of the program (registration to the CPD program, international research, international community ties, publications, etc…)

4 Build alliances at the hospital and national health system levels

- Continue contributing to the hospital certification process, supporting the hospital quality department through improvement actions, Professional Practice Evaluations, or hospital quality indicators;
- Develop new CPD programs for post PHARE-M cycles focusing on providing reminders of the QI method and tools, benchmarking, measuring and writing for publications;
- Participate in conferences of health authorities or working groups aimed at care quality improvement and patient involvement in healthcare to promote this QI LLC method;

635 **Figure 1 -** Example of multidisciplinary consultation process at a pediatric CFC



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639 **Table I** - Description of the steps of the multidisciplinary consultation process

No.	Step	Who	What	Length (min)	Protocol
1	Installation of the patient	RN	- Setup in the dedicated room - Collection of new elements since the last visit - Verification of the results of examinations performed in the community or at the hospital - Needs for administrative documents (transport passes and certificates) - Reminder of the hygiene rules (wearing a mask) - Validation of the day's clinic visit circuit	5 - 10	Hygiene — CR
2	Consultation w/nurse	RN	- Taking of measurements (weight and height) - Recording of the assessment in the patient's electronic record - Taking stock of the treatments prescribed and taken - Care (implantable device, blood draw, etc.) - Events in the life of the patient to be prepared - Responses to the patient's/parent's questions	20 - 30	Measurement protocol (height and weight) according to the patient's age
3	Respiratory assessment	PT	- Implementation of the hygiene protocol - Taking stock of the physiotherapy practiced in the community and review of instrumental aids - Taking stock of physical and sports activities - Physiotherapy session with sputum collection for sputum culture - Assessment of bronchial congestion - Recording of the assessment in the patient's electronic record	40	
4	PFT (pulmonary function test)		- Measurement of respiratory function - Recording in the patient's electronic record	10	Recommendations of the American Thoracic Society

5	Other scheduled intervention		- Psychological assessment (psychologist), social assessment (social worker), or nutritional assessment (dietician) - Or individual therapeutic education session - Recording of the assessment in the patient's electronic record	30 - 40	
6	Medical consultation	Physician	- Additional examination - Clinical examination - Review of all treatment - Response to the patient's/parent's questions - Referral to the referent professional - Planning of the next visit and need for additional examinations to be performed at the hospital or in the community - Preparation of prescriptions - Recording in the patient's electronic record - Signing of medical certificates	35 - 45	End of the course of consultation to benefit from assessments performed by the other professionals recorded in the patient's electronic record
7	Departure of the patient	Admin. Sec. or RN	- Scheduling of the next appointment - Review of organization for departure (transport, nutritional need, and support) - Verification that the patient has all useful documents - Instructions for events by the next visit - Once the patient leaves the room, disinfection before accommodating the next patient.	30	Disinfection protocol

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A Quality Improvement Program to improve nutritional status of children with Cystic Fibrosis aged 2-12 years old over a 3 year period at CF center Roscoff , Brittany

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ABSTRACT

Introduction

The Cystic Fibrosis (CF) center in Roscoff (Brittany) has been involved in therapeutic education programs (TEP) since 2006 and took part in the pilot phase of the French quality improvement program (QIP) since 2011.

Aim

To improve the nutritional status of children with cystic fibrosis aged 2-12 years old in order to optimize their health status as they enter adolescence.

Method

A multidisciplinary quality team was created in order to select and address a specific health problem among our pediatric population. Following analysis of yearly indicators for our CF center, our team chose to improve quality of care concerning nutritional status of children aged 2-12 years old. Factors influencing efficacy were studied, tools were developed to implement a new nutritional program, results were analyzed on a real-time basis.

Results

Over the 3 year period, all patients from 2 years of age, were monitored with the new follow-up program (2012: N=34; 2014: N=44). Each patient was followed up at every clinic visit, their BMI z-score was calculated to decide their nutritional risk and personalize their follow-up program consequently. Between 1/1/2012 and 31/12/2014, the mean BMI z-score of the open cohort improved from **-0.49** to **-0.22**. Since 2014, focus on nutrition using the newly-adapted program has become routine practice at each follow-up visit. Patients and parents expressed a high level of satisfaction (75% very satisfied).

Conclusion

The follow-up program aimed at improving nutritional status for children aged 2-12 years old was successfully implemented and integrated into routine practice; it was therefore extended to all children with CF (1 month - 18 years) in our center. The relationship among professional and patients and parents was strengthened.

Introduction / Background

The prognosis of Cystic Fibrosis (CF) patients is mainly related to their respiratory status. It is therefore vital to maintain the best possible respiratory function over time and especially during childhood to permit normal lung growth (49).

The direct relationship between nutritional status at the age of 2 years and FEV1 at 6 years is well established among children with CF (50).

In France, children have been followed - up for CF in specialized centers following newborn screening as of 2002. Systematic newborn screening exists in Brittany (region with the highest prevalence rate of CF in France) since 1989. Our patient cohort of 142 patients includes 70 children <18 years old. Children are first seen at our center at the age of one month for diagnosis. Follow-up visits are then programmed regularly with experienced professionals.

Therapeutic patient education (TPE) as defined by WHO in 1985 (51) as "helping patients and their parents to acquire or maintain the competencies they need to manage as well as possible their lives with a chronic disease" is implemented in our CF center since 2006 and programs have been specifically designed for parents of young children (1month-5 years old), for children from 6-10 years old and their parents and also for adolescents (11-16 years old) (Fig.1).

Our CF center Roscoff participated in the pilot phase of the PHARE-M[§] QIP (52) in 2011-2012. Following initial training, review of our 2010 data showed a BMI z-score average of -0,49 for patients aged 2-12 years old. The French CF registry had data for children <18years old but no data for the group 2-12 years old. The national median BMI z-score for < 18 years in 2010 was -0.35, and in our center for the same age group was -0.5 (53). Our multidisciplinary team chose to address the nutritional status of 2-12 year-old CF patients as our results for this age group showed a very large variation in BMI z-score with a mean value of -0.49 (range: -3.5 z-score to +1.8 z- score). This significant variability in our values for this group of children thus left room for improvement. All children aged 2-12 years (34 patients) followed-up at our center were included in the program.

Our aim was to attain an average BMI z-score = 0 by end of 2014. We also expected an impact on FEV1 for patients (> 5 years old) at the end of the program in 12/2014.

[§] *Programme Hospitalier d'Amélioration des Résultats et de l'Expertise en Mucoviscidose – A hospital-based program for improvement of results and expertise in cystic fibrosis care

Method

A quality team comprising a pediatrician, a dietitian, an adult patient, a social worker, a physiotherapist, a nurse coordinator and a study-coordinator was established. The team participated in 4 training sessions organized by the National training-team for CF centers participating in PHARE-M pilot phase. The pediatrician also had the opportunity to participate in a similar Learning and Leadership Collaborative face-to-face meeting in the USA organized by the CF Foundation in Anaheim (2011).

Following these training sessions, our team evaluated nutritional indicators in our center based on annual data and analysis of patient records: BMI z-score, number of clinic and dietitian visits/year, number of stool fat analysis/year, number of nutritional supplements prescribed.

We used a tool, the Ishikawa fishbone cause and effect diagram (54), to determine positive and negative factors influencing nutritional status among our patients. The main factors involving *patient and family* identified by the team were: insufficient knowledge concerning nutrition and link with respiratory status, how to titrate pancreatic enzymes according to fat intake and symptoms, reluctance to do stool sampling and fear of nasogastric (NG) tube feeding. For *professionals* we noted the same reluctance to talk about NG tube feeding, difficulty in obtaining up-to-date information on weight gain or loss between clinic visits and need for more training on patient therapeutic education. The team then reflected on ideas for change and applied the PLAN-DO-STUDY-ACT (PDSA) cycle to design, implement and evaluate new tools (Fig.2):

PDSA1: Creation of an Excel flow chart to follow up each patient over 3 years with calculation of BMI z-score classified in color categories reflecting the “at risk” nutritional status of the child: red for severe risk (BMI z-score < -1,5); orange for moderate risk (-1,5 < BMI z-score < -0,5); yellow for mild risk (-0,5 < BMI z-score < 0); green for no risk (=/>0 BMI z-score).

PDSA2: Creation of a personalized folder for each patient comprising: a simplified explication of the link between nutritional and respiratory status according to Yen et al. [50] publication highlighting the close correlation between a good nutritional status at 2 y and subsequent pulmonary function; a color-coded BMI chart to be updated at every visit using national BMI curves for girls and boys from Ministry of health (55) on which the 4 colors were

added to correlate with the selected at-risk categories on the Excel flow chart (Fig.3-4); a list of ideas for 100-calorie-snacks (Fig.5) illustrated by the dietitian; Individualized weight gain goals with home weighing sheet for orange and red groups

PDSA3: Intensification of our follow up program according to the child's BMI color category including number of clinic visits, dietitian visits, calorie-intake evaluations, stool fat analysis, prescription of nutritional supplements.

Therapeutic patient education (individually adapted) was proposed to all patients and parents according to their needs and age group: for the 0-4 year-old group, the program for parents was finalized in 2011; for the 6-10 year-old group, the program for children and parents was created and implemented during the study period; for the 10-16 year-old group, the program has been implemented since 2010.

Difficult cases in red zone were specifically reviewed at multidisciplinary staff meetings for analysis of individual causal factors (positive and negative) and discussion of the next step to be implemented. For all patients, psycho-social support by both team psychologist and social worker was offered and early discussion concerning NG tube feeding took place systematically with all families.

Satisfaction among patients and parents was assessed using a paper survey given to the patient/parent at a clinic visit (75% responded).

A visual display area (poster) was set-up in the out-patient and in–patient departments so that all the patients, families and professionals could be kept up to date on progress.

Results

All pediatric patients aged 2-12 years old followed up at our centre (34 patients) were enrolled in January 2012. Each child coming to a clinic visit at or after their second birthday was subsequently enrolled. All children were kept in the program for 3 years even after their 12th birthday; therefore the cohort increased to a total of 44 patients by December 2014. One child was excluded after one year as he was accepted on lung and liver transplant list in another centre. All except one patient had pancreatic insufficiency, 44 % were girls, 88% were diagnosed by new-born screening. Eighteen percent were colonized by Pseudomonas aeruginosa, 9% by MRSA (Methicillin resistant staphylococcus aureus) and 9% by Burkholderia cenocepacia or Inquilinus limosus.

Impact on patient health outcomes

The mean BMI z-score of the open cohort of children (34 at the start and 44 at the end of the program) progressed from -0.49 (SD=0,89) in December 2011 (just before starting the program) to -0.22 (SD=0,97) in December 2014. Comparison of our entire pediatric group of patients (0-18 years) with the national median showed a progression in median BMI z-score for our center from -0.5 to -0.26 z-score over the 3 years whereas the national figures progressed from -0.32 to –0.28 z-score (53)

The progression is also shown in the percentage and number of patients in each color category over the 3 years (Fig.7). Moreover, average FEV1 for children > 5 years showed no decline through this 3 year period at 85,5% despite increasing age of the cohort (mean age at the start of the program: 10,5 and 13 at the end of the program). (Fig.7)

Impact on the process of care

This new follow-up program included increased number of clinic visits for patients in red and orange zones. These were difficult to achieve as our center is in a rural area making transportation a limiting factor. For this reason, the program was adjusted in 2013 with fewer clinic and dietitian visits and increased telephone contacts (Fig.8).

Seven patients who were not improving their BMI z-score despite close follow-up – two were stable, five were deteriorating – were screened for other diagnoses related to nutritional status such as diabetes, coeliac disease and helicobacter infection (56,57). Early-stage diabetes was detected and treated for 3 patients, one patient was treated for helicobacter infection and one patient had supplementary

investigations for suspected coeliac disease (not confirmed on biopsy). These screening tests are now part of our routine check-up

Impact on the team

The project was well received by all the professionals involved. For the first 18 months, the pilot team met very regularly to plan and discuss progress, prepare results and presentations. Over the following 18 months, meetings were more focused with often just 2 or 3 members (dietitian, pediatrician and study coordinator). The study coordinator entered all the data from the patient clinic visit on a real time basis so results were available at each meeting.

The multidisciplinary team received training on patient therapeutic education at a national training Institute. The majority had already received training prior to the program, the others received training throughout the program.

Patients in red zone were presented more frequently at the weekly multidisciplinary staff meetings for input by all members. Outcome of discussions was entered into their files.

The quality improvement program was presented once to the hospital management/administration, twice to the multidisciplinary team, and was selected as a subject for examination by the external health authorities audit team as an example of our hospital's improved quality of care.

Other benefits for patients

Patients and parents were very involved in the program and motivated to improve their position on the colored BMI curve. The patient therapeutic education program (6-10y) developed during this period was rapidly applied and was a support to the program.

The process, program and results were displayed in both out-patient and In-patient Departments so all patients and parents had a simple visual summary of the program with update on results.

Satisfaction among patients and parents was assessed using a paper survey given to the patient/parent at a clinic visit (75% responded): results showed that 75 % were very satisfied overall especially concerning individual folder with calorie sheet (70%), information given about the program (66%), and concerning intensified follow-up of children in orange/red zones (48% very satisfied, 46% moderately satisfied).

Inspiration for other CF centers

The presentation of our work at French PHARE-M training sessions and post training sessions allowed us to share our tools, process of care and results with the other teams involved in the QI program. The tools were put on the PHARE-M website and were used by teams in other centers wishing to improve their patient BMI z-score.

We took the opportunity to present our work at 2 ECFS Conferences and were selected in 2012 and 2015 for the poster session. Moreover, the pediatrician was invited to present the program at the European quality management training course held in 2015 and 2016 in the form of video sequences to illustrate the steps of the method: 5 point analysis - selection of global aim – PDSA cycles and results ([58]).

Discussion

CF center Roscoff succeeded in improving the nutritional status of young children with CF thereby also maintaining good respiratory function and thus giving them a better start into adolescent and adult life. We did not achieve our initial target (median : 0 z-score) but did improve the nutritional status over the 3 year period. Comparison of our entire pediatric group of patients (0-18 years) with the national median showed a progression in median BMI z-score for our center from -0.5 to -0.26 z-score over the 3 years whereas the national figures progressed from -0.32 to –0.28 z-score (53). Statistical analysis was not carried out as the cohort was open and numbers insufficient.

Patient education played an important role in the program allowing parents and children to acquire skills and autonomy. Intensification of follow-up according to the “at risk” status of the child was instrumental and systematic screening for coeliac disease, early diabetes and helicobacter infection was implemented to identify individual causes of poor weight gain. The dietitian’s involvement was a key-role as her time was increased in order to see more children at clinic visits, to analyze calorie-intake, carry out education sessions, coordinate with the multidisciplinary team, enter data according to color zone and design new educational tools. The cohesion of the team around the physician leader ensured consistency of actions and was even enhanced throughout the project.

C. McDonald (59) describes a similar nutritional risk screening tool for 2-20 year old patients with CF, based on weight and height velocities using an algorithm to attribute points which then determine risk. Color codes were also used for patient and

parent motivation. On publication no data were available to determine impact on patient outcome.

Our experience with 2-12 year-old patients and their families show that nutritional outcome can be optimized through close follow-up including patient and family education along a "pathway" during childhood (60). In CF, nutritional status is dependent on pancreatic enzyme adherence at home and on learning how to titrate the dose according to fat intake and symptoms (57). Training for the staff was useful to foster the importance of patient education. Pulmonary function of patients older than 5 showed no decline during the 3 year follow-up despite increasing age of the cohort which favors a better prognosis for their adolescence and adult life (a decline in FEV1 of 1,4% per year was described by Welsh et al increasing to 2,6% per year during adolescence) (61).

BMI is not the sole indicator of good nutritional status

The cohort was heterogeneous including for example "tall thin family-pattern" children who had excellent growth in height following a curve at +2 or +3 SD with good weight gain following a median curve, good bone and lean body mass index but had however a BMI in the orange / red zone. The data for these patients explains the wide range of SD in our final results. In fact, only once these children's growth in height flattened off at the end of puberty did we see an improvement in BMI z-score (example Fig.3). This is one of the reasons explaining why we did not attain BMI z-score=0 at the end of the study as 2 "tall-thin" patients stayed in the red zone throughout the study period.

Adjusting doses of pancreatic enzymes

For 24 children receiving relatively high doses of pancreatic enzymes (>10000U/kg) but still in orange or red zones or presenting signs and symptoms of persistent fat-malabsorption, we combined use of 2 different pancreatic enzymes, active at different PH s (5,5 and 7) thus at different zones in the gastrointestinal tract, without increasing the total dose in order to maximize fat absorption. Our hypothesis is that it is probable that not all patients achieve a PH at 7 in the duodenum due to dysfunctional bile salt secretion in CF. For 46 % of patients for whom a mix of the 2 types of pancreatic enzymes was prescribed, we noted a substantial improvement in BMI z-score (average +0.7) within the following 12 months. This impact could lead to a further research study.

Prospects

The program continues after 2014 as new techniques and new-change ideas continue to be implemented.

Performing continuous glucose monitoring led to early intervention with insulin therapy (0,25U/kg of long acting insulin) following diagnosis of significant glucose intolerance or early diabetes (Fig.4). This monitoring was greatly facilitated by use of the FREESTYLE device as children did not have to do any finger-prick controls.

Children's technique for spirometry test was often quite deficient with inconsistent results depending on the child's motivation that day. For this reason a specific module was created in the patient therapeutic program to prepare 5 year old children for the first test with the physiotherapist assisting at the examination to ensure the best possible technique. Subsequently lung function evaluation included LCI (lung clearance index) performed yearly as this test is much less dependent on technique/motivation to obtain realistic results.

Benefits for the quality team

The team followed the framework proposed by PHARE-M; there was good cohesion and implementation as all professionals were kept up to date in the program. The follow-up indicators were updated at each visit on a real-time excel chart which motivated all actors to encourage the best possible results for their patients. The team experienced some difficulties in maintaining regular meetings which were sometimes replaced by smaller more focused discussions.

Conclusion

We have demonstrated that the program is easily integrated into normal clinical practice and has been extended to all pediatric patients (1 month - 18 years old) as of 1/2015. This patient – centered process including individual patient therapeutic education and individual goals helped maintain the dynamic of care which continues up to now.

We wish to thank P LESNE (Adult Patient) for his valuable input.

Fig.1: Patient Education program at Roscoff CF Centre

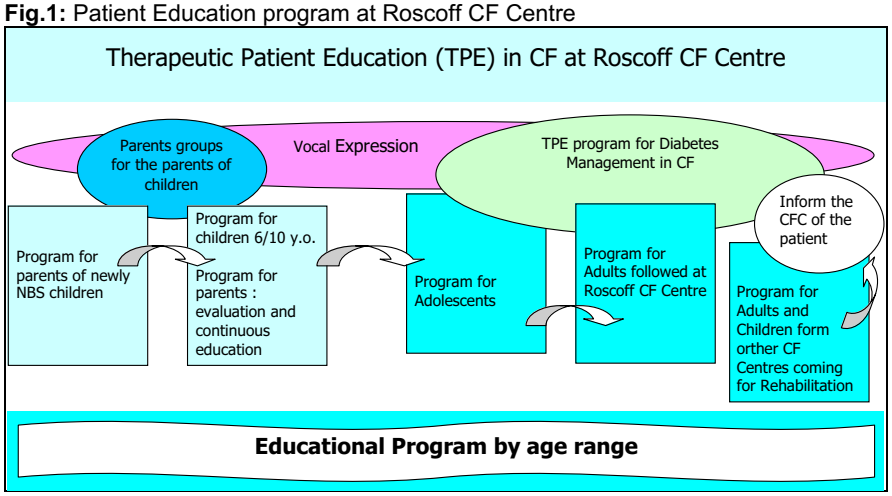


Fig. 2: PDSA Cycles at Roscoff CF centre

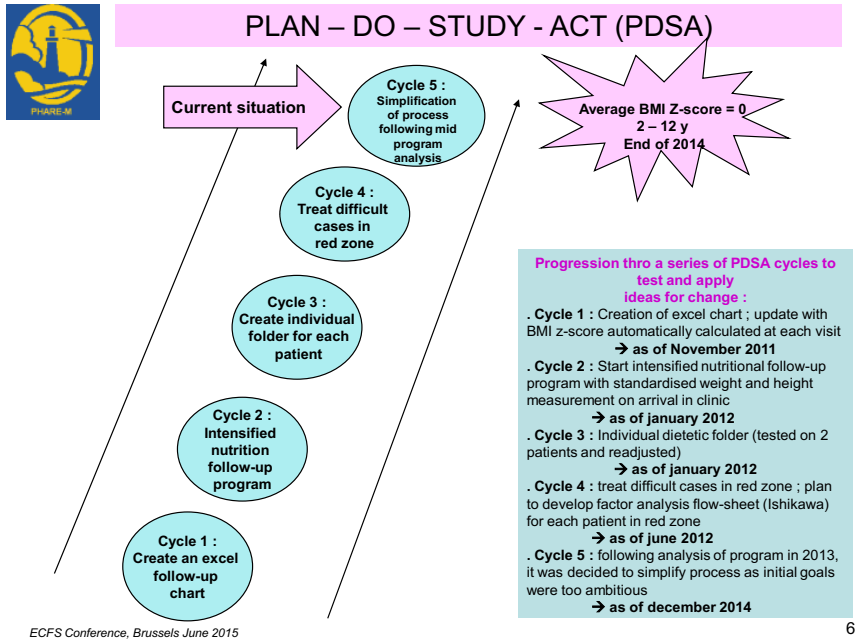
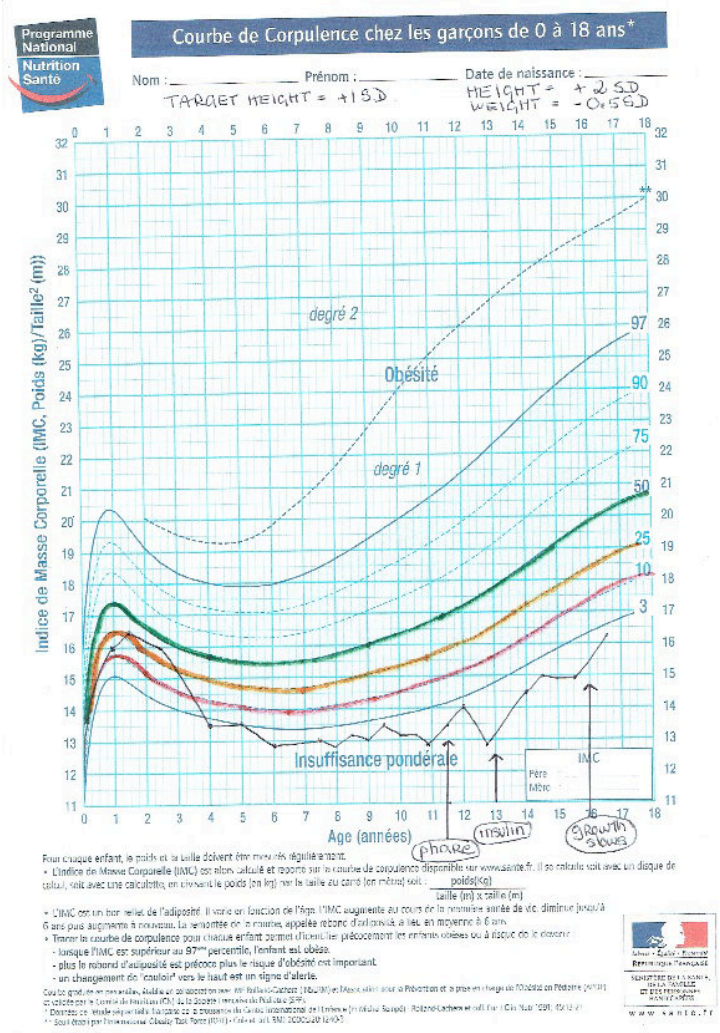
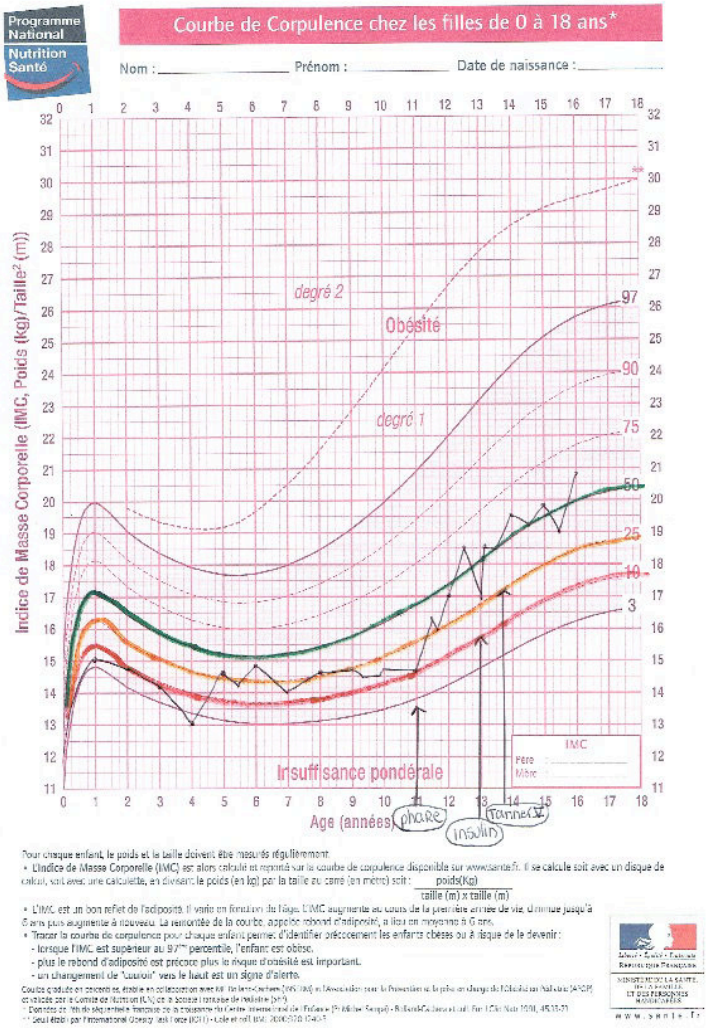


Fig 3-4: Examples of BMI color-zones on Health Ministry BMI curves (respectively for a Boy and for a Girl)





Pour chaque enfant, le poids et la taille doivent être mesurés régulièrement.

• L'indice de Masse Corporelle (IMC) est alors calculé et reporté sur la courbe de corpulence disponible sur www.sante.fr. Il se calcule soit avec un disque de calcul, soit avec une calculette, en divisant le poids (en kg) par la taille au carré (en mètre) soit : $\frac{\text{poids(kg)}}{\text{taille (m)} \times \text{taille (m)}}$

• L'IMC est un bon reflet de l'adiposité. Il varie en fonction de l'âge. L'IMC augmente au cours de la première année de vie, diminue jusqu'à 6 ans puis augmente à nouveau. La remontée de la courbe, appelée rebond d'adiposité, a lieu en moyenne à 6 ans.

• Tracer la courbe de corpulence pour chaque enfant permet d'identifier précocement les enfants obèses ou à risque de le devenir :
- lorsque l'IMC est supérieur au 97ème percentile, l'enfant est obèse,
- plus le rebond d'adiposité est précoce plus le risque d'obésité est important,
- un changement de "couloir" vers le haut est un signe d'alerte.

Courbe établie en concertation avec [illegible] et l'Association pour la Prévention et la prise en charge de l'Obésité en Nutrition Santé (APOP), et validée par le Comité de Nutrition (CN) de la Société Française de Pédiatrie (SFP).

** Seuil établi par l'International Obesity Task Force (IOTF) - Cole et coll. BMJ. 2000;320:1240-3

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Fig. 5: 100-calorie Snacks for children

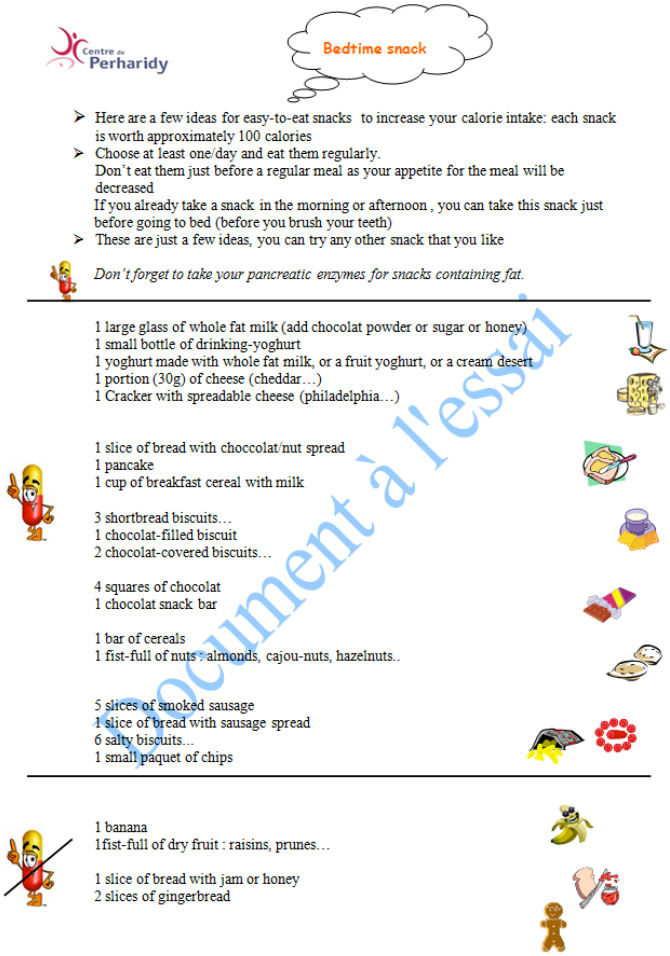
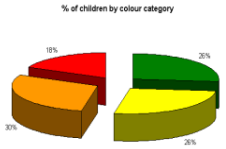


Fig.6: Initial intensified follow-up by color category

THE NEW PROCESS FOR INTENSIFIED FOLLOW UP OF PATIENTS DEPENDS ON COLOUR CATEGORY

NUTRITIONAL STATUS OF CHILDREN (2-12 y) AT START OF PROGRAM



Red : severe malnutrition (> - 1.5 z-score)
Orange : moderate malnutrition (- 1.5 => - 0.5 z-score)
Yellow : mild malnutrition (- 0.5 => - 0 z-score)
Green : normal nutrition status (> o z-score)

	N° of clinic visits/y	N° of dietician visits/y	N° of calorie intake evaluations/y	N° of stool fat samples/y	Prescription of nutritional supplements	Individualised weight gain goals with home weighing	Tube feeding if goal not attained
Green	4	1	1	1	-	-	-
Yellow	4	2	1	1	-	-	-
Orange	5 – 6	5 – 6	2	2	+	+	-
Red	6 – 8	6 - 8	3	2	+	+	+

Fig. 7: Number of patients, mean BMI z-score and FEV1% by color category

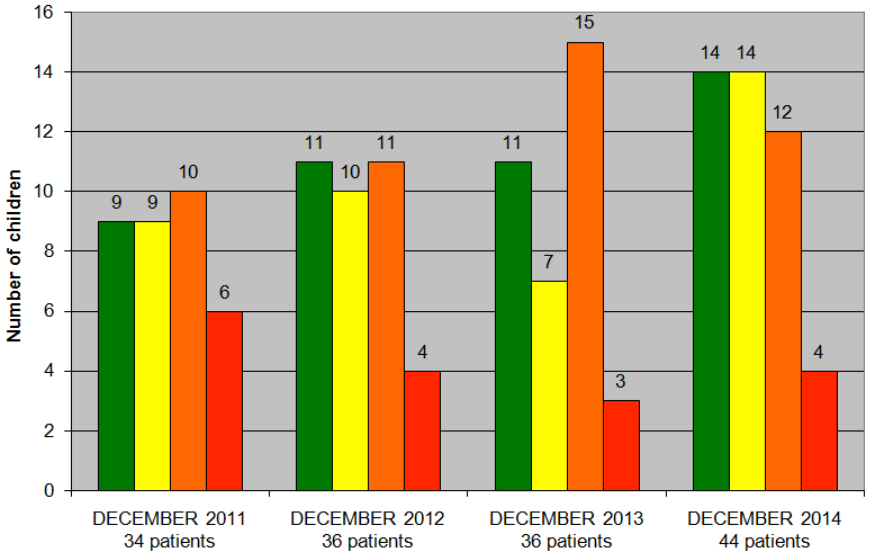


FIG 7 (continued)

December 2011 (34 children)
* Average Z-score : - 0.49
* Average FEV1 : 85.64 %, "mean age of children > 5 years old : 10.5 years (range 5-12.5)"

December 2012 (36 children)
* Average Z-score : - 0.49
* Average FEV1 : 82.71 %

December 2013 (36 children)
* Average Z-score : - 0.45
* Average FEV1 : 88.64 %

December 2014 (44 children)
* Average Z-score : - 0.22
* Average FEV1 : 85.22 %, "mean age of children > 5 years old : 13 years (range 6-15)"

Fig.8: Simplified follow-up process by color category after 2013

Simplified process (2013)

	N° of dietician visits/y	N° of calorie intake evaluations/y	N° of stool fat samples/y	Prescription of nutritional supplements
Green	1 => 1	1 => 1	1 => 1	/
Yellow	2 => 2	1 => 1	1 => 1	/
Orange	5 -- 6 => 4	2 => 2	2 => 2	100% => 100%
Red	6 -- 8 => 5 -- 6	3 => 2 -- 3	2 => 2	100% => 100%

A quality improvement program for adolescents with cystic fibrosis: focus on psychosocial skills.

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Abstract

Introduction: The two pediatric cystic fibrosis centers (CFCs) in Paris (Robert Debré) and Nantes, France, have been developing therapeutic patient education (TPE) programs since 2006 and have been engaged in the pilot phase of the quality improvement program (QIP) named the Hospital Program to Improve Outcomes and Expertise in Cystic Fibrosis (PHARE-M) since 2011.

Objective: To improve the FEV1 of the cohort of adolescents to prepare them for their optimal transition to an adult CFC.

Method: The two CFCs formed a multidisciplinary quality team and used the analysis of causes of insufficient respiratory function taking into account the adolescents' psychosocial factors. At the Nantes CFC, the approach was centered on adolescents' body image and their motivation to take care of themselves by assigning specific aspects of patient follow-up to each professional in the team. At R. Debré, an individual cause-and-effect diagram identified for each patient the medical and psychosocial factors that could account for insufficient respiratory function. Personalized actions were offered to each patient.

Results: in 2014, the median FEV1 (Forced Expiratory Volume in 1 Second) of the adolescent cohort exceeds 90% at the 2 CFCs (Nantes and R. Debré). Between 2011 and 2014 both centers improved their ranking for FEV1% in adolescents in the Registry histograms. At R. Debré, the personalized process allowed to reinforce equality of care, offering to all the opportunity to benefit from TPE sessions and coaching with an adapted physical activity teacher. The psychologist developed a specific tool to support the patient-centered process.

Conclusion: The link between TPE and QIP was strong at our two centers enhancing patient centered care and targeting an optimal transition to an adult program.

Introduction

The prognosis of cystic fibrosis is mainly associated with respiratory status. In the current lack of curative treatment, the objective must be to maintain good respiratory function over time. During adolescence, patients are more likely to see a decrease in their FEV1, the main indicator of their respiratory status [62].

As in all chronic diseases, adolescents with cystic fibrosis have more or less significant difficulties in complying with treatments and finding motivation to take care of themselves [63]. These difficulties may have repercussions on their respiratory status [64].

Adolescents with cystic fibrosis have generally been followed up for many years in cystic fibrosis centers (CFCs), even since birth since newborn screening has been generalized in France in 2002. They have progressively acquired a great deal of knowledge on the disease and the treatments, and are gradually gaining autonomy, both as regards their treatments and their life plans. They transition to an adult program between ages 18-20.

In 2007, the French National Authority for Health (HAS) published recommendations for therapeutic patient education (TPE) [65]. The TPE definition from the World Health Organization in 1998 [66] is: "helping patients acquire or maintain the competencies they need to manage as well as possible their lives with a chronic disease." TPE programs require an authorization issued by the French Regional Health Agencies (ARS), renewed according to a quadrennial evaluation based on the guidelines prepared by the HAS [67].

The two pediatric CFCs in Paris (Robert Debré) and Nantes have developed similar therapeutic education programs allowing children and their parents to acquire and evaluate skills during individual and group sessions.

In 2009, the French Cystic Fibrosis Registry's report by center indicated that the median FEV1 value for adolescents aged 13-17 at the two centers was below the national median value. The two CFCs participated in the pilot phase of the QIP PHARE-M [7] launched in France in 2011. Their common objective was the improvement of the median FEV1 value of their adolescent patients. The 2 teams decided to work on the psychosocial factors that could affect the respiratory status of these patients and on strengthening these patients' psychosocial skills in connection with the actions already undertaken as part of their therapeutic education programs.

This article seeks to assess in 2015 the effects of the actions implemented in the two CFCs during the QIP and particularly their impact on the FEV1 value in their adolescent cohorts.

Methods

The methodology of the PHARE-M QIP consisted of:

- the constitution of a quality team in the CFC: "lead" physician, nurse, psychologist, physiotherapist, dietician, and a patient's parent;
- the participation of the quality teams in 4 training meetings organized by the PHARE-M national team;
- the analysis of the CF center functioning according to the 5Ps assessment: Purpose, Patients, Professionals, Processes, and Patterns;
- the adoption of an improvement theme expressed by a goal on a patient outcome for a population of at-risk patients and a deadline to achieve it;
- the identification of leverage factors and barriers to attain this goal written on a fishbone or cause and effect diagram;
- the definition of PDSA cycles to implement change actions and measure their results on secondary indicators.

Experience at the Nantes CFC

Local context and method

Located on the west coast of France, our CFC follows around one hundred children aged 1 month to 18 years. Most professionals in our multidisciplinary team have been working at the CFC for several years. Furthermore, the head physician is responsible for promoting and developing a national therapeutic education program in cystic fibrosis. In 2006, our CFC established a well-structured therapeutic education program entitled "Becoming competent when growing up with cystic fibrosis" [68]. This program consists of individual therapeutic education sessions, incorporated into the children's periodic clinic visits, and of group sessions. The objectives of the sessions are chosen based on parents' and children's skills assessment so that they may be centered on the needs identified. The skills to be acquired include self-care and psychosocial skills (Figure 1).

A parent of an adolescent and a quality engineer from the quality department of the Nantes University Hospital were included in our PHARE-M quality team. At the first PHARE-M training session, the quality team set up the following goal: "to improve the

median FEV1 value from 78% in 2011 to 85% in 2015 for adolescents aged 15-18".
In addition to the patient outcomes analysis, we conducted 2 satisfaction surveys: one among the CFC's parents and one among the professionals. Our fishbone listed the causes and levers regarding our improvement goal (Figure 2). The team decided to prioritize the focus on self-esteem, body image, relationship between the healthcare providers and the adolescent, their motivation for self-management, early detection of pulmonary exacerbations and their access to leisure and sports activities. We listed avenues for improvement in each of these areas and assigned them to every professional in the multidisciplinary team:

- For the physiotherapist, focus on the patient's attentiveness to their bodily sensations, involving them in their drainage and postural development.
- For the coordinating nurse, focus on the adolescent rather than their parents during the clinic visit, programming the next visit with them and supporting them in achieving their own projects at school, on vacations or in the community.
- For the psychologist, assessing and reinforcing their self-esteem, helping them manage their relationships in their community and the changes inherent to adolescence. The adolescents were also asked to respond to an anxiety/depression/coping questionnaire and the CFQ-R questionnaire.
- For the dietician, assessment of energy expenditure, nutrition regimens and their digestive symptoms.
- For the social worker, socio-economic and cultural assessment in order to facilitate their access to sports and leisure.
- For the art therapist, in connection with the psychologist, improving their self-esteem through creative activities.
- For the physician, a systematic discussion with the adolescent alone, working on the management of their exacerbations, checking their vaccinations, identifying possible issues with tobacco and alcohol and talking about fertility and sexuality.

We determined indicators to be followed up throughout the year in an Excel workbook in the patient record. At the weekly multidisciplinary staff meeting, the professionals scheduled the clinic visit program for the adolescents coming next week according to the needs identified. Every month, the quality team meeting allowed to discuss the indicators and their traceability in the electronic patient record. Annually the quality team analyzed their relevance and the adjustments to be made.

Results

Impact on the patients

The median FEV1 of adolescents aged 15-18 followed up at Nantes CFC and enrolled in the PHARE-M (n = 26) went from 78% in 2011 to 90% in 2015 (Table I). In addition to the above values, our center also improved its national ranking among all French CFCs, as showed in the histograms issued by the Patient Registry, moving from the bottom third to a central position (Fig.4).

Table I: Evolution of the median and mean FEV1% of the cohort of adolescents aged 15-18 at the pediatric CFC of Nantes

	2011 N = 26	**2012 N = 23**	**2013 N = 23**	**2014 N = 23**	**2015 N = 26**
Mean FEV1	78%	85 %	85 %	82 %	86 %
Median FEV1	75%	86%	87%	82%	90%

The adolescents' responses to the questionnaire on quality of life/coping/anxiety/depression showed that quality of life was good for most of them (average score of 150). Half of the patients did not have anxiety/depression (score below 7), 40% were considered to be "uncertain" (score between 8 and 10), and two were "certain" (score above 11) [69].

The adolescents' satisfaction was demonstrated through an interview with professionals, or through their involvement in the illustration of the CFC bulletin with the art therapist's support.

Impact on the team and the process of care

This structured QI project was well received by the team already used to "working together" in therapeutic education group sessions. Professionals expressed their satisfaction in working together on new actions charged with dynamism. Each professional being responsible for a given set of indicators, this led to refine each one's role and refrain from overlapping during the clinic visit, asking the patient/parent the same questions multiple times. Over the 3 years, the secondary indicators were gradually adjusted and became more precise and more numerous (Figure 3). The process of care became standardized and was regularly assessed. Progressively, the new process of care was generalized to all the patients followed at the center. Some issues were raised related to the mesosystem level, such as a lack

of consultation rooms and slots, and work is going on with the administration to solve these difficulties.

Participation of a parent of an adolescent

At first a mother of a 13-year-old child was involved in the program. She participated in our meetings and gave her point of view on the indicators. She stressed that she was speaking in her own name and not on behalf of the parents' group. After a year, she wished to stop her participation and a mother of an 11-year-old adolescent replaced her, who happened to be a quality engineer. Her contribution from both the perspective of a mother and a professional is still going on.

Experience at the Paris Robert Debré CFC

Local context and method

The Paris (Robert Debré) CFC is a pediatric CFC in Île-de-France following around 170 patients. Since newborn screening was established, the Robert Debré CFC has been managing patients diagnosed with CF from the northeast area of Paris. The families are socially and culturally diverse, mirroring the territory in which the CFC operates. They come from 25 different countries, and many of them are in a difficult or even precarious socioeconomic situation.

Therapeutic patient education (TPE) has been developed at the CFC since 2005. The TPE program has been gradually formalized and strengthened, and received an authorization from the Health Regional Agency in 2011. The TPE program develops along the childhood and the adolescence ages, with skills assessment phases alternating with educational sessions. The sessions offered are most often individual sessions, but group sessions are also organized at certain ages: parents of young children (aged 1-3); children themselves near the end of elementary school and their parents.

The 5P analysis showed our center's patterns: a stable and motivated team; a TPE program that operates smoothly; a multicultural population; and patients with a good nutritional status. Our position in the bottom quarter of the histograms for the FEV1% of patients aged 13-17 in the 2011 patient registry report led us to set the goal of improving the FEV1% value for these adolescents by 5% by 2013 and reaching a median FEV1 value ≥ 85% by 2015. In order to achieve this goal, the quality team chose to develop patient-centered actions and to target firstly the adolescents aged

13-17 with an FEV1 below 80%. Thus we used the fishbone tool to analyze, not a system or a process, but the situation of each patient individually in order to identify the personal factors that could negatively influence their respiratory status. These adolescents had individual interviews with different professionals at the CFC. Experiences were then reflected upon at a multidisciplinary staff meeting bringing together physicians, coordinating nurses, physiotherapists, a dietician, a psychologist, and a social worker. Discussions among professionals enabled to identify the factors that could impact the patient's FEV1 value and to build a fishbone diagram displaying the barriers or difficulties in different areas: medical factors, nutrition, physiotherapy and sports, psychological or social factors, TPE… as illustrated on a patient example in Figure 5.

Social and psychological factors are shown of particularly importance in adolescence [70]. The psychologist decided to structure the interviews with adolescents and explore the different areas of their lives more comprehensively. As few tools are available, except the quality-of-life questionnaires, she created an educational assessment tool (Figure 5) centered on adolescent's "feelings" in connection with the various aspects of their life such as family, physiotherapy, sleep, meals, hospital, body image, friends, medications, future. This tool is as a star with a dozen branches, each of them representing one aspect. During a "face-to-face" interview, the adolescent placed an X on each branch corresponding to their level of "well-being" or "dissatisfaction" or "sadness" for each item. Based on this visual appraisal, the interview continued with open-ended questions to clarify the reasons for satisfaction or discontent. The social worker also met with each adolescent individually for a review of their situation and social needs, concerning their family and its resources, cultural background, identification of carers, sports activities, possibility of going on holidays, existence of a personal project or a dream.

The multidisciplinary team imagined for each adolescent personalized strategies based on the difficulties identified. These proposals were discussed with the adolescents, to build with them concrete actions such as relaxation sessions offered to very anxious adolescents, support in schooling, family mediation, help in expressing and fulfilling a dream. Motivational interviews took place to foster adherence to treatments and to help the adolescents develop their own solutions to overcome the difficulties encountered [71]. The main objective of improving the

FEV1 of adolescents aged 13-17 was thus supplemented by other indicators related to the secondary objectives:

- to develop sports activities with the intervention of an APA teacher, quantify the patient's physical activity, encourage the patient, and assess him/her regularly;
- to increase bronchial drainage thanks to instrumental aids;
- to strengthen self-management and psychosocial skills through their participation in the therapeutic patient education (TPE) program;
- to help the patient express and fulfil a dream or a project.

The FEV1% value and the secondary objectives were the indicators followed for the patients during periodic clinic visits.

Results

Impact on the patients

When the PHARE-M program started, 40 adolescents aged 13-17 were followed up at the CFC. Among them, 18 had an FEV1 value below 80%.

For each of them, we made an analysis in a multidisciplinary staff meeting, prepared a cause-and-effect diagram and implemented a personalized action plan. All these patients met with the psychologist for an interview with the "feelings star" tool. This psychosocial self-assessment tool (Figure 6) highlighted certain problems and needs, especially in less-often explored areas such as sleep and body image. It also allowed care adherence difficulties to be addressed. The example of a patient's self-assessment with the feelings star is showed in Figure 5.

At the end of 2015, the main objective of improving pulmonary function was achieved, with a progressive increase in the FEV1% value of the open cohort of patients aged 13-17 (Table II). However it is necessary to notice the variability of patients' evolutions, since some patients improved while others deteriorated during the study. Concomitantly, the rank of our CFC in the national registry improved, moving from the bottom quarter in 2011 to the top quarter in 2015 (Fig.7).

Table II: Evolution of the FEV1% of the cohort aged 13-17 (R. Debré pediatric CFC)

	2011 **N = 40**	**2012** **N = 35**	**2013** **N = 36**	**2014** **N = 39**	**2015** **N = 38**
Mean FEV1	**81.7%**	**84.1%**	**91.7%**	**92%**	**90.5%**
Median FEV1	**84%**	**89%**	**95%**	**97%**	**92%**

For the secondary objectives, the results varied: by the end of 2013, the 18 adolescents with an FEV1 below 80% had benefited from TPE sessions; 14 out of 18 had been trained in the use of instrumental aids for respiratory physiotherapy and 15 out of 18 had made an assessment of their sports activity and coaching with an APA teacher. But only 5 out of 18 patients achieved the objective "fulfilling a dream". The APA teacher's intervention led to an assessment concerning patient satisfaction, implementation of advice given, and possible changes in behavior. This study showed a high satisfaction score among patients (8.1/10); a perceived benefit in terms of a decrease in dyspnea and fatigue; better attention to hydration; and an increase in sports club registrations, which went from 56% to 65%. The use of instrumental aids and the benefit perceived by the patient are still being assessed. Finally, these actions, first initiated for adolescents aged 13-17 with an FEV1 value below 80%, were then extended to this entire age range, regardless of one's FEV1 value.

Impact on the team and the process of care

The PHARE-M was positively received by the team and built a positive team dynamic. The team's cohesion was strengthened. This program empowered each professional and recognized their specific skills. Thanks to the program, the entire team better recognized the psychosocial impact of the disease on the adolescents. The psychologist's and social worker's roles within the team were particularly highlighted, with their increased involvement and participation in the multidisciplinary staff meetings. Finally, the relationship between the team and the patient was often improved, with the team gaining a more comprehensive vision of the patient and their needs, especially in the psychosocial area.

A more structured multidisciplinary analysis of the patient's situation was implemented. The patient-centered approach, already developed within the

framework of the TPE program, was generalized. New tools were developed, such as the "feelings star". As the initial analysis showed that the adolescents were unaware of the social worker's role as a "resource person", a specific session was established for all patients with her, to discuss patient rights at school or at the university and the possible status of disabled worker.

Discussion and prospects

The two CFCs in Paris (Robert Debré) and Nantes chose to improve the FEV1 in adolescents by strengthening their psychosocial skills through a patient-centered approach, in connection with their respective therapeutic education programs. The goals were achieved for both teams in terms of patient outcomes and satisfaction, and in terms of teams' functioning, interdisciplinary work and development of innovative actions. In the two centers, the FEV1% stabilized at a median value of 90% for the population of adolescents after four years. The improvement in the national ranking of both CFCs also suggests an acceleration of their progression compared to the overall national improvement in FEV1% for this age range.

Limitations

It is difficult to attribute these results exclusively to the PHARE-M, as an overall improvement was observed in the respiratory function of CF patients (cf. the annual French national registry data [72; 73]). Furthermore, our cohorts were open, not very numerous and heterogeneous, and various individual evolutions were observed.

Other benefits induced by PHARE-M

Within the framework of the PHARE-M QIP, the participation of a quality engineer from the quality department in Nantes Hospital and the involvement of a parent were key assets for the team. The PHARE-M methodology focuses on the follow-up of indicators, in "real time" and not exclusively based on registry data issued with a lag of one or more year. Assessing the suitability of the indicators lead to readjusting them on a regular basis. Thanks to that, the quality improvement program becomes a process that continues over time [74]. Certain difficulties were noted by the teams regarding information traceability in the patient record, the regularity of the "quality" staff meetings, and the teams' long-term motivation. The annual Quality Improvement experience-sharing days, organized after the training year for the CFCs having participated in PHARE-M, seem essential to maintain a dynamic of continuous quality improvement.

Impact on the quality of care

Over the years, improvements in CF care have been made at our CFCs. The role of the social worker and the psychologist became more important for the adolescents. New professionals were incorporated into our teams, with their specific skills: a physical activity teacher at Robert Debré and an art therapist at Nantes. When incorporated into patient care with specific follow-up indicators, creative activities can improve adherence to the treatment thanks to the increase in adolescents' satisfaction with the team [75]. Among the adolescents followed up at the Nantes CFC, quality of life was most often good and anxiety scores were most often normal; this differed from the results found in the literature [76 ; 77].

Synergy between QIP and patient education

The physicians in charge of the PHARE-M at these two centers were also in charge of TPE programs and members of the GETHEM French national working group for the development of TPE in cystic fibrosis in France. Work and reflection on the adolescents' FEV1 led to identify the skills to be strengthened within this population. Their self-management of care knowledge and skills seemed generally satisfactory; however, their psychosocial skills were often fragile and deserved to be strengthened before the transition to the adult program. At Nantes, this transition is structured at key times, such as discussions about transition since 15 years of age and the "Are you ready?" assessment inspired by the Canadian questionnaire [78], six months to one year before the transitioning process. At Robert Debré, "pre-transition educational assessment" had been used for several years between 16 and 18 years of age leading to educational sessions according to the needs identified. The two teams created a common adolescent assessment approach, based on the existing adult model [79]. This key period for patient follow-up was the subject of a specific quality improvement program in California, United States, in which both teams found similar approaches to determine whether an adolescent is ready to transition [80]. Moreover, some psychological needs were identified in the parents regarding empowering their adolescents, supporting them, managing emotions… It is thus essential to involve them in the transition process. Helping the parents support the adolescent, redefine their role and express their fears and hopes are important objectives in the transition process and may help them for a quiet transition to adulthood with CF [81].

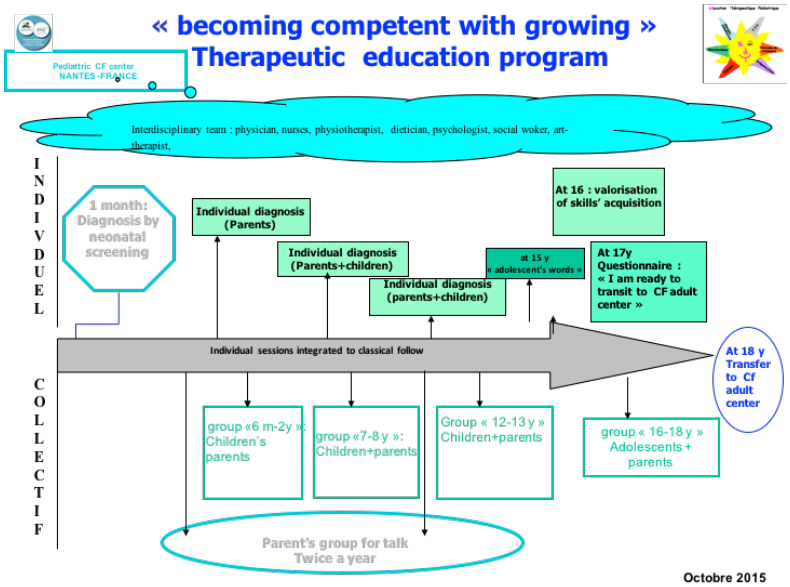
Conclusion

The PHARE-M provides tools and a methodology which structures the QIP towards the optimization of the process of care. The experience of the two CFCs shows that PHARE-M relies on the teams' culture, in this case, the educational programs for the development of the adolescents' skills. Through the PHARE-M, the CFCs combined a systematic approach on processes and an individualized approach centered on each adolescent. This "patient-centered quality process" maximized the QIP's effects and allowed consideration of the patient's needs. The strong involvement of psychologists and social workers in the TPE programs and the PHARE-M QIP strongly benefited to the adolescents and their families.

Acknowledgement

We would like to thank the healthcare teams at the Nantes and Robert Debré pediatric CFCs, as well as the parents and the adolescents for their participation.

331 Fig. 1: "Becoming competent when growing up with cystic fibrosis" program



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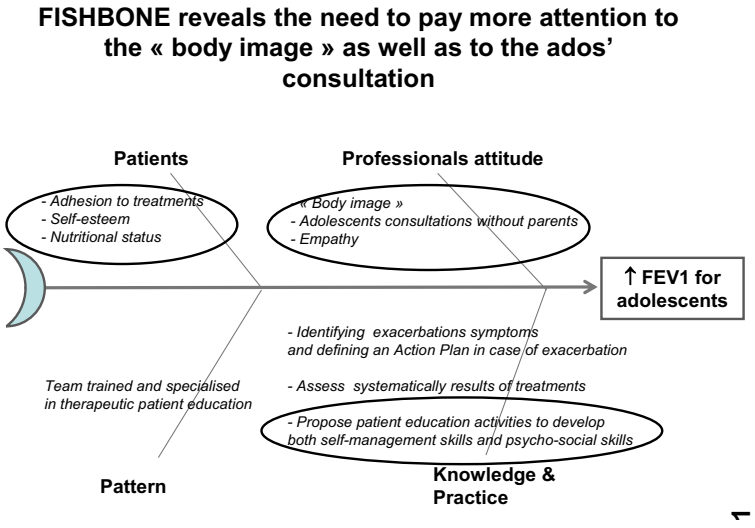
333 Fig. 2: Nantes CFC fishbone diagram



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336 Fig. 3: Nantes CFC 2016 indicators

INDICATORS 2016 Patients born in 1998, 1999, 2000 & 2001		Patients concerned
Physiotherapist & Sport Educator	Contact with physiotherapist at home	All
	Education on auto-drainage	All
	Discussion on sports and physical activities	All
	Contact with personal doctor or physiotherapist about home spirometry	All
Nurse	Questioning about what she/he wants to talk	2001
	Proposition of a session about “How competent you are”	2000
	Realization of the session about “How competent you are”	2000
	Discussion on the transition to the Adult program	2000
	Questionnaire “Are you ready?”	1998
MD	Education on Pulmonary exacerbation and Action Plan	All
	Discussion about Patient’s project of life	All
	Discussion about Genetics – Fertility - Sexuality	All
	Patient vaccination status	All
	Discussion about Tabaco - Alcohol - Drug	All
	Consultation MD alone with the Adolescent	All
Psychologist	Annual Assessment	All
	QoL – Anxiety – Depression questionnaire at inclusion	2001
	QoL – Anxiety – Depression questionnaire before transitioning	1998
Dietician	Annual food survey	All
	Session of dietary counselling	All
	Proposal of a session of dietary assessment skills	2001
	Realization of a session of dietary assessment skills	2001
Social Worker	Education on social rights	1998
	Discussion about Hobbies	All
	Proposal of “a Dream”	All
	Realization of “Your Dream”	All
Art Therapist	Presentation and discussion about inclusion in Art Therapy	All

337

338

339 Fig. 4: Evolution of Nantes FEV1 % ranking for patients aged 13-17

340 • in 2011

27

~~ VEMS (%) médians - patients de 13-17 ans ~~

Figure 26. VEMS (%) médians par centre

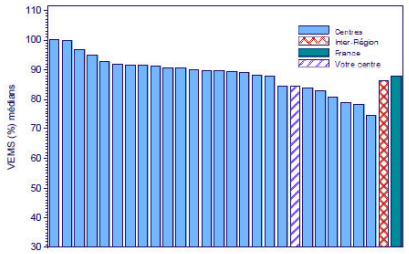
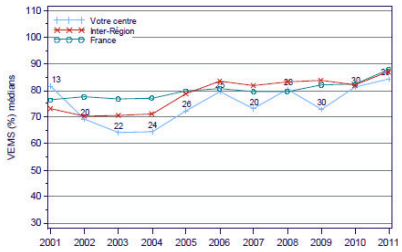


Figure 27. Evolution des VEMS (%) médians entre 2001 et 2011



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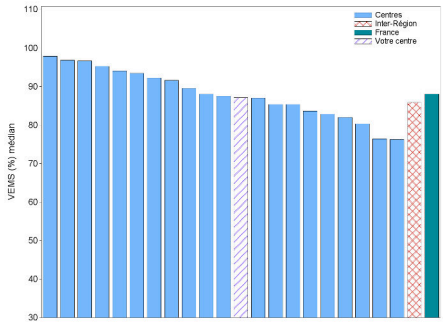

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345 Fig. 5: Cause-and-effect diagram for a patient at the Robert Debré CFC

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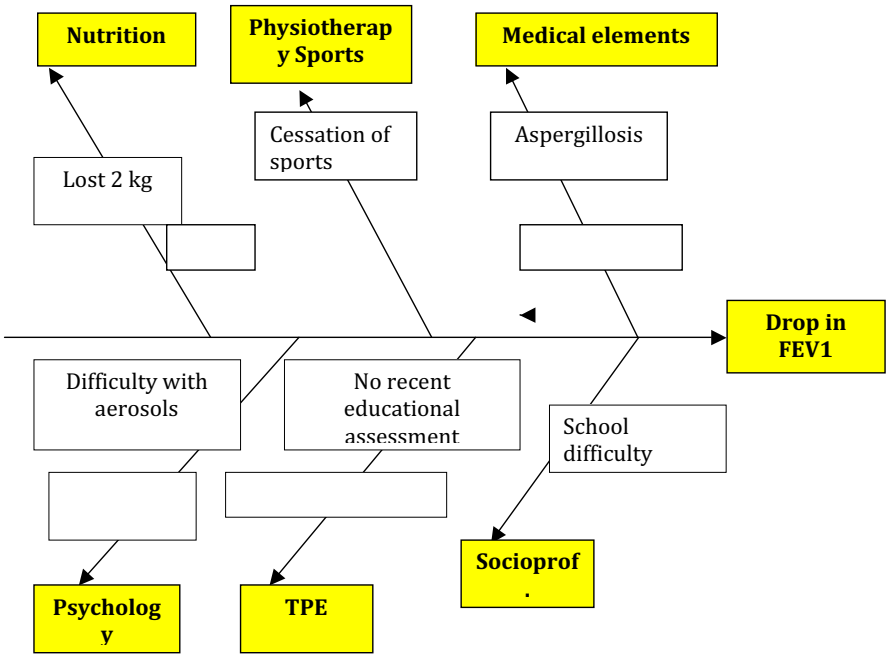

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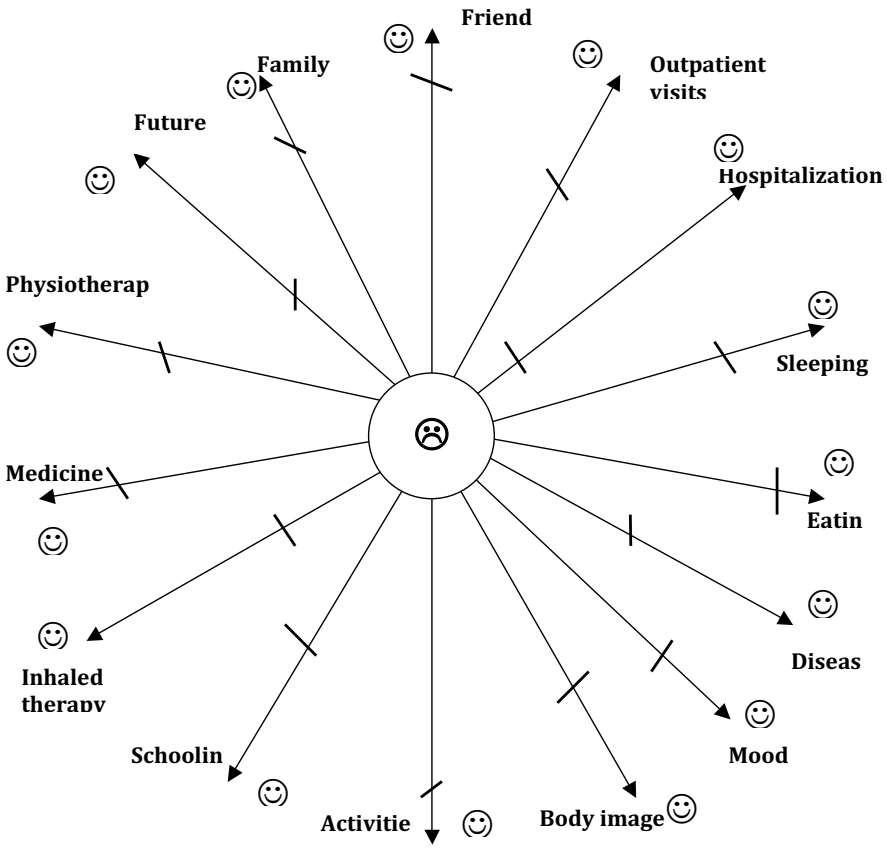
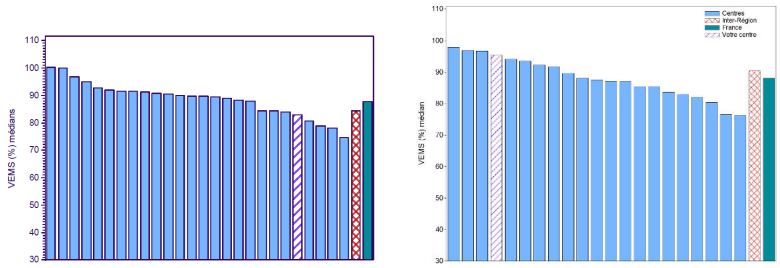
Fig. 6: "Feeling star", psychosocial self-assessment tool from the Robert Debré CFC



355 **Fig.7:** Ranking of Paris R. Debré CFC for adolescents aged 13-17 in 2011 and 2014



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A Quality Improvement Program to Reduce the time on the lung transplant waiting list at the Nantes University Hospital.

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Abstract

Background - In 2010, the time on the lung transplant waiting list in Nantes University Hospital (NUH) was 9.2 months, compared to a French national median of about four months. The NUH transplant unit performs both heart and lung transplantations, which can be seen as competing activities. To fix the problem, the adult Cystic Fibrosis (CF) team decided to engage in the French CF Quality Improvement Program (QIP PHARE-M) in 2012.

Objectives - i) To reduce the time on the lung transplant waiting list at the Nantes Transplant Unit by increasing the number of lung transplants per year twhile maintaining a five-year survival rate above the French national average. ii) To improve the organization of the lung transplant access process and the quality of the waiting time for patients.

Method - A quality controller was involved as the QIP referent to coach the CF quality team, analyze the pre-transplant process, and set up meaningful measures. Benchmarking was performed with other transplant units, and staff discussions were held with the Transplant Team (TT) to assess the outcomes of rejected donor lungs. Negotiations were made with the hospital administration. Plan, Do, Study and Act cycles were used to redesign the pre-transplant assessment in connection with the CF centers (CFC) referring patients to the NUH transplant unit.

Results - i) The flow of patients has been reorganized, decreasing the time spent in surgical intensive care by increasing the number of beds in the intensive care unit, and a chest physician has been recruited ii) The number of organs rejected has been reduced iii) Lung transplant activity has increased to 20-25 transplants per year, and the median waiting time was reduced to 3.5 months for patients transplanted in 2014 and to 1.85 months for patients transplanted in 2015 iv) Added-value activities including education, information, and psychosocial support are now offered to patients during the waiting time.

Conclusion - The QIP PHARE-M, including coaching by a quality-engineer, has helped our adult CF center address its specific lung transplant issues and redesign the lung transplant process for both local patients and patients referred by other CFC.

Introduction

The French national median time on the lung transplant waiting list reported by the French Biomedical Agency (ABM) in 2012 was 4.4 months, while it was 9.2 months for the Nantes University Hospital (NUH). In 2010-2011, the lung transplant activity was at 15-20 transplants per year, including 60% in CF patients,

As the only transplant center in western France, our centre provides a much needed service for transplant particularly for remote areas where traveling to Paris for a transplant is logistically complicated given the time required to arrive at the transplant center during a call. Half of CF patients who have received a lung transplant in Nantes have been referred from other CF centers in France.

The surgery department is unique in that our surgeons at once practice lung transplants, heart transplants, and assisted circulation, as well as scheduled lung and heart surgery. These activities compete with each other, and certain necessary choices are made, not always in favor of lung transplants. On an ethical level, we felt that it was impossible to continue to work with such a discrepancy in our waiting times, including a risk of death on the waiting list greater than the French national average. The survival of our transplant center was at stake. When we joined the QIP PHARE-M, we decided to choose an objective that was original but close to our hearts: to reduce the time on the lung transplant waiting list in Nantes by increasing the number of transplants while maintaining the quality of patient management.

Objectives

The primary objective was to reduce the time on the lung transplant waiting list at the Nantes Transplant Unit by increasing the number of lung transplants per year to achieve the objective of 30 transplants/year, while maintaining the quality of patient management and a five-year survival rate above the French national average (55.7%).

The secondary objective was to improve the organization of the lung transplant access process and the quality of the waiting time for patients, both those at our CFC and those referred by other CFCs, to better prepare them and better meet their needs.

Method

A working group was formed within the CF multidisciplinary team comprising one CF coordinator nurse, one physiotherapist, two psychologists, two pulmonologists, one patient referee who had not undergone a transplant, and one quality controller in charge of coaching the group and helping it analyze pre-transplant processes. This group, called the quality team, worked in accordance with the recommendations and techniques for quality improvement of the QIP PHARE-M. The quality team participated in four face-to-face sessions and six webinars. Secondly, two secretaries were included, their presence being required to manage pre-transplant reviews and the waiting list.

We prepared a fishbone diagram to list the different causes of the problem linked to the main headings: patients, professionals, material resources, other CFCs that refer their patients to us for a transplant, and management processes. An analysis of the transplant process was performed that described the different steps of the process, from pre-transplant to post-transplant follow-up: initial consultation, pre-transplant review, registration on the waiting list, and call for transplant. All these steps were the subject of a team reflection aimed at streamlining the process.

At the same time, information concerning waiting times was disseminated to raise awareness among the different players in the care chain (anesthetists/intensivists, surgeons, pulmonologists, and cardiologists) and negotiations were made with the hospital administration with the help of our Head of Department to alert the medical direction of the situation and ask for more support. The NUH transplant Unit had a reputation for being more demanding than most French centers regarding the quality of grafts accepted. A thesis written by Dr T. Madjer examined the outcomes after six months of recipients of grafts that had been rejected at the NUH because they were deemed to be of poor quality, then accepted at another transplant center.

A satisfaction survey on the experience of the pre-transplant review (PTR) and then the transplant waiting time was sent to 40 patients who had undergone a transplant in the previous three years or who had done a pre-transplant review in the course of these previous three years. The aim was to gather these patients' opinions on the

points that could be optimized to improve the quality of their time on the lung transplant waiting list. The questionnaires were sent by email, with a secondary reminder by post. The results were analyzed anonymously.

Plan, Do, Study and Act cycles were described to structure actions for change, test them, and evaluate their results.

Results

The fishbone diagram identified the points to be improved at the CFC and in the thoracic and cardiovascular surgery department at the Nantes Hospital (Figure 1).

All the players involved in the care journey developed a heightened awareness of the need to reduce the waiting period, accompanied by a renewed motivation to improve quality in the lung transplant process.

The thesis work showed that the FEV1 and survival of patients who had undergone a transplant at Nantes were comparable to those of patients who had undergone a transplant at another center with a graft that had been rejected in Nantes as a poor graft. These results allowed the team to expand its acceptance criteria slightly. At the same time, each donor lung rejected as a "poor graft" was discussed at the weekly transplant staff meeting, allowing contrasting opinions to be expressed. The surgeons agreed to adopt the volume reduction technique, which had not been practiced up to that time, to accept lungs that were morphologically too large and reduce their size (by lobectomy or peripheral resection) to render them morphologically suitable (Ref. 1). As a consequence, the rate of lung proposal refusal for volume missmatch decreased from 26% in 2010 to 21% in 2012.

On the basis of the process described (Figure 2), the hospital administration got involved to make the decision to allocate additional resources, namely:

- The opening of two additional intensive care beds;
- The reorganization of the downstream healthcare network to quickly move patients having undergone a transplant out of surgical intensive care and into pulmonology intensive care, to keep from compromising the schedule of surgeries in the operating room, which requires patients undergoing heart operations to stay 24 hours in the surgical intensive care unit; and

- The acquisition of the machine required for ex vivo lung graft reconditioning, allowing the quality of certain lungs to be improved and allowing them to be transplanted when they do not meet the initial acceptance criteria. In theory, this will allow an increase in the number of usable grafts and thus the number of transplants performed. This technique is in the process of being acquired, and the staff are in the process of being trained (Refs. 2 and 3). However, this technique was not in place at the time of the PHARE program.

Certain patient education actions were carried out with the creation of tools such as the memo card (Figure 3) so that a graft is not lost because of patient unavailability. The memo card reminded the patients of the instructions: to give notice in the event of a change in telephone number, to pay attention to their mobile phone battery, to notify the transplant team if they are hospitalized, and to stay up to date on their vaccinations and anti-HLA Ab monitoring (a lack of recent immunological monitoring necessitates a crossmatch, which can only be organized with a geographically close graft and can thus lead to a transplant being cancelled).

To keep from compromising care quality with an increase in the number of transplants and a corresponding increase in the follow-up load, post-transplant follow-up was reorganized with the other CFCs in the region. Alternating follow-up between our transplant center and the patient's CFC of origin was thus established: it starts one year after the transplant and can be suspended at any time on the opinion of the transplant center if a problem is identified with the relay team. The transplant center remains the center responsible for the patient.

Several actions were undertaken to carry out this alternating post-transplant management.

- Theoretical training was conducted at all the relay centers, followed by immersion training of several days per team at our center.
- Alternating follow-up was progressively established with the CFCs in the region that refer their patients to us for a transplant, after the CFC teams were trained in the unique features of the follow-up of transplant recipients.
- Support was provided with the institution of a time for exchanges in the form of videoconferences one to two times per year with these teams.

- Our team traveled to meet with the main relay team, which strengthened relationships.

The satisfaction survey on the experience of the pre-transplant steps and waiting time was sent to 40 patients who underwent a transplant, and 17 responses were collected. The survey showed good overall patient satisfaction. It essentially revealed a lack of information on social needs. Certain actions were established (Figure 4):

- A tool to identify social needs was created, and
- A possible consultation with the social worker was scheduled.

This survey made us aware of the difference in transplant preparation between the pre-transplant patients followed up at our center and the patients followed up elsewhere and referred for discussion of a transplant. The latter all benefitted from an initial consultation with twice the usual time for exchanges to conduct an initial study of the record and give them information on the transplant process, its challenges, its risks, and the course of the care journey. By contrast, the patients followed up at our center received this information in the course of their consultations. However, the patients felt that this dedicated time to talk about the transplant, often with their relatives, was important. Thus, we established a clearly identified transplant information period for the pre-transplant patients followed up at our center in the form of an additional double-length consultation.

Following a review of our practices, each patient was assigned a referring physician, which had not been the case earlier, when the patients could be seen by different physicians in the course of the pre-transplant consultation, then the PTR week. This assignment of a head physician in charge of presenting each patient's pre-transplant record at the transplant staff meeting and monitoring each patient's subsequent evolution made the journey smoother.

We also instituted a PTR restitution consultation that had not been systematic before this study and that seemed necessary to us.

Since 2012, the time on our lung transplant waiting list has reduced considerably. The median waiting time for transplant recipients went from 9.2 months in 2008-2011

to 5.6 months in 2012-2013, then 3.5 months in 2014 and 1.85 months in 2015 (as of 31/07/2015).

Discussion

When in 2011-2012 we became aware of the major discrepancy between our team's median waiting time and the waiting time at other centers, we found it difficult not to talk about it with the patients on our list whose condition was the most severe. Some of them chose to leave our list to be registered at the Foch (Suresnes) center, which then had a median waiting time of around one month, well below the French national average. This departure of a few patients, combined with an increase in the number of transplants performed associated with the PHARE-M program, reduced the number of patients registered on our waiting list. We went from a list of around 20 patients to six in late 2015. Once the old patients who had been registered for a long time had disappeared from the list (following a transplant, death, or a transfer to another list), our waiting list was self-regulating, with comparable numbers of registrations and transplants per year.

This must be compared to the reduction in the French national median waiting time due to an increase in the number of transplants in France. This was mainly linked to a work conducted on the expansion of the graft acceptance criteria that increased the French national number of transplants from around 180 double lung transplants in 2009-2010 to around 260 in 2012-1013 (Figure 5).

It is important to note that the median figures reported by the French Biomedical Agency are always delayed, while the median waiting times reported for Nantes are real-time figures. Thus, the median waiting time given in 2012 by the Agency concerned the years 2007-2009. The median reported in summer 2015 concerned the years 2010-2013 and also reduced to 2.7 months (Figure 6) (Ref. 4).

It is important to balance these figures with several datas that could have impact our results. First, we saw a decrease of the refusal rate from 2011 (96%) to 2014 (86%) and the main reason for it is a significant decrease of refusal for morphological reason. Howerver, there was a remarkable variation over the past 6 years in the total number of lung proposed to our team: 247 in 2010, 478 in 2011, 532 in 2012 and 355 in 2014. This might be due to the implementation of extended donor criteria at that

period and it clearly can have consequence on the analysis of our rate of refusal. Secondly, the number of patient who underwent lung transplantation under the High Emergency rule increased from 5 patients in 2010 to 8 patients to 2012. It dropped to 4 in 2014 and 5 in 2015. Finally, we saw a variation in the number of patients listed in Nantes (18 in 2010, 10 in 2011 and 19 in 2012).

The PHARE-M process includes patients in the working group. We asked a referent patient who had not undergone a transplant and whose state did not foreseeably require a transplant in the next five years to participate. The working sessions in which she participated were chosen deliberately on the basis of her interest and state of fatigue. This young woman observed that her participation had stirred up certain emotional reactions in line with the reality she faced in advance, despite the efforts made to choose a patient not expected to require a transplant for some time. However, she said that she appreciated this collaboration and found it enriching. Perhaps we should have chosen a patient who has already undergone a transplant, or included another patient, to further enrich the discussion around the experience.

Conclusion

Our team was committed to participate in the PHARE-M improvement program recognizing the need to change in order to improve the service to our patients. With this in mind, our team reduced the median time on the lung transplant waiting list in Nantes. Now we are close to the French national average. The acquisition of the ex vivo lung graft reconditioning technique that is expected to start in early 2016 will position Nantes as a transplant center determined to continue this program with its associated technological innovations. This program also allowed us to review our management processes and qualitatively improve our patients' waiting time and pre-transplant journey. Our program improved tremendously in all these areas and through this publication we would like to encourage other programs o work on similar or more difficult projects.

Figure 1: Fishbone diagram identifying the points that can be improved at the CFC and in the thoracic and cardiovascular surgery department at the Nantes CHU.

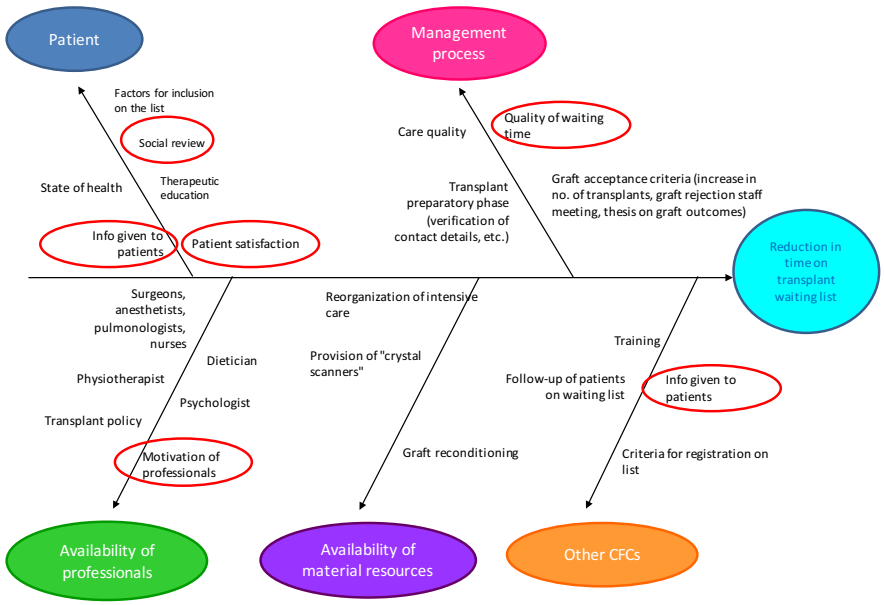

Figure 2: Transplant process from initial consultation to post-transplant follow-up. TTU: Thoracic transplant unit belonging to the thoracic and cardiovascular surgery department (10 beds), managed at once by anesthetists/intensivists, pulmonologists, and cardiologists.

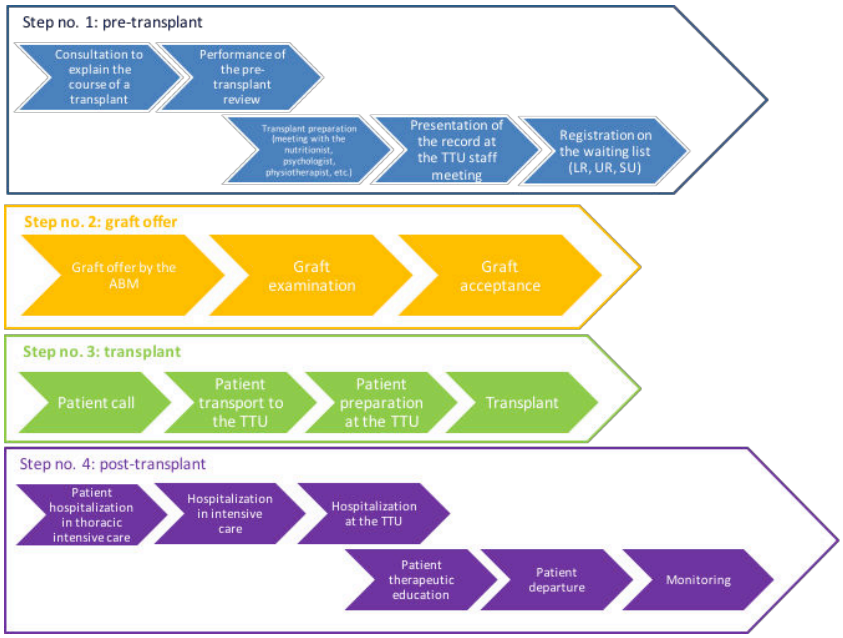

Figure 3: So-called "Memo Card" tool with essential reminders given to the patient at the time of registration on the waiting list.

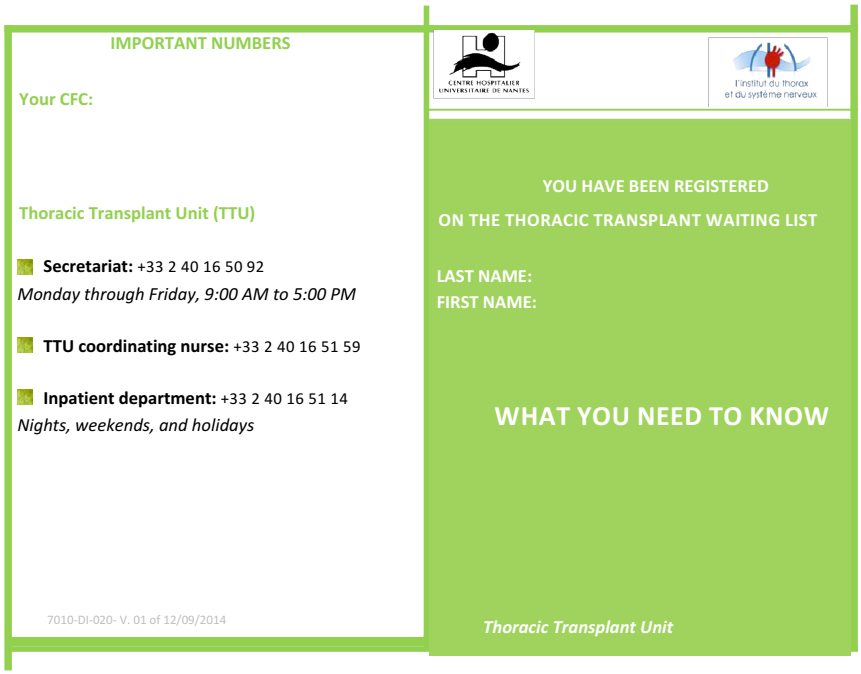

Figure 4: Graphic prepared based on the survey carried out in patients and showing their information needs based on the responses in the 17 questionnaires returned.

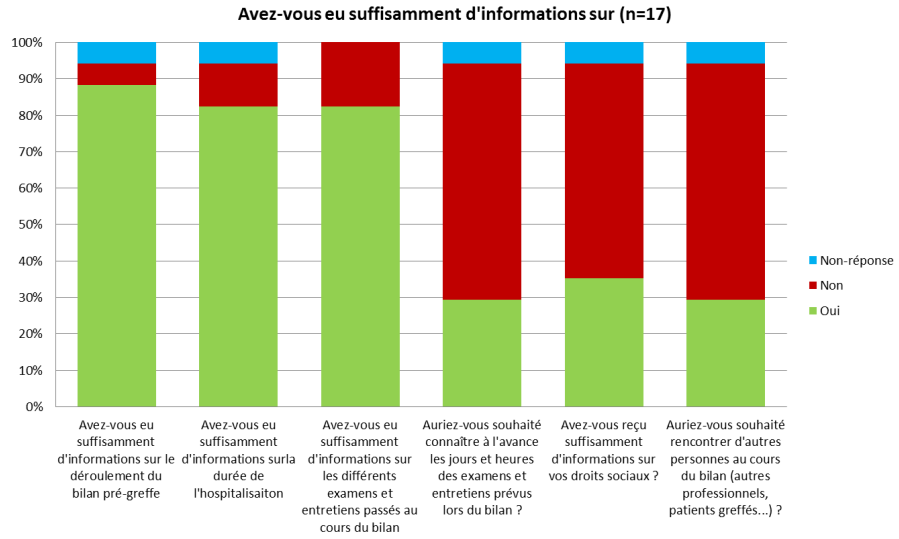

Figure 5: Graphic provided by the French Biomedical Agency on the changes in lung and heart–lung transplant activity in 1997-2014

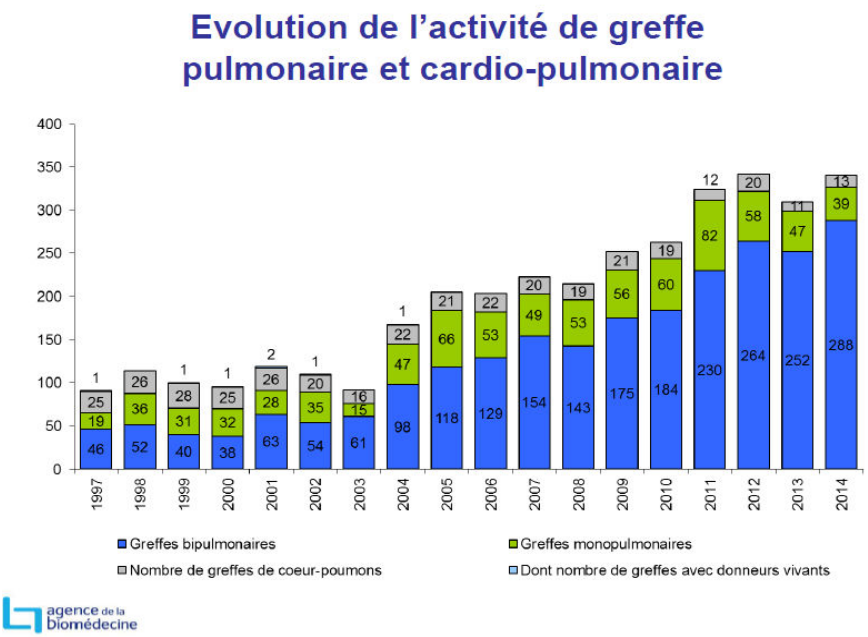

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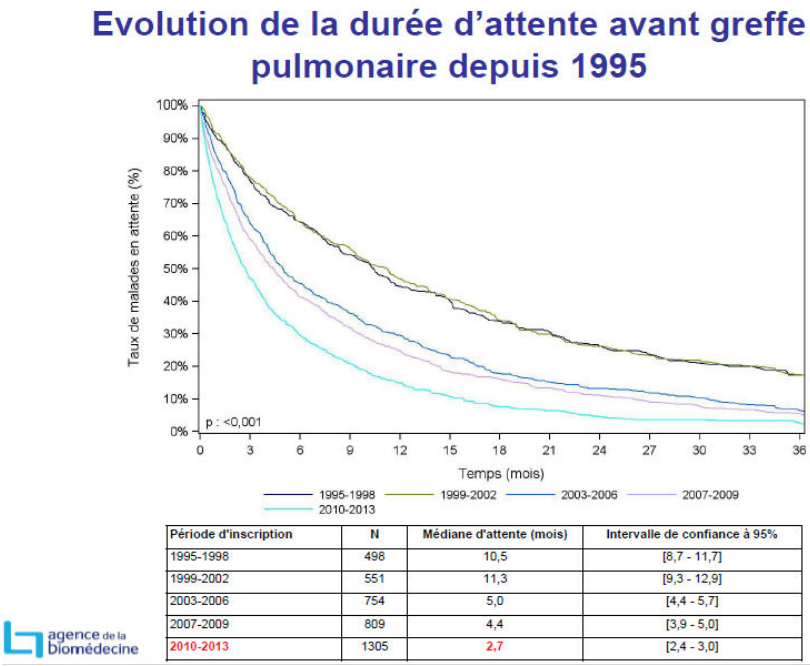
Figure 6: Graphic provided by the French Biomedical Agency on the changes in the waiting time before a lung transplant from 1995 to 2013.



Période d'inscription	N	Médiane d'attente (mois)	Intervalle de confiance à 95%
1995-1998	498	10,5	[8,7 - 11,7]
1999-2002	551	11,3	[9,3 - 12,9]
2003-2006	754	5,0	[4,4 - 5,7]
2007-2009	809	4,4	[3,9 - 5,0]
2010-2013	1305	2,7	[2,4 - 3,0]

	2010	2011	2012	2013	2014	2015
lung proposal n	247	478	532	355	199	281
acceptance n (%)	21 (9)	20 (4)	34 (6)	27 (8)	27 (14)	26 (9)
refusal n (%)	224 (91)	458 (96)	498 (94)	327 (92)	172 (86)	255 (91)
refus morpho n (%)	64 (26)	79 (17)	110 (21)	7 9 (22)	37 (19)	52 (19)
High emergency n	6	5	8	8	4	5
listed patients n	18	10	19	16	21	19

Mis en forme: Anglais (E.U.)

Quality of care in cystic fibrosis: assessment protocol of the French QIP PHARE-M*

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** Programme Hospitalier d'Amélioration des Résultats et de l'Expertise en Mucoviscidose (*Hospital Program to Improve Outcomes and Expertise in Cystic Fibrosis*)*

Abstract

Background

The PHARE-M care quality improvement program, modeled on the US Cystic Fibrosis Quality Improvement Program, was introduced at 14 cystic fibrosis centers (CFCs) in the French Cystic Fibrosis Network between 2011 and 2013. The pilot phase assessments attested the progressive adherence of the teams and improvements in care management. The PHARE-M Performance research project aims at assessing in 2015 the impact of the PHARE-M program on patient health indicators at trained versus untrained centers. It also sought to identify contextual factors that could account for variability in the performance of the PHARE-M among the trained centers.

Method

A mixed methodology combining:

- a quantitative experimental study: a comparison, using a mixed model for repeated data (from 2011 to 2015), of the average changes over time in forced expiratory volume in one second (FEV1) and body mass index (BMI) between two groups of patients included in a closed cohort (non-transplant patients, continuous follow-up at one participating CFC, and a CF-causing mutation), one having benefitted from the PHARE-M program and the other not having done so, and
- a realistic study: a characterization of the impact on care management and an identification of mechanisms through which the PHARE-M intervention improved the team's effectiveness in different CFC contexts; this required modeling the intervention, context, and impact on care management with respect to the criteria of the chronic care model (CCM); this was done using a self-administered questionnaire given to professionals and patients/parents supplemented with focus groups.

Discussion

Although the study population was controlled, it may be difficult to establish a causal relationship between the differences in the changes over time in patient health indicators in the two groups of patients and the PHARE-M intervention as it is often the case in complex interventions rolled out in adaptive environments. The analysis of factors associated with variations in the impact of the PHARE-M at the different trained CFCs required the adoption of instruments validated in other contexts; these could be useful for assessing the performance of other interventions in healthcare practices at CFCs in France.

Keywords: cystic fibrosis; quality improvement program; quantitative study; patient registry; qualitative study;

Background

Cystic fibrosis is the most common rare disease affecting the Caucasian population; it afflicts around 6,500 individuals in France, 29,000 in the United States, and 11,000 in the United Kingdom. It is an autosomal recessive genetic disease caused by mutations in the cystic fibrosis transmembrane conductance regulator (*CFTR*) gene. Among all identified *CFTR* gene mutations, a list of mutations responsible for cystic fibrosis symptoms has been established and is regularly reviewed by the CFTR2 expert group [82]. Cystic fibrosis mainly affects the respiratory and digestive systems. The thick mucus in the bronchi brings about chronic inflammation and repeated infections, leading to chronic respiratory failure, the major cause of death. The majority of patients have pancreatic insufficiency and show poor nutrient absorption, resulting in an at-risk nutritional status associated with a poorer respiratory state [83]. Since the 1960s, the US Cystic Fibrosis Foundation (CFF) has identified multidisciplinary patient management at specialized centers as an essential factor in care improvement; this has led it to establish criteria for the accreditation of cystic fibrosis centers [84]. In the late 1990s, an increase in the number of adults suffering from cystic fibrosis led the CFF to clarify certain criteria for adult centers by stipulating care management by specialized physicians and a specialized team and a formalized process of transition from a pediatric center to an adult program. The accreditation process not only validates centers but also *"fosters continuous improvement efforts within care centers,"* as *"the expectation that each care center have a QI program in place was added to the accreditation and oversight process in 2004."* In the 2000s, following the publication by the US Institute of Medicine, of the report on the Quality Chasm [85], the CFF launched a benchmarking study across the US CFCs, which showed a difference of several years in the median survival age between the ten centers having the best patient outcomes and the other centers (unpublished study). This led the CFF to develop and implement a Quality Improvement Program (QIP) in the form of Learning and Leadership collaboratives [86, 87, 88] with the academic support of The Dartmouth Institute Microsystem Academy (TDIMA). A supplement in BMJ Quality and Safety has been published in May 2014 to present the success of this QI initiative [89].

In 2002, following the generalization of newborn screening in France, the French Ministry of Health designated 49 cystic fibrosis centers (CFCs) [90] and in 2006, the

French National Authority for Health (HAS) published the National Diagnosis and Treatment Protocol (PNDS) in Cystic Fibrosis to establish a framework for multidisciplinary care at CFCs. The French public health insurance guarantees that every CF patient is reimbursed 100% for care and authorized drugs related to cystic fibrosis. In 2006, within the framework of the 1st National Plan for Rare Diseases, two centers of expertise for cystic fibrosis were labelled (CF-CERDs), in order to implement six priorities across the CF Network: care expertise, information systems and epidemiology, quality of care, clinical research, network organization and coordination. The Nantes/Roscoff CF-CERD, consisting of the CFCs at the two hospitals in Nantes and Roscoff as well as the transplant center in Nantes and the rehabilitation center in Roscoff, developed its action plan contributing to 5 out of the 6 priorities, covering themes such as therapeutic patient education (care expertise), quality improvement in care processes, information and communication systems, and clinical research on transplantation and in human and social science. The agreement signed by the heads of all CFCs in 2007 included a commitment to *"participate in a quality assessment and improvement program to be offered by the CF-CERDs in collaboration with the French Cystic Fibrosis Society (SFM) and the patient organizations in the next five years"*.

In 2011, the French national team at the Nantes/Roscoff CF-CERD transposed the PHARE-M quality improvement program from the US CFF QIP model. It was launched in September 2011 with a pilot phase (2011-2012) involving seven volunteer CFCs, which underwent two external assessments, leading to certain adjustments to the initial program. This adjusted version was deployed during a regional expansion phase (2012-2013), including seven more CFCs before its national deployment [91]. The main adjustments consisted in more practical exercises during face-to-face meetings (less theoretical presentations), more on-site coaching to help the quality teams analyze their processes of care, and the designation of a PHARE-M referent in each local team to keep focused on the QI work. These two years are called the “experimental phase”, which involved 14 CFCs.

The two evaluations at the end of the one-year pilot phase showed the progressive adherence of the teams and improvements in care management, but a limited impact on patient health outcomes. They also highlighted that the adherence to the program mainly depended on the motivation of the multidisciplinary team (MDT), especially its lead physician. The lack of resources at some CFCs was raised to account for variations

in the teams' engagement as the level of available staff seemed to influence the extent to which the team was effectively enlisted. The participation of a patient or parent in each local quality team varied depending on the cultural context of the centers, some being used to share information with patients/parents, having a patient group in the CF center for years, others being involved in patient therapeutic education while others were acting in a more partenalistic model of care. The support received from the hospital quality department in two hospitals was emphasized as a factor that facilitated the adoption of quality tools by the teams. The recommendation of the assessor was to evaluate the impact of the program on patient outcomes by 2015.

Given the innovative nature of the QIP PHARE-M in France, the cultural differences and various organizational contexts at the CFCs, an assessment of the impact of PHARE-M at the CFCs engaged in the experimental phase was expected after three years to continue the enrollment in the program. Will it show favorable changes in the patient outcomes in the group of CFCs engaged in the PHARE-M compared to the other CFCs? What impact on care management can be observed in 2015? Was the period sufficient to show improvements in the two areas? In which contexts is the impact of PHARE-M observed to be the strongest? The PHARE-M Performance research project, submitted at a call for projects of the French Ministry of Health and selected for funding in December 2012, aims at providing answers to these questions.

Method

1- A mixed methodology

The rationale of the PHARE-M Performance project is to show evidence of the performance of the PHARE-M program on patient outcomes and care management.

The study is based on a mixed methodology inspired on the one hand by epidemiology, using data from the French Cystic Fibrosis Registry, and on the other hand by the British guidelines on "Process evaluation of complex interventions" [92] :

1) *a quantitative study* to compare the changes over the 4 years in the patient health indicators of a closed cohort, using data from the French Cystic Fibrosis Patient Registry, between CFCs having benefitted from the intervention during the experimental phase and CFCs not having benefitted from the intervention up to 2015; and

2) *a qualitative study* to analyze the contextual elements and mechanisms brought into play by the PHARE-M intervention that could account for a difference in impact among

trained CFCs either on patient health indicators or on care management assessed according to the criteria of the chronic care model [93].

2- Quantitative Study

2-1- Design

- observational,
- national and multi-center, and
- before/after and here/elsewhere: a comparison of patient health indicators before and after the "PHARE-M training" program at "PHARE-M Group" centers versus "Control Group" centers.

2-1-1- Primary and secondary endpoints

- FEV1%
- BMI as an absolute value and as a Z-score (standardized normal distribution of the BMI for children under two years of age)

For this research in particular, the value selected for these indicators is the only value appearing in the French CF Registry for a given patient and a given year. It will be analyzed by category of patients defined by age, sex, age at diagnosis, and possibly severity of disease expression, treatment, and certain social characteristics (data appearing in the Registry).

2-1-2- Study population

A closed cohort was formed to identify the study population including the patients followed up at CFCs who met the following inclusion criteria according to the 2012 Registry data:

- patients seen at a CFC in 2012
- patients having two of the CF-causing mutations of the CFTR2 list published on Feb 2012
- patients not having received a transplant in 2012

A patient left the cohort if he or she no longer met the inclusion criteria after the annual data were updated in the Registry (2013, 2014, and 2015), i.e.: if he or she was a carrier of a mutation excluded from the CFTR2 list updated on 13/08/2015 [82]; if he or she was followed up at a CFC engaged in the PHARE-M in 2014 or 2015; if he or she changed CFC in the course of the study and in doing so, changed CFC group; if he or she received a transplant between 2013 and 2015 (data up to the transplantation were taken into account), or if the patient died between 2013 and 2015 (data up to the death were taken into account).

The cohort was divided into two groups: the "PHARE-M Group" and the "Control Group":
- The "PHARE-M Group" consisted of the patients followed up at one of the 14 CFCs trained in the PHARE-M in the experimental phase (1,309 patients).
- The "Control Group" consisted of the patients followed up at the CFCs not having benefitted from the intervention in the same period of time (2,490 patients).

2-2- Pairing of the two "PHARE-M" and "Control" Groups

A preliminary analysis of the cohort formed from the 2012 Registry data showed significant differences between the two groups of patients, before the PHARE-M intervention, in terms of: 1) distribution by age, 2) distribution by age at diagnosis, and 3) distribution by FEV1% value (see Table I).

Consequently, a 1:1 pairing of the patients from the Control Group was decided in an attempt to eliminate certain confounding factors that could be attributed to the type and size of the CFC to which the patient was assigned: each "PHARE-M patient" was associated with a "control patient" followed up at a center of the same type (pediatric, adult, or mixed) caring for a total number of patients belonging to the same interval ([1;50], [51;100], [101;150], [151;200], or [> = 200]). Reunion island CFCs were excluded from the Control Group to reduce heterogeneity in CF care. All "eligible" control patients for each patient in the PHARE-M Group were selected, and one control patient was randomly drawn from that group of eligible control patients (without replacement). The patients in the PHARE-M Group were paired in a random order.

At the end of the process, 1,104 patients remained in each of the two paired groups. The Control Group included 20 CFCs. No paired control patients were found for 205 "PHARE-M patients". As data are collected in the French Cystic Fibrosis Registry for all patients, exposure variables are identical in both groups. Completeness is similar: for FEV1, 20.2% and 24.5% of missing data corresponding to the children below 6 y.o., for whom this measure is not taken, and 0.6% and 3.5% for ZBMI, in the PHARE-M group and the Control group respectively. The two groups had a similar distribution by age (see Fig. 1). However, there remained a significant difference in average age at diagnosis (PHARE-M paired group: 1.9 years; control paired group: 2.5 years; p value: 0.0123); this could be due to the fact that newborn screening was implemented in the 1990s in Brittany, and that seven (out of the 14) CFCs in the PHARE-M Group are located in this region. Furthermore, a significant difference in FEV1% of +3.89% (p value = 0.0012) remained in favor of the PHARE-M patient group before the intervention (see Table II).

2-3- Analysis of the primary endpoint between the two groups

Changes over 5 years in patient health indicators are measured for 2011 (baseline), 2012, 2013, 2014, and 2015; each patient served as his or her own control. A difference in the rate of decline is expected between the two population groups, PHARE-M and control (see Fig. 2). Changes over time in FEV1% will be modeled and compared in the two groups using a mixed model for repeated data with adjustments for potential confounding variables. Measurements for a subject i at time j is given by the following model, where ε_{ij} are the normally distributed residual components with mean zero and covariance structure Σ :

$Y_{ij} = \beta_0^P + \beta_1^P t_{ij} + \varepsilon_{ij}$ for the PHARE-M group

$Y_{ij} = \beta_0^C + \beta_1^C t_{ij} + \varepsilon_{ij}$ for the CONTROL group

$cov(\varepsilon_{ij}, \varepsilon_{ik}) = \sigma_{jk}$

The covariance structure Σ is given by the σ_{jk}. It allows taking into account correlation between measurements on a same subject. Correlation is assumed to be null between subjects. The choice of a covariance structure will be data driven, but we can expect that the correlation between two measurements will only depend on the time lag between them. The most realistic covariance structure should be the so-called Toeplitz covariance matrix. A special case of the Toeplitz model is the first-order autoregressive model.

The question here is to investigate whether the two slopes are parallel or not, that is to test whether $\beta_1^P = \beta_1^C$ (H_0) versus $\beta_1^P \neq \beta_1^C$ (H_1).

Using this model, the slopes (i.e. decline in FEV1) in the two groups will be calculated and compared. Changes over time in BMI will likewise be analyzed by comparing the changes in the two groups from 2011 to 2015, taking into account the Z-score for children under two years of age. The average trends will be calculated and analyzed for different patient categories (such as age, sex, age at diagnosis, severity of disease expression, treatment, and certain social characteristics in the Registry). The changes over time in indicators will be presented for the "PHARE-M Group" population by CFC for crossing with the results of the qualitative study.

2-4- Audit of the quality of the data included in the primary endpoints' calculation

The patient data measured by the CFCs (height, weight, and FEV1 [per L]) for 2012 and 2013 underwent an on-site quality audit at the 14 CFCs in the PHARE-M Group. It

was the first on-site audit ever performed to establish the quality of these indicators. The objective was not to comprehensively audit all data for the patients included in the study. Rather, the objective was to comprehensively identify the different causes of error due to failures in the processes of measuring and/or selecting the values transmitted to the Registry in order to identify avenues for improvement of the quality of the data in the Registry. The sample of patients whose data were audited thus had to reflect the distribution by age range of the patients at each CFC (20 records/CFC) in order to cover the different measurement procedures defined by international benchmarks [94, 95, 96] and the data selection rules defined by the French Patient Registry Steering Committee, and to offer every opportunity to reach saturation of the various causes of error [97]. They will be taken into account in the interpretation of the results of the quantitative study.

3- Qualitative Study

3-1- Design

The design refers to the modeling of the intervention [92] including the contextual elements and the mechanisms shown in Figure 3.

The PHARE-M intervention consisted of establishing, training and coaching a quality team (QT) at each CFC comprising a number of professionals from the multidisciplinary CF team and 1 parent or patient from the CFC's caseload. The members of the QT have been trained in quality methods and tools and coached in changing care processes. The PHARE-M intervention should have directly impacted the ability of the local QT to master QI methods and tools, lead changes in the care processes, and should have generated good appreciation of the utility of the QT efforts. This direct impact of PHARE-M is identified under the heading "QT effectiveness". QT effectiveness may not only be the result of the PHARE-M intervention but may have been modulated by internal mechanisms, such as the composition of the QT (number of members and disciplines enlisted), its functioning (rigor in the QI work, decision-making, clarity of the roles…) and the parent or patient engagement. Those mechanisms are represented as impacting QT effectiveness (Fig. 3). Beyond the ability to master the QI methods and tools, the PHARE-M intervention was expected to have an impact on the quality of CF care delivered at the CFC. The Chronic Care Model [93] was deemed appropriate to account for quality of CF care across the 6 dimensions: existing improvement goals,

multidisciplinary care, self-management support, decision support (use of evidence-based guidelines), use of information system and electronic patient record, and organization of resources in the patient's community of life. Finally, an indirect impact of the PHARE-M intervention is expected on the trend in patient outcomes' evolution as measured in the quantitative part of this study. Moreover, some elements in the CFC contexts, which are external to the PHARE-M intervention and preexisted to its introduction, may have had a major impact both on the adherence of the team to the QI work and on its outputs. The contextual elements that have been brought in this study include the composition of the MDT, the leadership, the patient-centeredness of care, the innovative culture of the team, and the support from the hospital quality department.

The qualitative study will test these hypotheses using a questionnaire to be self-administered, in 2015, to all members of the MDT at the 14 CFCs and to the patients/parents participating in the quality teams.

Quality of care has been defined according to the criteria of the Chronic Care Model [93]; as this model has not been popularized in France nor in cystic fibrosis, we adapted it with 47 items aimed at characterizing CF care. *Table III presents a list of these items.*

QT effectiveness has been described in the studies by Lemieux-Charles [98] and Shortell [99]: it is characterized according to 27 items *(see Table IV).*

QT Internal factors that may have modulated the QT effectiveness: QT functioning [98] is characterized by 22 items classified in 4 categories 1) the organization at work, 2) the decision-making process, 3) the shared improvement goals, and 4) the ability to communicate and get external support. Studies by L. Lemieux-Charles defined these items to analyze the impact of adopting quality improvement practices on the internal functioning of a team. We use the same items to analyze if the team's functioning could modulate its effectiveness *(see Table V).*

The engagement of the patient/parent as characterized in Carman's framework [100] is assessed by a list of 31 items, prepared as part of this research *(see Table VI).*

The context elements include: the composition of the multidisciplinary team at the beginning of the PHARE-M intervention (2011) because it might have been a limiting factor in assigning staff to the QT; the culture of the microsystem to which the QT belongs [99] i.e. the organizational culture *(see Table VII)* and patient centeredness and leadership style *(see Table VIII);* the alignment of the PHARE-M QIP with the hospital

quality policy as described within the framework of the European QUASER study [101] using eight open questions in an interview with a head of the hospital quality department *(see Table IX).*

Focus groups with the members of each QT were conducted by the Clinical Research Assistant, designed around four open-ended questions: 1) What changes in the organization of the CFC can be attributed to the PHARE-M? 2) What difficulties were faced at the CFC? 3) What successes were achieved? and 4) What lessons from this experience after 3 to 4 years? The results of these focus groups involving the 14 CFCs will be put in perspective with the results of the survey conducted by one assessor of the pilot phase who interviewed the 7 first CFCs on the following themes: 1) PHARE-M applicability, 2) participation of patients and parents, 3) functioning and coordination, 4) perceived benefits and costs, 5) effect on the team, 6) effect on care management, and 7) recommendations for PHARE-M national deployment.

3-2- Development of the instruments of the realistic study

The self-administered questionnaire was developed from the instruments (cited above) translated into French, and new items prepared as part of this research to characterize quality of CF care and the degree of engagement of the patients or parents. The whole questionnaire is proposed to the members of the quality teams. A limited part of the questionnaire is proposed to the members of the MDT not on the quality team. The questionnaire has been prepared from January to June 2014 with clinicians from the Nantes/Roscoff CF-CERD and experts from the Health Education and Practice Laboratory (LEPS) at the Sorbonne Paris Cité University - Paris 13 Bobigny. It has then been tested between July and September 2014 in three teams from the Nantes/Roscoff CF-CERD (pediatric, adult, and mixed) with 29 respondents from all disciplines and the patients/parents participating in the QT. As a result of these tests, the questionnaire has been slightly adapted, essentially by rewording parts of the French translation and adding free text fields (*Questionnaire available upon request to the corresponding author*).

4- On-site investigations

The investigations conducted by the clinical research assistant at the 14 PHARE-M centers take place over the course of 2.5 consecutive days per CFC. The questionnaire is self-administered successively under the supervision of the clinical research associate according to a schedule established with the team at the site, with no possibility of

communication or consultation among respondents. The questionnaires and responses are managed in SurveyMonkey Software and subsequently exploited using SAS and Excel Software. The focus group is conducted at the end of the visit. Each focus group is recorded using audio equipment and transcribed in writing.

4-1- Analyses of responses and validation of the questionnaire

Responses to the items of the questionnaire are processed anonymously. Each item receives a score on a Likert scale from one to four based on the degree to which the respondent agrees or disagrees with the proposition: “Completely disagree; Disagree; Agree; Completely agree”. "No" and "Unknown" responses are assigned a score of 0. The score is reset to 100 points and can thus be totaled by theme of the questionnaire and category of respondents. An initial descriptive analysis of the responses by CFC is returned to each quality team in the month following the on-site investigation, via a web conference, in order to validate the interpretation of the scores for the different themes and identify avenues for or obstacles to continuous care quality improvement at the CFC. A Cronbach's alpha test will be performed on all responses collected at the centers. Since the anticipated number of respondents is around 130 people in total for the 14 teams, this test will not allow the questionnaire to be modified for use in a larger population of respondents. It mainly aims to validate the French translations of the parts of the questionnaire coming from previous studies in English and discuss the use of the parts created within this research study.

A second level of descriptive analysis will be performed by aggregating the responses (all CFCs, by professional discipline, for resource patients/parents, and for professionals) to search for potential associations between quality of care at the CFC 3 years after the PHARE-M intervention and the effectiveness of the QT and/or the engagement of parents/patients and/or contextual elements.

After the publication of the Registry report presenting the 2015 data, changes in indicators from 2011 to 2015 will be crossed with the results of the realistic part of the study, in an attempt to identify any association in relation with more favorable changes over time in patient outcomes. A "signature" set of factors associated with a maximum/minimum impact of the PHARE-M will be sought.

4-2- Analyses of the content of the focus groups

The content of the focus groups will be exploited (coding, categorization), processed (analysis, validity), and interpreted according to the standard thematic content analysis

protocol [102]. This will be done by grouping and counting within the framework developed during the pilot phase assessment.

4-3- Regulatory matters

Regulatory authorizations were granted for the quantitative research part focused on the patients' personal health data: a favorable opinion from the Ethics Committee of the Brest University Hospital (CHU) (session on 13 May 2014) and a notification of authorization by CNIL for a change in data processing stipulating the addition of a new recipient of the Registry data within the framework of a care quality improvement program (DR2015040 on 16 February 2015).

Conclusion/Discussion

Scope of the study and generalization

The research program aims at identifying the impact of the PHARE-M quality improvement program three years after the intervention at the 14 trained CFCs, situated in different organizational and cultural contexts. It uses a mixed methodology crossing the results of a quantitative analysis based on registry data and the results of a qualitative study designed in accordance with the recommendations for research on complex interventions.

The scope of the PHARE-M intervention and thus of the research concerns the management of a singular disease in a care network organized since 2002, which represents a relatively controlled scope. Therefore, the influence of contextual elements on the PHARE-M program's impact can be analyzed independently from other confounding factors associated with different organizations for the management of various diseases or different hospital departments running diverse specialties.

Fourteen centers volunteered to engage and test the PHARE-M program; they were not randomized. Moreover, initial assessment highlighted that team motivation is a determinant of the speed of adherence to the program. This pattern of our research, focusing on an experimental phase having enlisted volunteer centers, is to be considered in interpreting the results and developing recommendations for a successful roll- out of the PHARE-M program in the national network.

Finally, the research study on the PHARE-M intervention has a study design that could be applied in the assessment of other complex interventions at healthcare settings.

Hence, this research study could inform the assessment of interventions concerning the care of rare and/or chronic diseases and the instruments needed for such assessment.

Limitations identified and initial lessons

As a result of the experimental study based on Registry data, a study population paired between two groups (intervention and control) was defined to eliminate certain confounding factors, especially factors linked to patient age distribution. Despite this pairing, significant differences remained in terms of patient age at diagnosis and primary endpoint (FEV1%) between the two groups before the intervention, in favor of the intervention group. These initial differences could have a favorable effect for the rate of decline in FEV1% in four years in the intervention group [103, 104]. The question is to investigate whether the slopes are parallel or not. The difference in FEV1% will be taken into account using two different intercepts in the model, one for the intervention group and one for the control group. The patients belonging to either the "PHARE-M" group or the "Control" group will be identified in the Patient Registry with respect to their group for further analysis of their health outcomes.

Moreover, on-site quality audits of the Registry data included in the calculation of the primary endpoints showed discrepancies, mainly due to the CFCs' interpretation of the rule for selecting the values to transmit to the Registry [97]. The volume of the discrepancies identified in the data audited could be attributed to the change of the rule applied from the 2011 registry survey. This audit points out the need for a certification process to enable a larger use of this database in epidemiologic studies or for public health or pharmacovigilance purposes.

The survey conducted for the qualitative study of the multidisciplinary teams at the 14 centers should include around 130 respondents, including at most 14 patients/parents. This number of respondents might seem low for having enough statistical power in the statistical validation of the survey instruments, especially for the parts of the questionnaire developed within this research. The survey instruments could be improved within the framework of subsequent research studies aiming, for example, at comparing quality of care between centers trained in the PHARE-M and centers untrained in the program, or at making an assessment of the quality of care before/after another intervention. Therefore, this questionnaire represents an instrument that could have further uses in the network.

Expected results in terms of quality improvement of care

If the research study enables to identify factors promoting the adoption of the PHARE-M QIP and the maximization of its impact at CFCs, attention must be paid to the contextual elements to be worked on before or in parallel with the introduction of this program at the remaining CFCs. In the United States, the CFF has conducted "Leadership Collaborative" programs to develop leadership on multidisciplinary teams. The availability of the MDTs staff at the European standards for the number of patients followed could also represent a pre-requisite for their participation in the PHARE-M. The quality of care assessed after three years within the CFCs trained to PHARE-M might also enable to identify new avenues for improvement, including some beyond the scope of the clinical microsystem such as the Information System or the generalization of Guidelines.

Table I — Distribution by age, age at diagnosis and FEV1% of the 2012 study population between the two groups of the study cohort before pairing.

Comparison of the two groups	PHARE-M (N=1051)			Control (N=1962)		
Comparison of Ages	Avg.	Med.	Max.	Avg.	Med.	Max.
Age of patients (years)	15.0	13.0	62	18.0	17.0	74
Age at diagnosis (years)	2.0	0.1	51	3.2	0.2	71
Comparison of FEV1%	Avg.	LLM	ULM	Avg.	LLM	ULM
FEV1%	83	81,55	84,45	75,48	74,33	76,64

Table II — Comparison between the PHARE-M Group and the paired Control Group

Comparison between PHARE-M Group and Control Group		PHARE-M (N=1104)	Contrôles (N=1104)	Patients PHARE non paired (N=205)	Comparison between PHARE-M Group and Control Group (proc TTEST)
Gender	Men *n (%)*	582 (52.72)	564 (51.09)	93 (45.37)	
	Female *n (%)*	522 (47.28)	540 (48.91)	112 (54.63)	
Age	Average	15.57	16.05	14.48	
	Std Deviation	10.73	11.00	10.51	
Age (classes)	00-04 *n (%)*	182 (16.49)	175 (15.85)	32 (15.61)	
	05-09 *n (%)*	209 (18.93)	206 (18.66)	42 (20.49)	
	10-14 *n (%)*	213 (19.29)	204 (18.48)	48 (23.41)	
	15-19 *n (%)*	169 (15.31)	168 (15.22)	38 (18.54)	
	20-24 *n (%)*	125 (11.32)	130 (11.78)	19 (9.27)	
	25-29 *n (%)*	93 (8.42)	84 (7.61)	10 (4.88)	
	30-34 *n (%)*	53 (4.80)	68 (6.16)	4 (1.95)	
	35-39 *n (%)*	36 (3.26)	34 (3.08)	6 (2.93)	
	40-44 *n (%)*	7 (0.63)	18 (1.63)	1 (0.49)	
	45-49 *n (%)*	9 (0.82)	11 (1.00)	4 (1.95)	
	50-54 *n (%)*	4 (0.36)	3 (0.27)	0	
	55-59 *n (%)*	4 (0.36)	2 (0.18)	0	
	60-64 *n (%)*	0	0	1 (0.49)	
	70-74 *n (%)*	0	1 (0.09)	0	
VEMS	Nmiss	223	270	49	
	Average	83.00	79.11	85.06	p=0.0012 (S)
	Std Deviation	23.96	25.81	21.92	
ZBMI	Nmiss	7	39	2	
	Average	-0.17	-0.14	-0.18	p=0.5171 (NS)
	Std Deviation	1.05	1.15	1.11	

409 Table II (Followed): Comparison of Age at diagnosis between PHARE-M and Control

Age at diagnosis (years)

	Control	PHARE-M	Patients PHARE non paired
Nmiss	33	39	2
Average	2.49	1.85	2.47
Std Deviation	6.34	5.33	6.30

	P-value*
Comparison of Age at Diagnosis between PHARE-M and Control Groups	0.1317

***Test de Wilcoxon**

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412 Table III — Criteria for quality of CF care derived from the chronic care model

IG — Improvement Goals at the CFC	1 — There are improvement goals at the CFC
	2 — These goals, if they exist, are the subject of both indicators and an action plan at the CFC
	3 — The CFC has tools to follow up this action plan in the form of a dashboard
	4 — To your knowledge, this action plan has been discussed with management and validated
SMS — Self-Management Support - Therapeutic Patient Education	1 — To your knowledge, there is a therapeutic education program for patients at the CFC authorized by the French regional health agency (ARS)
	2 — In your opinion, the professionals at the CFC are well trained in TPE
	3 — More than 80% of the patients/parents attended at least one TPE session in the last year
	4 — The total time spent by the professionals on TPE is sufficient
	5 — There are no obstacles to implementing TPE at the CFC
	6 — The team is involved in the studies of one of the French national groups on therapeutic education via face-to-face participation or regular reporting of information
	7 — The CFC has priority objectives for developing TPE
	8 — If yes, the CFC has indicators to follow up the achievement of these priority objectives
MM — Multidisciplinary management	1 — To your knowledge, the multidisciplinary team at the CFC comprises all the disciplines recommended by the French National Diagnosis and Treatment Protocol (PNDS): specialist physician, nurse, physiotherapist, psychologist, secretary, and social worker
	2 — The number of staff in all disciplines is sufficient for the number of patients followed up
	3 — In your view, the multidisciplinary team seems stable over time (the professionals' turnover rate is below 20% in a year)
	4 — The members of the multidisciplinary team have a great deal of expertise in managing cystic fibrosis
	5 — The multidisciplinary team meets often enough to perform a summary of the records of the patients who have come to the CFC
	6 — During these multidisciplinary meetings, the team generally reviews the records of the patients with a scheduled visit to the CFC
	7 — During these multidisciplinary meetings, the team regularly examines the patients' educational needs and the outcomes of the educational sessions held
	8 — The scheduled consultation is genuinely multidisciplinary: the patient meets with at least the physician, the nurse, and the physiotherapist
	9 — The scheduled consultation allows the patient to meet with a professional other

	than the ones mentioned above, as required (dietician, psychologist, or social worker)
	10 — The scheduled consultation allows the patient to benefit at least once per year from a TPE session on a priority objective for him or her
	11 — When a patient requires it, the CFC is able to call upon a network of referent professionals in other disciplines with knowledge of cystic fibrosis (geneticist, endocrinologist, ENT, gastroenterologist, etc.)
	12 — It is possible to be managed at the CFC on a 24/7 basis
	13 — Patients who arrive at the hospital emergency department are managed in accordance with a protocol established by the CFC with the emergency department for patients suffering from cystic fibrosis
	14 — The team regularly holds a meeting to discuss its functioning and the problems at the CFC in order to improve care management
DS — Therapeutic decision support (guidelines)	1 — The team manages the availability of guidelines (nutritional, respiratory, hygienic, etc.) in a way that they are accessible to all professionals
	2 — The team has defined an internal reporting procedure to insure that care management recommendations (guidelines) updates are accessible to the team
	3 — The team systematically verifies for each patient that the latest recommendations are applied and/or offered to him or her
	4 — The team uses alerts on the population followed up to verify that the latest recommendations for care are applied to the eligible patients (e.g. glucose tolerance test alert, vaccination alert, examination alert, etc.)
	5 — The team has optimally organized the multidisciplinary consultation process (circuit, schedules, chain of professionals, cross-contamination, hazards, etc.) to deliver high quality of care.
	6 — The team has optimally organized the process of responding to telephone or email messages from the patients and families
IS — Patient information system	1 — The team uses an electronic cystic fibrosis patient record
	2 — The team has an electronic patient record system that allows it to view changes in the patient health outcomes (nutritional and respiratory outcomes) over the course of several years
	3 — The team uses the electronic patient record system during the multidisciplinary staff meetings
	4 — The team displays information from the electronic patient record during the multidisciplinary meeting (graphs of changes over time, reports from previous consultations with different professionals, etc.)
	5 — The team uses the electronic patient record system both to create alerts on applying recommendations for the patient and to compile statistics on the population followed up

	6 — The team uses the electronic patient record system to include biology results
	7 — The team uses the electronic patient record system to include imaging results
	8 — The electronic patient record system helps in selecting patients for clinical trials
	9 — The electronic patient record data are automatically transmitted with a good degree of reliability (minimal verifications, corrections, and additions) to the French Cystic Fibrosis Registry
SN — Staff in the networks in the community	1 — The CFC has organized a network of professionals in the patient community for managing care at home
	2 — The CFC organizes regular trainings for professionals in the patient community
	3 — The CFC regularly evaluates the professionals caring for CF patients in the community
	4 — The CFC assesses the health providers of devices managing CF patients
	5 — The CFC assesses the needs for home care and its distribution between professionals and carers for a balanced organization of home care
	6 — The CFC provides the patients with offers of sports activities, creative activities, and psychological support near their place of residence

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414 Table IV — Effectiveness of a quality team (QT)

Command of the quality process and tools	1. The teams that implement a quality process have a clear vision of the area on which to focus their improvement efforts and the expectations to be met. When you started the project, did you have such a vision?
	2. The quality teams sometimes use a method for making progress, such as a guide to follow step by step which helps them organize their work. Did your team use such a structured method?
	3. Did your team make one or more changes in its way of working?
	4. Did the team analyze data to ensure that such change(s) indeed represented an improvement?
	5. Did the team try to understand variations in the CFC processes and the reasons that could account for them (variations over time or between professionals, time of year, patient characteristics, etc.)?
	6. Does the team routinely have data allowing it to make a state of play and identify problems?
	7. Did the team have to develop a system to collect specific data (such as questionnaires, audits, interviews, or measurements) to identify problems and assess the responses provided?
	8. Did the team establish a data collection system to continue to manage quality or monitor the new processes established?
	9. Was the team able to rely on a referent professional to coordinate the meetings and work of the quality team?
	10. Was the team able to rely on a referent professional to collect and analyze data?
	1. The team was able to perform measurements to define and assess changes within the framework of tests.

Capacity to drive change	2. After testing a change, the team succeeded in discussing the outcomes observed and learning from this test.
	3. The team succeeded in analyzing the outcomes of the test to propose new changes or adjustments to be tested.
	4. During the process, the team was able to easily incorporate and adapt ideas for changes to meet the organization's needs.
	5. The team was able to enlist sufficient knowledge and skills to drive change under good conditions.
	6. The team could find sufficient assistance in the hospital to support changes.
	7. The team could sufficiently rely on the support of the French national team to make changes at the CFC.
Effectiveness perceived by the quality team	1. The performance of the PHARE-M steering team met my expectations.
	2. I was satisfied with my experience as a member of the quality team.
	3. I believe that my participation was useful and positive for the work of the team.
	4. I would be willing to participate again on a similar team to work on quality improvement.
	5. I believe that the work of the quality team was useful for improving quality.
	6. The outcomes achieved through the work of the quality team meet the organization's needs for improvement.
	7. It is necessary to maintain an ongoing quality improvement process to continuously improve care at the CFC.
Effectiveness perceived by the rest	1. I believe that the work of the steering team was useful for improving quality at the CFC.
	2. I believe that the entire team at the CFC was enlisted and contributed to quality improvement.
	3. I believe that the outcomes achieved collectively meet the organization's needs for improvement.
	4. I believe that it is necessary to maintain an ongoing quality improvement process to continuously improve management at the CFC.

Table V — Internal functioning of the quality team (QT)

Strictness of organization and clarity of roles	1. The leader was clear and explicit on how he or she wanted the team to work.
	2. The leader reviewed the steering team's work and asked how we were going to go about it.
	3. The leader also requested the opinion of the other members of the team.
	4. The leader's behavior reflected the importance he or she placed on the quality team functioning well.
	5. Our team could have been better at seeking help and securing more skills to do the work.
	6. Sometimes it seemed that we were working or going about the matter in the wrong way.
	7. Roles were so unclear that the work of different individuals seemed to overlap.
	8. The members of the team had different outlooks and experiences and came from different disciplines.
De	1. Most of the members of the team had an opportunity to participate in decision-making.

	2. We appreciated our differences, which shaped our decisions.
	3. The contribution of each member of the team was heard and taken into consideration.
	4. We examined many different ideas before making a decision.
	5. Our team possessed sufficient resources and skills and applied them well enough to work properly.
	6. Our team worked well enough to accomplish its mission satisfactorily.
Clarity of objectives	1. The members of the team were in agreement on the objectives of the project.
	2. The achievement of the objectives guided the activities of the members of the team.
	3. The members of the team did what was expected of them.
	4. The members of the team were all focused on the achievement of the same objectives.
Communication and cooperation	1. There was a great deal of cooperation between the different hospital departments.
	2. In this hospital, most departments and services have a hard time sitting down at a table and solving problems together.
	3. The people I worked with were comfortable with suggesting changes and improvements.
	4. Our team received all the information required to plan and organize its work.

Table VI — Engagement of the patients/parents on the quality team (QT)

Information and activation of the patients/parents	1. The patients and parents are educated regularly (annually or more often) by the team about general subjects concerning cystic fibrosis care and research.
	2. The patients and parents are rather familiar with general cystic fibrosis information: research, progress made, and Registry data.
	3. The CFC team has educated the patients and parents about the PHARE-M's importance and aim.
	4. A good relationship between the patient or parent recruited and the team is indispensable for the patient or parent to participate in the PHARE-M.
	5. The patient or parent recruited is well informed of the challenges (10 commitments) of management quality.
	6. The presence of a patient or parent on the steering team is a given and an asset.
	7. The place of a parent or patient is not on a quality team, because he or she does not have enough training or education.
	8. The place of a parent or patient is not on a quality team, because he or she already has too many personal problems to manage.
	9. The patient or parent recruited possesses the qualities to become a member of the steering team.
	10. The patient or parent recruited must have developed coping skills (see therapeutic education standard: knowing how to manage emotions and stress; solving problems, making decisions, and

	making choices; knowing how to communicate and being adept in relationships with others; and knowing how to put oneself in the place of others).

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Empowerment of patients/parents to allow them participate in the QT	1. The participation of a patient or parent depends on the systematic reimbursement of his or her travel expenses.
	2. The participation of a patient or parent should be facilitated by the reimbursement of other expenses: child-care, lost working hours, etc.
	3. The participating patient or parent does not represent all patients.
	4. The patient or parent was selected by the team based on a list of specific criteria (cultural level, capacity to communicate, availability, etc.).
	5. The patient or parent is motivated to improve management for all.
	6. The patient or parent is also motivated to improve his or her own management by participating in the program.
	7. It is important to communicate with the other patients or parents concerning the role of the patient or parent on the steering team.
	8. It would be necessary to include several patients or parents to ensure that more different points of view are represented.
	9. The patient or parent must be knowledgeable about the disease and its management beyond the requirements of his or her own care.
	10. The patient or parent must be knowledgeable about the general functioning of the hospital.
	11. The patient or parent must know how to communicate with the professionals by taking a step back and drawing general lessons from his or her own experience.
Capacity for effective contribution of the patients/parent	1. The PHARE-M national organization created good conditions for incorporation of the patient or parent.
	2. The participation of a patient or parent on the team at French national training and information meetings (four French national face-to-face "EPE" meetings) is indispensable.
	3. The patient or parent participated and contributed as much as the professionals during the French national "EPE" meetings.
	4. The patient or parent's regular participation at quality team meetings at the CFC is indispensable.
	5. The patient or parent participates in and contributes significantly to the work of the steering team.
	6. The patient or parent's ideas and proposals are generally taken into account by the steering team.
	7. The atmosphere of work of the steering team meeting is better and more productive when the patient or parent is present.
	8. The pace of work is slower when the patient or parent is present at the steering team meeting.

	9. Certain decisions made by the steering team are inspired by the patient/parent.
	10. The process of incorporation and participation of the patient or parent should be reviewed and improved for the continuation of the PHARE-M.

Table VII — Organizational culture

Organizational culture:
Research studies have defined four types of organizational culture, arising from both the organization's external environment and internal management: a "familial" type, an "entrepreneurial" type, a "prescriptive" type, and a "productive" type.
The five rubrics below describe the characteristics associated with these different types of organization.
You have 100 points to distribute among the four proposals based on the degree to which they resemble your organization. For example: If the CFC resembles Description A a great deal and Description B a little, and does not resemble Description C or Description D at all, assign 70 points to Response A and the 30 remaining points to Response B.

§1. Character	1. Organization A is very familial, like a big family. People seem to share a lot of themselves.
	2. Organization B is very dynamic and entrepreneurial. People seem to want to venture off the beaten path and take risks.
	3. Organization C is very structured and formalized. Procedures govern people's work.
	4. Organization D is very focused on production, with the concern being that the work gets done. Individuals are not very personally involved.
§2. Management	5. Organization A's director(s) are warm and attentive. They try to develop people's potential and act as mentors or guides.
	6. Organization B's director(s) take risks. They encourage people to be innovative and to try out new ideas by taking risks.
	7. Organization C's director(s) enforce rules. They expect people to strictly apply policies and procedures.
	8. Organization D's director(s) resemble coordinating coaches. They help people achieve the organization's objectives.
§3. Cohesion	9. Organization A's factors for cohesion are loyalty and tradition. Dedication to the organization is high.
	10. Organization B's factors for cohesion are the race for innovation and development. There is a desire to be the first.
	11. Organization C's factors for cohesion are hierarchical rules and establishment policies. Maintaining suitable functioning is important here.
	12. Organization D's factors for cohesion are the achievement of objectives and the performance of required tasks. This vision of production is shared.
§4. Emphasi	13. Organization A emphasizes human resources. Having strong cohesion and a high sense of morale are important.
	14. Organization B emphasizes growth and acquisition of new resources. Being ready to rise to

	new challenges is important.
	15. Organization C emphasizes permanence and stability. Complying with rules and performing operations smoothly are important.
	16. Organization D emphasizes competition to achieve objectives. Measuring results is important.
§5. Recognition of efforts	17. Organization A recognizes all its members' efforts equally. It is important that everybody in the pyramid, from the very top to the very bottom, is treated as equally as possible.
	18. Organization B rewards individual initiative. Those who have the most ideas and perform the most innovative actions receive the most recognition.
	19. Organization C modulates recognition based on rank. The higher your position, the more your efforts are recognized.
	20. Organization D rewards the achievement of objectives. Individuals who demonstrate leadership and thus help achieve objectives are recognized.

Table VIII — Patient-oriented culture and leadership

Patient-oriented organization	1. Our organization works to properly identify patient needs and expectations.
	2. The professionals handle patient requests promptly.
	3. Patient complaints are analyzed to identify recurring causes and prevent problems from being replicated.
	4. The organization uses data from the patients themselves to improve services.
	5. The organization uses data regarding patient satisfaction and/or patient expectations to improve services.
Leadership at the CFC	1. The leader develops interesting/exciting opportunities for our organization.
	2. The leader proposes new and even innovative ideas to improve management services and processes.
	3. The leader drives the organization to meet patient needs and ensures management/care safety.
	4. The leader takes into account the needs of both the service and the staff during major changes within the organization.
	5. The leader builds close, positive relationships with the other departments in the hospital.
	6. The leader builds close cooperative relationships with other organizations outside the hospital.

Table IX — Open-ended questions to the hospital's quality department

1. What are the priorities of the hospital's quality department?
2. Support for care services in quality improvement: was another quality program developed for another disease or another care service?
3. How are patients included in the different committees and groups working to improve quality in the hospital?
4. How is quality measured (main indicators)?
5. What training programs in quality tools and methods are promoted by the hospital?
6. How was the quality department informed of the PHARE-M (by whom and when)?
7. What were the reasons for the quality department's engagement (or non-engagement) in the PHARE-M, in support of the CFC? In the case of engagement, what resources and time were dedicated to supporting the CFC?
8. How is the PHARE-M perceived by the quality department management in terms of coherence with hospital policy, perceived effectiveness, and other matters? If necessary, the example of another quality improvement program rolled out in the hospital can be cited.

Figure 1 — Distribution by population age between the two groups (PHARE-M and control), paired in 2012 data.

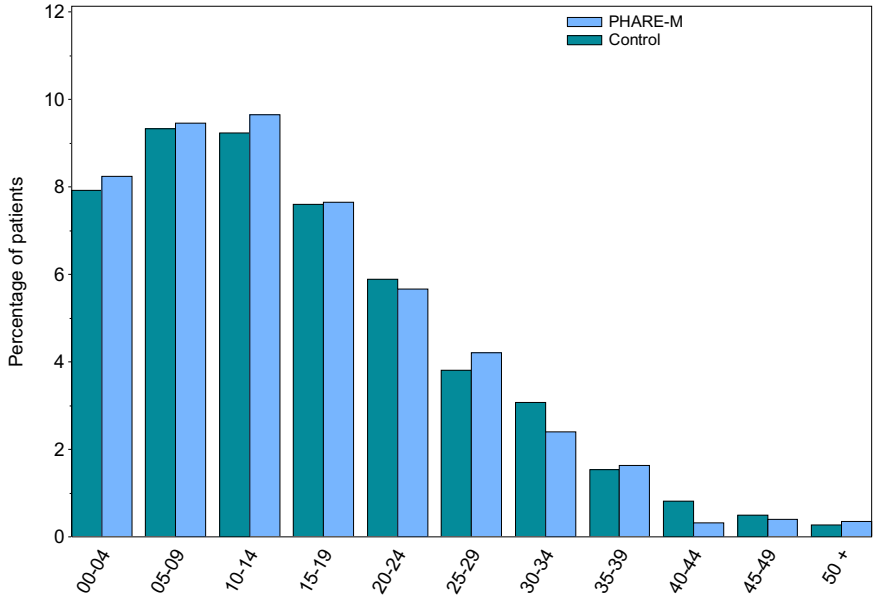


Figure 2 — Representations of the analysis of the primary endpoint

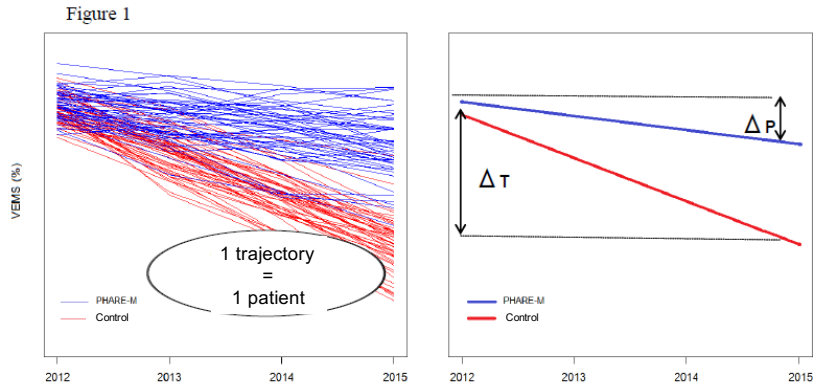
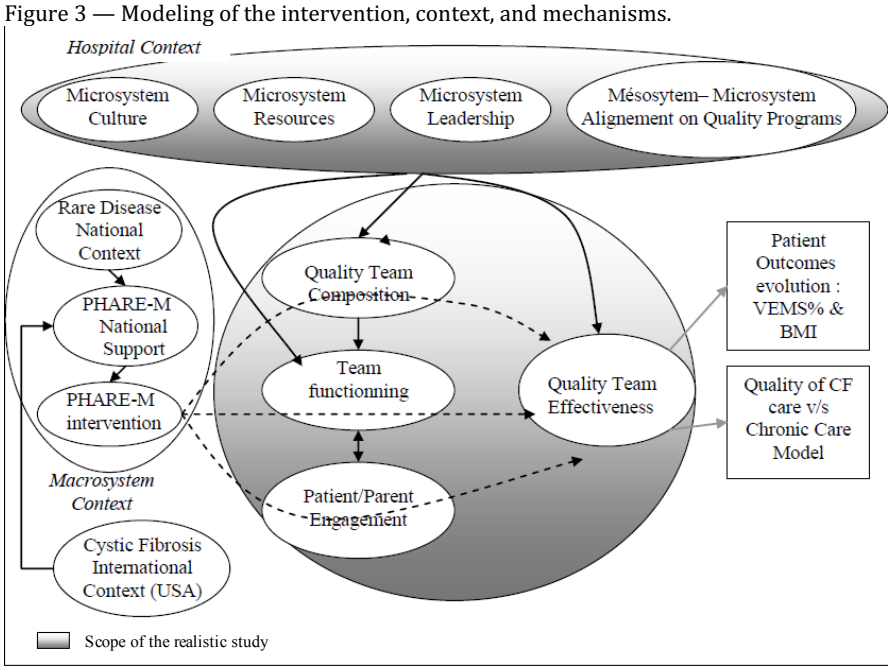


Figure 3 — Modeling of the intervention, context, and mechanisms.



Lessons from the On-Site Quality Audit of Data Transmitted to the French Cystic Fibrosis Registry

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Abstract

Background:

The French Cystic Fibrosis Registry takes a census of the population of patients and records their annual data transmitted by Cystic Fibrosis Centers (CFCs). Quality of patient data has been a focus in the past years, with the implementation of automated controls before data integration.

Objective:

To assess, at the 14 CFCs trained in the quality improvement named *Hospital Program to Improve Outcomes and Expertise in Cystic Fibrosis (PHARE-M)*, the quality of the 2012 and 2013 data transmitted to the French Registry with respect to the rules established to obtain forced expiratory volume in one second (FEV1%) and anthropometric data.

Method:

The clinical researcher selected 20 patients at each CFC from age ranges corresponding to different visit frequencies and measurement procedures in order to reach saturation of error causes. The control consisted in comparing source data, pulmonary function tests (PFTs), patient records, and data in the Registry.

Results:

The audit focused on 242 patients, 2,455 consultations and 1,855 PFTs. Less than 5% of data concerning weight, height, or FEV1 (L) in the patient records files had discrepancies with source data. Discrepancies on patient height between patient records and PFT files were found in 11% of cases. For one hundred and ten patients (45%), anomalies were found between the patient record and the Registry for the FEV1% and the associated anthropometric measurements mainly related to the interpretation of the selection rule of the venue corresponding to the “best spirometry in the year” and the reference standard used (local standards versus Knudson reference equations). For the 33 children in the age range of 6-17 years old (27% out of 120 children records controlled), the FEV1% value in the Registry presented an average deviation of + 4.25% (min. = -9.3%; max. = +16.9%; median = 4%) with the value from the Patient record.

Conclusion:

This first on-site quality audit of the data transmitted to the Registry pointed out variability in the measurement process at the CFCs. The rule for selecting the data for the Registry was applied differently at some CFCs, and various local References for the FEV1% calculation were used. Avenues for improvement have been identified.

Keywords: cystic fibrosis, registry, quality audit, measurement recommendations.

Background

History of the French Cystic Fibrosis Registry

The French National Cystic Fibrosis Observatory was established in 1992. Its initial objective has evolved into a comprehensive census of the population [105], allowing it to become the French Cystic Fibrosis Registry [106] certified by the French National Committee of Rare-Diseases Registries in 2008. It falls in the group of six countries whose Registry is classified as grade A based on criteria of comprehensiveness of the census population and precedence [107]. It is funded and managed by the_patient organization Vaincre la Mucoviscidose with the support of the Patient Registry Steering Committee (PRSC) including the organization's medical & scientific directors, clinicians, demographers and epidemiologists. The objectives of the French Registry are:

- To take a comprehensive census of people suffering from cystic fibrosis by including data on diagnosis (French Association for Screening and Prevention of Child Handicaps and CFTR-France), death (CépiDc — INSERM) and transplantation (HEGP);
- To have annual data concerning the patients followed up at healthcare centers in France (mainland France, Réunion Island, and Guadeloupe);
- To help improve knowledge of the medical and social characteristics of the population suffering from cystic fibrosis and to assess the impact of therapeutic advances on the evolution of state of health and survival;
- To assess the socioeconomic cost of this disease in terms of treatments and management and to anticipate changes in this cost; and
- To have information to shed light for the choices of parents and patients and the strategic choices of associations and other institutional partners.

The data transmitted to the Registry by the CFC teams once per year in the annual survey, according to various procedures, concern: semi-anonymous patient identification, diagnosis of cystic fibrosis, medical follow-up, social data, long term therapies prescribed, anthropometric data, pulmonary function data, and bacteriological data. The main survey is supplemented by thematic surveys: Pregnancy, *Burkholderia cepacia*, and Enrollment in Clinical Trials. Quality of patient data has been a focus for the PRSC in the past years, leading to the increase of automated controls of completeness and consistency of data before their integration in the Registry. The Registry is used for epidemiological and socioeconomic studies. Since 2006, reports by center have been issued to compare the outcomes at each CFC to the French national averages and to the outcomes at the other

anonymized centers. In 2013, the French Registry recorded 6,329 patients [[108]], 6,275 (99.1%) of whom had been seen by a CFC at least once in the year. For the first time in the history of CF in France, the number of adults exceeded the number of children or adolescents in the Registry (50.6% were adults).

PHARE Performance research project

The PHARE-M quality improvement program (QIP) was launched in 2011-2013 in 14 CFCs willing to engage in the approach (Fig.1). The research project, named PHARE-M Performance, funded by the French Ministry of Health in 2012, aims to assess if, in 2015, there is a measurable positive discrepancy in the trend of patient outcomes (Forced Expiratory Volume in 1 second, or FEV1, and Body Mass Index, or BMI) between patients followed up at CFCs involved in PHARE-M in 2011-2013 and patients followed up at CFCs not involved until 2015 (control group) [[109]]. A closed cohort was formed in 2012 for this research project including patients meeting the following criteria: genetic criteria (two CF-causing mutations [[110]]), uninterrupted follow-up at a CFC belonging to one of the two groups (trained or not trained in the PHARE-M), and no lung transplant. The annual Registry values for FEV1% and BMI are used as primary endpoints to determine the performance of the PHARE-M program by assessing the three-year trend (2012-2015) between the two groups of patients. The FEV1% and BMI values are calculated by the Registry software from patient's FEV1 in liters (FEV1 L), height and weight values transmitted by the CFC. The Knudson reference equations are used to obtain the FEV1% value. Thus, best research practice led to assess the quality of the data (FEV1 L, height and weight) transmitted to the Registry to calculate FEV1% and BMI Z-score for the population enrolled in the study cohort.

Some additional hypotheses led to clarify the audit's objectives and scope:

- The recording of reliable data in the Registry is one essential aspect of quality improvement and as such, the CFCs from the PHARE-M group must be audited so that they take actions for improvement, if necessary;
- The CFCs from the "Control" group will only be known by the end of 2015, since all those that engaged in the PHARE-M between 2013 and 2015 are excluded; thus it is difficult to audit data in this group during the course of the research project;
- The research project is not a substitute for a national Registry data quality audit, which may be decided and framed at the national level by the PRSC, should the audits

conducted within the framework of this research study point to the need for such a process;

- The audits conducted on a sample of patient records at 14 CFCs should identify all the possible causes of error or, at least, all the main causes of error, in order to reach saturation of error causes.

Objective

Within the framework of the PHARE-M Performance research project, audit at the 14 CFCs of the PHARE-M group, on a sample of patients enrolled in the closed cohort of the study, the quality of the data transmitted to the Registry for the years 2012 and 2013, with respect to the rules established to obtain the height, weight, and forced expiratory volume in one second in liters (FEV1 L). A secondary objective is to assess on the sample of patients the difference on the FEV1% values between the CFC patient record and the Patient Registry, and if any, analyze the causes.

Method

Patient data submitted for the audit

All data submitted for the audit were from the Registry database and hard-copy or electronic patient records and examined during on-site visits by the CRA (clinical research assistant). They include:

1) Data for patient identification by the Registry and by the CFC patient record management tool:

- Patient Registry Identification No.
- Initials of Last Name, First Name
- Date of Birth
- Gender
- No. of the CFC following the patient

2) Data for enrollment in the closed cohort of the study:

- Mutations in the CFTR gene
- Status with respect to transplant
- Status with respect to death

3) Measured data used to calculate indicators:

- Date of measurement

- Anthropometry: weight and height
- FEV1 in L

4) Data calculated based on general population benchmarks:

- FEV1 as a percentage of the expected theoretical value for age, height, and gender

Rule for the data to be transmitted to the Registry

The data transmitted annually to the Registry by the CFC teams must meet the rules established by the PRSC. Since the 2011 survey, the spirometry and anthropometry data to be transmitted to the French Registry must correspond to the visit at which the best forced expiratory volume in one second (FEV1) in the year has been measured, and no longer to the last visit of the patient in the year, as had been done until the 2010 survey. This rule has been worded as follows in the 2011 and subsequent questionnaire: *"Please specify the best spirometry values for the year. If there has been no spirometry: Check 'Spirometry not done' and indicate the date and the most recent anthropometry values for the year."* Realizing the ambiguity of the wording, and given the fact that the three software used by a number of CFCs for the follow-up of CF patients automatically select the visit corresponding to the best FEV1 measured **in liters** for the patient, the Quality Control Team decided to take the following rule to designate the reference FEV1 value that should have been transmitted to the Registry: "Select the visit at which the best FEV1 L value in the year has been measured".

Selection of the sample of patients for the audit

The sample of patients whose data will be audited should reflect the distribution by age ranges of patients at each CFC in order to cover all the measurement procedures as defined by the international guidelines [111], [112], [113]. Thus, it must offer every opportunity to reach saturation of error causes. The audit also has to report the CFCs context in terms of amenities and local CF Patient software, including the nature (manual or automated) of the interface with the Patient Registry software, as there might be explanations regarding the quality of the data transmitted to the Registry.

The patients were selected in each CFC according to the following steps:

1. Through an email sent to the Registry administrator, the head physician at the CFC authorizes the CRA to access the patient data undergoing the quality audit.

2. The Registry administrator prints the list of patients at the CFC forming part of the population enrolled in the PHARE-M Performance research project, on which the personal data to be audited appear as they appear in the Registry.
3. The list is sent via a secure Internet connection to the head physician at the CFC and the CRA simultaneously.
4. From this list, the CRA randomly selects 20 patients, one by one, traveling through the different age ranges, until the number of 20 is reached (cf. Table I): these patients' records will be audited in the period of time allotted to the CRA (8 hours/CFC).
5. The list of patients selected is sent to the head of the CFC so that they may prepare the corresponding patient records for the CRA visit.

Procedure for the on-site audit

1. During the on-site visit, the CRA uses the list of patients from the Registry to record the progress of the audits conducted, indicate the discrepancies in values observed, and write possible corrections that will be submitted to the CFC head physician.
2. The audit is conducted on two levels:
    - On the CFC internal level: comparing the PFTs and source files, the information reported in the patient record, and the information appearing in the consultation report;
    - On the Registry level: for each patient, assessing the data in the patient records for all their visits in the year, identifying the visit at which the best FEV1 L value for the year has been measured and comparing it with the data appearing in the Registry for that patient.

   This dual audit identifies on one hand measurement *discrepancies* and on the other hand *anomalies* for the selection of the FEV1 L value and the associated weight/height values.

3. Once the audit has been completed, the list containing the requests for correction in the Registry is printed out by the CRA and presented to the head physician for signing preceded by the statement "I acknowledge that I have read the requests for corrections to be made to the Registry. Unless I specify otherwise within a period of one month, I authorize the Registry administrator to make the necessary corrections." A copy of the document is left to the physician on the same day.
4. At the end of the audit, a report of the visit by the CRA is given to the head physician. This report includes: an ethnographic assessment of the presentation of the patient records (classification, storage, and retention), difficulties encountered during the audit, factors having facilitated the work of data control, and recommendations for improvement.
5. After the period of one month, the anonymized list of patients with a request for correction is sent by the CRA to the Registry administrator to make corrections.

The CRA was in possession of a number of files equal to the number of CFCs visited: these files were circulated among the CRA, the Registry administration, and the CFC's physician. The means ensuring personal data security focused on file storage (on an external hard disk stored in a safe at the CRA's office) and file access audit, one per CFC (access protected by password or delivery by email accompanied by an access code delivered by SMS or telephone). The procedure for circulating data among the Registry, the Clinical Research Assistant (CRA), and the CFC received CNIL authorization [114].

Results

The fourteen CFCs underwent the data quality audit between July 2, 2014, and June 24, 2015. This section presents the results on the two levels, the CFC level and the national Registry level. The discrepancies and anomalies found are reported by type and source of data, with the frequency of occurrence.

Number of patient records audited

According to the 2013 Registry data, 1,292 patients met the inclusion criteria in the research project population for the 14 CFCs in the PHARE-M group. Among these patients, 280 records (21%) were selected from the different age ranges according to the population distribution at the 14 CFCs (see Table I). The population selected also had the same sex ratio as the study population.

For 2012, 13 patient records (5%) could not be consulted because they were archived off the CFC premises and could not be accessed within the period of time allotted for the visit. For 2013, seven records (2.5%) could not be found. Twenty-five available records could not be audited owing to a lack of time. In total, 242 records were audited (87 patients 18 years of age or older, and 155 patients under 18 years of age) (see Table II).

The audits focused on 2,455 consultation reports for the years 2012 and 2013: 754 visits concerning adults and 1,701 visits concerning children or adolescents in the two years. The number of consultations corresponds to an average of five visits per patient each year (standard deviation: 3.6 to 7.3), all age ranges combined. The 155 children/adolescents and the 87 adults whose records were audited made an average of 5.5 and 4.3 visits per year, respectively. During these 2,455 visits, 1,855 PFTs were performed to produce source files for FEV1 data (L and %) incorporated into the patient record.

Local level: Patient records at the CFCs

At the 14 CFCs, patient records were presented in the form of a hard-copy record and an electronic file. The hard-copy record contains examination documents including PFT source files. The electronic record is managed in the Hospital Information System (HIS). At 11 of the 14 CFCs, a software dedicated to cystic fibrosis is used concurrently with the HIS: Five CFCs used the MucoDoméos software, three used the Gulper software, and three used the eMuco software.

Discrepancies in the patient records (see Figure 2)

For the 2,455 consultation visits:

- In 67 cases (3%), the consultation report, in which weight and height measurements are recorded, was not found;
- In 45 instances (2%), including 43 instances for adult patients, weight was not recorded in the consultation report; weight anomalies of up to plus or minus 5 kg were identified in 22 cases (1%): these were linked to errors in entry or position of the decimal point;
- In 62 instances (2.5%), including 35 instances (1.5%) for adult patients, height was not recorded in the consultation report; height anomalies of up to 2 cm more or less were identified in 52 cases (2%): these were linked to errors in entry or position of the decimal point;
- In 55 instances (3%), FEV1 results were not recorded in the consultation report; in five instances, the FEV1 value was only that measured after a short-acting bronchodilator was taken; and in 20 instances (1%), the FEV1 value differed from that in the PFT source file.

Table III summarizes the number of discrepancies identified in the records by nature and by CFC.

Discrepancies in the PFT source files (see Figure 3)

The organization, equipment, and practices concerning PFTs vary widely from one CFC visited to the next, as illustrated by the description below.

Organization: at seven centers, PFTs are performed, and measurements are taken, at the CFC by coordinating nurses (four), physiotherapists (two), or a PFT technician (one); at the other seven CFCs, PFTs are performed in a dedicated department of the hospital by nurses

or technicians; one CFC also takes measurements during patient home visits, in which case a physiotherapist takes the measurements; and at the seven hospitals where PFT follow-up is done at the CFC, the annual review takes place in a dedicated department in five cases.

PFT frequency: at certain CFCs, PFTs are systematically performed at each visit, i.e. three to four times per year; at other centers, they are performed only once per year for patients in a "good respiratory state". Mean age at initial PFTs is 4.5 years.

Equipment: Forty-four different spirometers are used throughout the 14 CFCs visited. Six are portable devices used in consultations by two CFCs; one is used for home consultations by another CFC; 14 devices are linked to a plethysmography chamber; different brands are represented: Jaeger® (25), Medisoft (nine), EasyOne™ (four), Dyn'R (five), and Spirodoc® (one); the plethysmography chamber is systematically used at one CFC, but never used at four other centers; three CFCs do not have a plethysmography chamber; nine CFCs use both methods; four CFCs systematically use the chamber during the annual review; four use it when it is free; and one mixed CFC uses it for adults only.

Practice: patients blow into the spirometer in a seated position at two CFCs and in a standing position at four other CFCs; at eight CFCs, both positions are used depending on patient age, chambers available or patient choice.

Updated height: at the 11 pediatric or mixed CFCs, height was updated at each consultation in the spirometry software; at the three adult CFCs, height was not updated at each consultation; and at eight CFCs, height and weight measurements were taken in both PFTs and consultations;

Standards for calculating FEV1%: 21 spirometers apply the Zapletal pediatric benchmarks [115] and ECCS/ERS adult benchmarks [116]; seven spirometers only use the Zapletal benchmarks, 14 only use the ECCS/ERS benchmarks, and two centers were unable to specify the benchmarks applied in their spirometer; at three pediatric CFCs, the ECCS/ERS standard was used as a benchmark; and, using the Knudson reference equations [117], the Registry recalculates the FEV1% based on the value in liters transmitted by the center.

Discrepancies between Patient records and PFT files: beyond the variability described, the following discrepancies were found:

- Forty-nine PFT source data (2.6%) were missing in the patient records while measurements had been taken and reported in the record; however, it was impossible to audit the value reported;
- Three hundred and thirty-eight discrepancies in weight measurement (18.2%) of up to plus or minus 1 kg were found in the PFTs with respect to the value in the patient records;
- Eight discrepancies on height data of up to plus or minus 2 cm; and
- Two hundred discrepancies on height data of up to plus or minus 1 cm were found between the PFT files and the patient records (11%); these were due to multiple height measurements in a single visit or no update of height in the spirometry data.

It should be noted that:

- Discrepancies on the weight data in the PFT file have no impact on the result of the calculation of FEV1 as a percentage;
- Discrepancies on height (and sex) do have an impact on the result of the calculation of the FEV1 as a percentage.

National level: results from the control of data transmitted to the Registry

The values appearing in the Registry and the values appearing in the patient visit record with the best FEV1 in liters for the year are compared. An *anomaly* is counted each time a difference existed between the values in the Registry and the values in the patient record with the best FEV1 L. When a patient (especially a small child or infant) has not done any PFTs during the year, only the anthropometric data have to be transmitted to the Registry and must be those measured at the last visit for the year. A difference between these values is then counted as an *anomaly*.

Various rules are applied by the CFCs to transmit the spirometry data and the associated anthropometric data to the Registry. The MucoDoméos, Gulper and e-Muco software programs semi-automatically transmitted data to the Registry by selecting the data corresponding to the best FEV1 in liters, for the years 2012 & 2013. Three CFCs did not use software specific to cystic fibrosis and thus transmitted the data selected manually from their Hospital Information System.

Given the variety of procedures for transmitting data to the Registry and the ambiguity of the wording of the rule in the document accompanying the annual Registry questionnaire

for the years 2012 and 2013, variability in the selection rules has been observed, causing *anomalies*. The *anomalies* observed are often "massive" as they generally result from the procedure applied at the CFC for all their patients.

The controls showed that:

- Nine CFCs transmitted the value in liters corresponding to the best FEV1 % for the year to the Registry;
- The CFCs using the CF software for semi-automatic transmission generally reported the data corresponding to the best FEV1 in liters for the year;
- The weight and height data transmitted to the Registry by the eMuco software are those appearing in the PFT data set, while the primary data measured by the nurse are most often recorded by the healthcare providers at the CFC in the consultation data set;
- Three CFCs transmitted FEV1 values measured after short-acting bronchodilators were taken, in line with their interpretation of the rule for selecting the “best spirometry of the year”;
- At one CFC, in 2013, a replacement staff member entered theoretical FEV1 values by age and sex, instead of patients' measured values;
- At another CFC, the data transmitted to the Registry corresponded to the last FEV1 for the year in 2012 in accordance with the rule valid up to 2010;
- One CFC did not report FEV1 values in liters in 2012 (only FEV1 % value).

In summary, in 110 patient records out of the 242 audited (45%), there were *anomalies* between the FEV1 L appearing in the Registry and the value that would have been expected according to the given selection rule (see Table IV). They mainly relate to the date of the venue not corresponding to the visit at which the best FEV1 value in liters for the year was measured. Other causes of *anomalies* derive from the conditions of FEV1 measurement (transmission of the value after bronchodilator), the absence of data (no transmission of FEV1 L) or an error in the value transmitted (theoretical value). Among those 110 patient records presenting *anomalies*, 33 were children aged 6 to 17 for whom further investigations were made.

Causes of anomalies concerning the data transmitted to the Registry in children

We decided to analyze the causes of the anomalies observed between the Patient Record data (PFT source) and the Registry data in 33 children records (out of the 120 children records controlled) and investigate the potential deviation of FEV1 % value resulting from this.

Impact of growth on FEV1 L and %

For the 33 children aged 6 to 17, table Va shows that:

- For all of them, the visit with the "best FEV1 L" is later in the year than the visit with the "best FEV1 %"
- All have grown up between the 2 visits, with height increases of up to 6 cm (average growth = 3.1 cm; median growth = 3.0 cm)
- All have increased their FEV1 L between the 2 visits, in parallel to their height increase, from 0,01 L to 0,49 L (average = 0.1 L; median = 0.06 L)
- All have decreased their FEV1 % between the 2 visits, in parallel to their height increase, from – 0.2% to – 11% (average = - 4%; median = - 3%).

For these 33 children, the choice of selecting the visit with the best FEV1 L or the visit with the best FEV1 % does have an impact on the value of FEV1 L transmitted to the Registry (average = 0.1 L; median = 0.06 L).

Impact of various standard references for the calculation of the value FEV1 %

For the 33 children aged 6 to 17, table Vb shows that:

- the different selection rules applied were either the best FEV1 L or the best FEV1 % or another value corresponding to an undetermined rule. Some standardization appeared when a CF software is used to transmit the data to the Registry;
- the Registry applies the Knudson reference equations to the FEV1 L value transmitted by a CFC: a discrepancy then appeared between the FEV1 % in the Registry and the FEV1 % in the patient record when local standards used were different from the Knudson reference values, even though the FEV1 L was identical in the 2 files.

For the 33 children in the age range of 6-17 years old, the deviation between the FEV1 % value appearing in the Registry and the FEV1 % value appearing in the Patient Record of

the visit with the “Best FEV1 L”, is an average deviation of + 4.25% (median deviation = + 4%; min. = -9.30%; max. = + 16.9%).

Standardization of Data transmitted to the Registry with the use of a CF Software

The example of 4 pediatric CFCs equipped with the 3 different CF software programed to select the visit at which the “Best FEV1 L” had been measured, shows (Table Vc):

- standardization of the data selection in these CFCs
- deviations on FEV1 % value remained when the local standard reference in the CFC was different from the Knudson reference value used in the Registry

Conclusion and Discussion

Conclusion

This first on-site quality audit of the data transmitted to the French CF Registry showed a great deal of diversity in terms of organization, information circuits, equipment, and practices concerning taking measurements as well as in the rules applied for selecting the values to be transmitted to the Registry. While *discrepancies* in the recording of each value for weight, height, or FEV1 in liters in the patient records are observed in less than 5% of cases – except for the height in PTF files, discordant in 11% of the 1855 PFTs, *anomalies* between the data appearing in the Registry and the data from the patient records occur for a great number of patients in the sample controlled (45%). The rule of selecting the annual visit when the “best spirometry in the year” was measured, enacted from the 2011 survey, was not homogeneously applied at the 14 CFCs for the years 2012 and 2013, except when the CF Software selected semi-automatically the visit at which the “Best value L” was measured. The use of local standard references instead of the Knudson reference value used in the Patient Registry explains additional deviations in the FEV1 % value between the Registry and the patient records when the FEV1 L values matched.

For the 33 children (age range 6-17 years) whose records presented anomalies, an average deviation of the FEV1 % value by +4.25% (median = +4%; min. = -9.3%; max. = +16.9%) was observed between the Patient records and the Registry. The impact of the applied selection rule appears to be more critical in this sample as it was observed that respiratory function as reflected by the FEV1 % value continuously declined during the 2 years 2012 and 2013 while these children were growing in height, even though their FEV1 value in L had increased.

Question of reliability of the primary endpoints

The reliability of the primary endpoints used for the research program, FEV1% and BMI, as appearing in the Registry and calculated from the data transmitted by the CFCs was the subject of this on-site audit conducted at 14 CFCs. The discrepancies and anomalies observed may challenge the interpretation of the results for the research program quantitative analysis, which intends to compare the trend of these indicators from 2012 to 2015 between two groups of patients, the PHARE-M group and the Control group. Even though the design of the audit only served the goal of reaching saturation of error causes and not statistical significance, it appeared that on a sample of children and adolescents representing about 33% of the total patients with anomalies, various causes could lead to an average deviation of + 4.25% in the value of FEV1% between the Registry and the Patient records (median deviation = + 4%; min. = -9.30%; max. = + 16.9%).

In general, registry data quality, unlike clinical research study data, is rarely audited at the source. However, one intend to use these data for epidemiological studies, phase 4 clinical studies, and care quality improvement follow-up, as well as for national or international comparisons. The European CF Patient Registry Working Group recognizes the current difficulties and limitations in the interpretation of variations in indicator values across the countries and emphasizes that their transparency may increase their reliability. In France, avenues for improvement have been identified on measurement processes and staff training, organization of data and suitable use of patient information systems, and clear definition and strict application of rules for transmitting data to the Registry.

Measurement recommendations and staff training

The best practice consists in measuring patient weight and height only once per consultation, at the start, applying the international recommendations for measurement [94]. The results of these measurements should be reported in the PFTs. For adults, the height check is to be done at least once per year, and the weight check at each consultation. A patient's self-report of their weight and height cannot replace measurement under the required conditions. Multiple measurements by various professionals during a single visit increases the risk of error and cannot compensate for the failure to provide a single measurement done by the required people under the required conditions. The safest way to organize height and weight measurements is to perform them all in one place equipped with devices (height gauges and scales) compliant with the standards and endowed with staff trained in measurement rules and regular monitoring of the devices.

Since the conditions for performing PFTs depend on the patient's circuit in the hospital, it may be unrealistic to aim to harmonize the organization of PFTs for all CFCs. The most reliable way to organize PFTs would be to ensure that the devices used in different places

are compliant with the standard Reference, pediatric or adult, calibrated, and regularly monitored under the responsibility of the PFT department, and are used by trained staff. Knudson reference values should be generalized.

Organization of data and suitable use of patient information systems

The quality of the organization of the data in the patient record, whatever the format (hard-copy or electronic), is a criterion of the French program of Financial Incentives for Quality Improvement (IFAQ) of patient management. The challenge of the CF electronic patient record is that of taking into account multiple interventions by various professionals in the course of the CF patient's visit while organizing the data collected in a database such that a given piece of information is recorded in a single structured field. Within the framework of an outpatient visit, the weight and height values measured must be entered just once by a qualified professional, and must be available in read-only real-time mode in the later steps of the patient's circuit. These electronic records have the advantage of including immediate consistency checks and warnings. In the future, it would be important to conduct a quality audit of software use.

Clear definition and strict application of rules for transmitting data to the Registry

Since 2011, the PRSC has recommended transmitting the data — FEV1 L, weight (kg, g), and height (cm, mm) — corresponding to the "*best spirometry in the year*". However, this recommendation was ambiguous, as it did not specify if it should be the best value for FEV1 in liters or as a percentage, and in growing individuals, the best value for FEV1 in liters most often does not correspond to the best value as a percentage. In our audit, selection in practice varied by CFC in 2012 and 2013.

The European Registry takes into account the FEV1 L value corresponding to the best FEV1 % value for the year[118]. This recommendation was not clearly adhered to in France as the CF patient software programs automatically selected the best FEV1 L value for the year. This issue is to be addressed by the PRSC. Any change in the selection rule would need to be largely explained and implemented in the software used by the CFCs to ensure its application and avoid misinterpretations.

The checks showed that standardization is achieved through automation with software programs selecting suitable data. The annual process of transmitting data to the Registry should be under the responsibility of an identified and trained person at each CFC. An audit

of the practices at each site should identify the operations required to check missing or aberrant data and validate the data before transmission.

Just one out of the 14 centers audited did not have any discrepancy in the data. At this center, measurements are taken only once. They are recorded in a software dedicated to cystic fibrosis patients which selects automatically the data to be transmitted to the Registry. The Knudson reference equations are applied in the CFC software. Finally, a person knowledgeable about the instructions is responsible for validating the data before transmitting them to the Registry.

444 **Table I** — Distribution by age range of the selected patients

Age ranges	Patients selected	% of patients selected	Study population	% of study population
0-2 years	28	10	86	7
3-6 years	34	12	163	13
7-12 years	67	24	307	24
13-17 years	52	19	258	20
18-25 years	77	27	328	25
26-35 years	15	5	105	8
> 35 years	7	3	45	3
Total	280	100%	1292	100%

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446 **Table II** — Distribution of the number of records audited by CFC

CFC	1	2	3	4	5	6	7	8	9	10	11	12	13	14	Total
Study population	108	86	71	77	70	163	208	67	108	57	88	41	61	87	1,292
Records selected	20	20	20	20	20	20	20	20	20	20	20	20	20	20	280
Inaccessible records for 2012	3	7				1		2							13
Inaccessible records for 2013		2						3			2				7
Number of records available for audit	17	13	20	20	20	20	20	17	20	20	20	20	20	20	267
Number of records audited	**12**	**10**	**13**	**18**	**20**	**20**	**20**	**17**	**14**	**20**	**18**	**20**	**20**	**20**	**242**

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448 **Table III**— Distribution by CFC of the types of discrepancies in the patient records

CFC	1	2	3	4	5	6	7	8	9	10	11	12	13	14	Total	%
No. of consultations audited	168	130	148	162	172	191	173	231	159	219	162	197	151	192	2,455	100
Report missing				2	25	1	6	10	15		8				67	2.7
Date missing from report	1	1													2	NS
PFT source file missing	3	1	14	2	6	3	3	2			15				49	2
Weight missing from report	2	1	4		16	18					3	1			45	1.8
Weight anomaly in patient record		1		1	1	18				1					22	0.9
Height missing from report	7	3		7	35				3		6	1			62	2.5
Height anomaly in patient record	2	3	3	2		11	1	13	7	2			4	4	52	2.1
FEV1 missing from report	8	13	7		4				5			18			55	2.2
Discrepancy in FEV1 (L) between report and PFT	2		3		3						3	3	1	5	20	0.8
FEV1 after bronchodilator only in report	3				1			1							5	0.2
Number of PFTs performed	128	49	147	104	173	177	149	144	83	149	147	102	68	236	1,855	100
Discrepancy in weight in PFT file	2		16	30	95	97		4	11	2	19	12	18	32	338	18,2
Height anomaly in PFT file			3		2			1					2		8	0,4
Discrepancy in height between PFT file and patient record		30		15	9	80	4	9	6	2	18	13	7	7	200	10,8
Discrepancy in gender in PFTs					1										1	NS

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450 **Table IV**— Discrepancies between the patient record data and the Registry data

CFC	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**	**11**	**12**	**13**	**14**	**Total**	%
No. of records audited	12	10	13	18	20	20	20	17	14	20	18	20	20	20	242	100
No. of records with anomalies	9	3	8	6	10	0	17	4	4	15	10	11	8	5	110	45
Selection of visit date	7		3	6	8		9	5	5	9	8	11	5	2	78	
Weight	4		4	6	11		9	5	4	9	7	11	4	2	76	
Height	3		2	5	1		16	3	5	8	7	8	6	2	66	
FEV1 (L)	4	3	3	6	8		23	5	5	28	7	11	5	4	112	
FEV1 (L) not transmitted	1														1	

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Table Va – Growth impact on FEV1 L and % for the 33 children presenting a variance btw Patient Record (PFT source) and Registry

33 Patients < 18 y.o.			Data of the Visit "Best FEV1 L"					Data of the Visit "Best FEV1 %"					Deviation between the 2 Visits		
n°	Year of Birth	Gender	Date visit	Height	Weight	FEV1 L	FEV1 %	Date visit	Height	Weight	FEV1 L	FEV1 %	Height	FEV1 L	FEV1 %
1	2001	F	nov-13	141	29	1,72	89	janv-13	135	25	1,65	96	6	0,07	-7
2	2005	F	oct-13	127	26	1,58	109	mars-13	124	24	1,53	113	3	0,05	-4
3	2005	H	déc-12	125	23	1,47	91	sept-12	122	22	1,37	102	3	0,1	-11
	2005	H	déc-13	130	25	1,54	96	oct-13	128	25	1,51	98	2	0,03	-2
4	2002	F	oct-12	127	23	1,08	72	févr-12	124	20	1,07	76	3	0,01	-4
5	2007	F	nov-13	109	19	1	104	juin-13	108	19	0,99	107	1	0,01	-3
6	1996	F	mai-13	160	57	2,84	84,8	févr-13	160	57,3	2,58	94	0	0,26	-9,2
7	2001	F	juil-13	159	43	2,5	93	janv-13	153	42,3	2,32	96	6	0,18	-3
8	2001	H	oct-12	138,5	31,2	2,12	102,7	juin-12	134	30,4	1,93	110	4,5	0,19	-7,3
9	2002	F	déc-12	130,5	27	1,89	105	juin-12	127	26,6	1,66	114	3,5	0,23	-9
10	2004	F	déc-12	131	26	1,33	77	juin-12	128	25,2	1,2	81	3	0,13	-4
11	2004	F	sept-12	128,5	25,1	1,55	93	juin-12	127	25,5	1,45	104	1,5	0,1	-11
	2004	F	sept-13	133	26,6	1,53	83,7	nov-13	133	27,5	1,39	84	0	0,14	-0,3
12	1996	H	déc-13	177,8	63,5	4,28	108	avr-13	174,5	60,3	4,16	110,5	3,3	0,12	-2,5
13	2001	F	sept-12	126,5	24,6	1,19	83,2	nov-12	125,5	24,8	1,17	83,5	1	0,02	-0,3
14	2001	H	avr-13	143,4	31,5	1,87	87,2	mai-13	142	32	1,86	89,5	1,4	0,01	-2,3
15	2003	H	nov-12	135	27	1,58	87,6	août-12	133,5	25,1	1,55	88,8	1,5	0,03	-1,2
	2003	H	août-13	138,5	28,3	1,58	81,7	janv-13	136	27,6	1,52	82,9	2,5	0,06	-1,2
16	2006	F	sept-12	115	18,4	0,88	79,7	mars-12	112	17,7	0,87	84,3	3	0,01	-4,6
17	2001	H	déc-13	133	27,9	1,08	62,5	avr-13	130	23,9	1,04	64,4	3	0,04	-1,9
18	2009	H	nov-13	109	16,7	0,87	89,5	juil-13	106	15,6	0,84	93,6	3	0,03	-4,1
19	2004	H	oct-13	132	27	1,54	91,3	avr-13	130	27	1,48	91,6	2	0,06	-0,3
20	2008	F	nov-13	111	16	0,89	89,1	janv-13	104,8	15,1	0,8	92,4	6,2	0,09	-3,3
21	1998	F	juin-13	158	56	2,84	107,5	sept-13	157	55	2,81	108,2	1	0,03	-0,7
22	2001	F	déc-12	150	40	1,9	83,1	févr-12	142	36	1,77	89,6	8	0,13	-6,5
	2001	F	avr-13	151,3	46,2	2,15	92,3	janv-13	150	44	2,14	93,4	1,3	0,01	-1,1
23	2007	H	oct-13	113	19	0,98	91,3	févr-13	108	17	0,96	101,4	5	0,02	-10,1
24	2001	F	avr-12	136	28	1,36	77,5	janv-12	133	26,4	1,31	79,4	3	0,05	-1,9
25	2002	F	oct-12	136	30	1,51	86,5	mars-12	132,8	28,4	1,46	88,7	3,2	0,05	-2,2
26	1998	F	nov-13	152,4	37,5	1,76	74,1	mai-13	150,3	35,2	1,7	74,3	2,1	0,06	-0,2
27	1998	H	déc-12	178	57	3,23	81	août-12	175	54	3,1	81,9	3	0,13	-0,9
	1998	H	déc-13	184	64,6	3,69	84,3	mars-13	180	59,5	3,5	85	4	0,19	-0,7
28	2004	H	avr-12	119	1	0,8	73,2	oct-12	122	1	0,67	76,24	3	0,13	-3,04
29	1994	F	nov-12	168	57	3,19	93,4	avr-12	168	53	2,94	94,2	0	0,25	-0,8
30	2001	H	déc-13	150	48	2,02	82,9	janv-13	142	1	1,82	87,3	8	0,2	-4,4
31	1997	H	août-12	160	63	2,94	100,2	janv-12	159	63	2,9	100,6	1	0,04	-0,4
32	1999	H	oct-12	140	35	2,24	112,1	janv-12	136	32	2,21	120,5	4	0,03	-8,4
	1999	H	avr-13	143	39	2,92	115	janv-13	141	37	2,43	119,2	2	0,49	-4,2
33	2005	F	oct-12	123	1	1,21	91,1	janv-12	118	1	1,18	99,8	5	0,03	-8,7
												Average	3,1	0,1	-4,0
												Median	3	0,06	-3

Table Vb – Deviation in FEV1 L and FEV1 % btw Patient Record (PFT source – visit when "Best FEV1 L" measured) and Registry for the 33 children among 120 controlled ones.

33 Patients < 18 y.o.			"Best VEMS L"		"Best VEMS%"		Data in the Registry		Selection & Reference to calculate FEV1 %	△ Registry v/s Expected	
n°	Year of Birth	Gender	FEV1 L	FEV1 %	FEV1 L	FEV1 %	FEV1 L	FEV1 %		FEV1 L	FEV1 %
1	2001	F	1,72	89,00	1,65	96,00	1.97	100,00		0,25	11,00
2	2005	F	**1,58**	109,00	1,53	113,00	1.58	111,00		0	2,00
3	2005	H	**1,47**	91,00	1,37	102,00	1.47	104,00	**CFC 1** Best FEV1 L and Zapletal Reference	0	13,00
	2005	H	**1,54**	96,00	1,51	98,00	1.54	95,00		0	-1,00
4	2002	F	**1,08**	72,00	1,07	76,00	1.08	74,00		0	2,00
5	2007	F	**1,00**	104,00	0,99	107,00	1,00	105,00		0	1,00
6	1996	F	2,84	84,80	2,58	94,00	2.43	89,00		-0,41	4,20
7	2001	F	2,50	93,00	2,32	96,00	2.41	93,00	**CFC 2**: Best FEV1 % ; CFC with Zappletal87; Annual Review in Pnumonology with Jaeger cabin & ECCS93 Quanjer Reference (adult); at Home with Spirodoc	-0,09	0,00
8	2001	H	2,12	102,70	**1,93**	**110,00**	1.93	110,00		-0,19	7,30
9	2002	F	1,89	105,00	**1,66**	**114,00**	1.66	114,00		-0,23	9,00
10	2004	F	1,33	77,00	**1,20**	**81,00**	1.2	81,00		-0,13	4,00
11	2004	F	**1,55**	**93,00**	1,45	104,00	1.55	93,00		0	0,00
	2004	F	1,53	83,70	**1,39**	**84,00**	1.39	84,00		-0,14	0,30
12	1996	H	4,28	108,00	4,16	**110,50**	.	111,00		-4,28	3,00
13	2001	F	1,19	**83,20**	1,17	83,50	.	83,00	**CFC 3**Transmission of FEV1 % only; ECCS93 Quanjer Reference in the spirometer at the CFC	-1,19	-0,20
14	2001	H	1,87	**87,20**	1,86	89,50	.	87,00		-1,87	-0,20
15	2003	H	1,58	87,60	1,55	**88,80**	.	89,00		-1,58	1,40
	2003	H	1,58	**81,70**	1,52	82,90	.	82,00		-1,58	0,30
16	2006	F	0,88	**79,70**	0,87	84,30	.	80,00		-0,88	0,30
17	2001	H	1,08	**62,50**	1,04	64,40	.	63,00		-1,08	0,50
18	2009	H	0,87	89,50	0,84	93,60	0.91	94,00		-0,04	4,50
19	2004	H	1,54	91,30	1,48	91,60	1.57	97,00		-0,03	5,70
20	2008	F	**0,89**	89,10	0,80	92,40	0.89	106,00		0	16,90
21	1998	F	**2,84**	107,50	2,81	108,20	2.84	108,00		0	0,50
22	2001	F	**1,90**	83,10	1,77	89,60	1.9	89,00		0	5,90
	2001	F	2,15	92,30	2,14	93,40	2.16	93,00	**CFC 4** Best FEV1 L after bronchodilator and Zapletal Reference	0,01	0,70
23	2007	H	0,98	91,30	**0,96**	101,40	0.96	101,00		-0,02	9,70
24	2001	F	1,36	77,50	1,31	79,40	1.38	84,00		0,02	6,50
25	2002	F	1,51	86,50	1,46	88,70	1.52	92,00		0,01	5,50
26	1998	F	1,76	74,10	1,70	74,30	1.85	78,00		0,11	3,90
27	1998	H	3,23	81,00	3,10	81,90	3.33	88,00		0,1	7,00
	1998	H	3,69	84,30	3,50	85,00	3.94	90,00		0,25	5,70
28	2004	H	**0,80**	73,20	0,67	76,24	0.8	63.9		0	-9,30
29	1994	F	3,19	93,40	2,94	94,20	3.18	102,00	**CFC 5** Random Selection of Best FEV1 L or Best FEV % maybe depending on the year and Zapletal Reference	-0,01	8,60
30	2001	H	2,02	82,90	**1,82**	87,30	1.82	87.3		-0,2	4,40
31	1997	H	2,94	100,20	**2,90**	100,60	2.9	100,00		-0,04	-0,20
32	1999	H	2,24	112,10	**2,21**	120,50	2.21	120,00		-0,03	7,90
	1999	H	2,92	115,00	2,43	119,20	2.69	126,00		-0,27	11,00
33	2005	F	1,21	91,10	**1,18**	99,80	1.18	99.8		0,03	8,70
									Average	-0,36	4,25
									Median	-0,02	4,00

Table Vc – Standardization of Data transmitted to the Registry with the use of CF Software.

Patients < 18 y.o.			"Best VEMS L"		"Best VEMS%"		Data in the Registry		Selection & Reference to calculate FEV1 %	△ Registry v/s Expected	
n°	Year of Birth	Gender	FEV1 L	FEV1 %	FEV1 L	FEV1 %	FEV1 L	FEV1 %		FEV1 L	FEV1 %
1	1997	H	**4,16**	108,00	4,11	110,00	4.16	108,00		0,00	0,00
2	2001	F	1,72	89,00	1,65	96,00	1.97	100,00		0,25	11,00
3	2005	F	**1,58**	109,00	1,53	113,00	1.58	111,00	**CFC 1**: Best FEV1 L and Zapletal Reference **Software Gulper**	0	2,00
4	2005	H	**1,47**	91,00	1,37	102,00	1.47	104,00		0	13,00
	2005	H	**1,54**	96,00	1,51	98,00	1.54	95,00		0	-1,00
5	2002	F	**1,08**	72,00	1,07	76,00	1.08	74,00		0	2,00
6	2007	F	**1,00**	104,00	0,99	107,00	1,00	105,00		0	1,00
35	2000	F	**2,43**	101,00	2,39	103,00	2.43	101,00	**CFC 6**: Selection of FEV1 L and Knudson Reference **Software Mucodoméos**	0	0,00
36	1999	F	**2,48**	91,00	2,27	92,00	2.48	91,00		0	0,00
	1999	F	**2,64**	86,00	2,41	87,00	2,64	86,00		0	0,00
37	2012	H	**1,21**	67	1,18	69	1.21	67	**CFC 7**: Selection of FEV1 L and Knudson Reference **Software e-muco**	0	0
38	2004	H	**1,69**	93	1,65	95	1.69	93	**CFC 8**: Selection of FEV1 L and Knudson Reference **Software : e-muco**	0	0
39	2007	F	**0,92**	89	0,72	93	0.92	89		0	0
40	2003	H	**1,76**	90	1,57	93	1.76	90		0	0
41	2005	H	**1,43**	90	1,25	93	1.43	90		0	0
42	2001	F	**2,25**	108	2,21	112	2.25	108	**CFC 9**: Selection of FEV1 L and Knudson Reference **Software Mucodoméos**	0	0
43	2004	F	**1,52**	107	1,51	114	1.52	108		0	0
	2004	F	**1,52**	98	1,5	106	1.52	99		0	0

Figure 1 — Location of the 14 CFCs involved in PHARE-M between 2011 and 2013

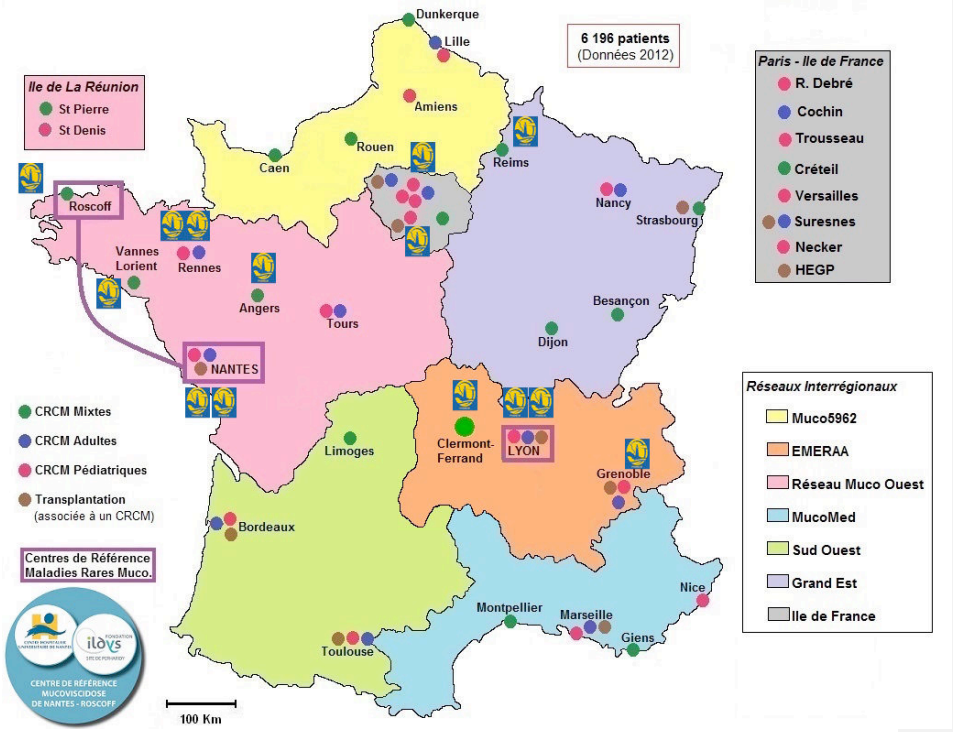


Figure 2 — Discrepancies observed in the patient records

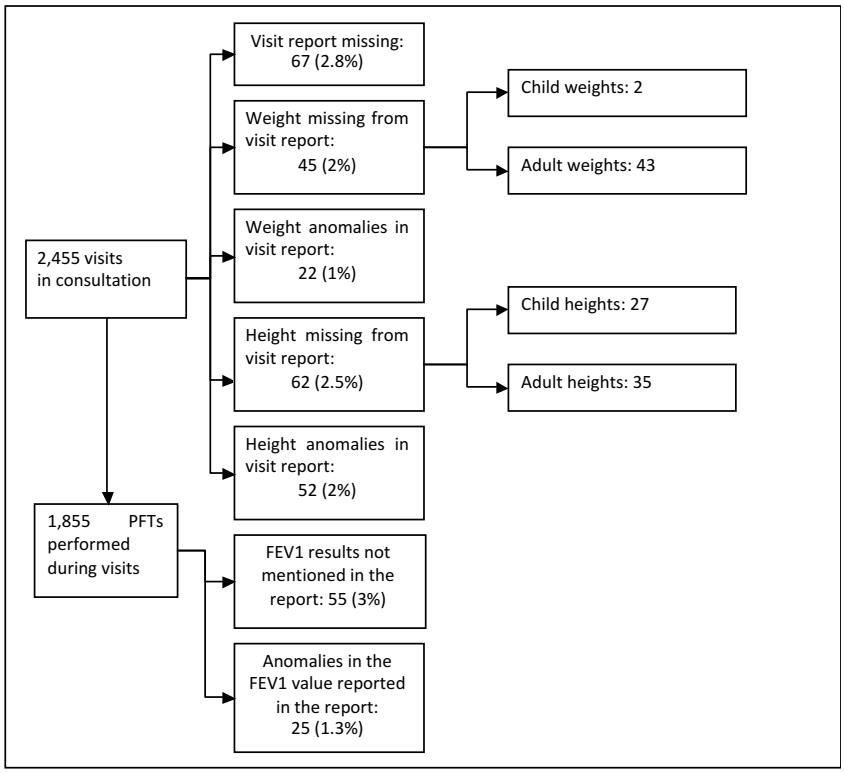
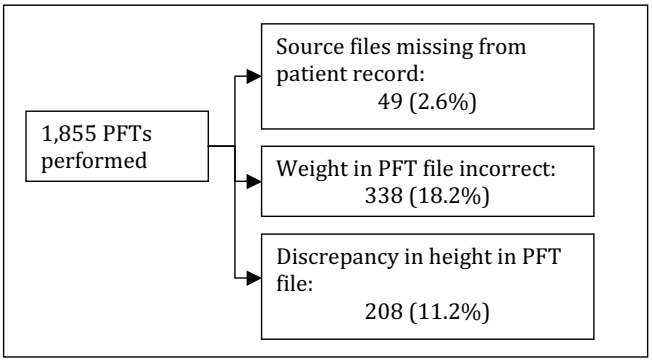


Figure 3 — Discrepancies found in the PFT source files



Lessons from patient and parent involvement (P&PI) in a Quality Improvement Program in Cystic Fibrosis care in France

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Abstract

Introduction

Quality Improvement Programs (QIP) in cystic fibrosis (CF) care have emerged as strategies to reduce variability of care and of patient outcomes among centres facilitating the implementation of Best Practices in all centres. The US CF Foundation developed a Learning and Leadership Collaborative program which was transposed in France in 2011. Patient and parent involvement (P&PI) on the local quality teams (QTs) is one dimension of this complex intervention. The conditions and effects of this involvement needed to be evaluated.

Method

In all settings, patients and parents were recruited by their centre care team. They were trained to QI method and tools and contributed their own expertise to improve the process of care. This involvement has been analyzed in the frame of the whole process evaluation. Observations and interviews conducted during the course of the first PHARE-M [**] training year explored the motivations of the patients and parents to participate and the vision of the health care teams. A research study was carried out after three years with the patients/parents and the professionals to assess the PHARE-M's effectiveness using a questionnaire to report their opinions on various components of the program, including their experience of P&PI. Responses were analyzed in view of identifying consensus and dissensus between the two groups.

Results

At the introduction of the program, P&PI was an opportunity for healthcare providers to reflect on their conceptions of these individuals both as patients and as healthcare system users. Curiosity about the teams' functioning, the various center organizations and outcomes led patients to overcome their initial barriers to participation. Seventy-six people including 12 patients/parents from the 14 pilot centres responded to the questionnaire after 3 years. Consensus between professionals and patients/parents was high on most items characterizing the performance of the QIP, QT effectiveness and QT functioning. Patients, parents and professionals agreed on the main characteristics of care such as an optimized organization, multidisciplinary care and patient-centredness. Regarding the use of patient electronic records, the use of care guidelines or the organization of support in the patient community, responses were not consensual amongst patients/parents and a source of dissensus between the two groups. All agreed that the PHARE-

[**] Programme Hospitalier d'Amélioration des Résultats et de l'Expertise en Mucoviscidose – A hospital-based program for improvement of results and expertise in cystic fibrosis care

M organization created good conditions for their involvement. In the end, both groups agreed that it was difficult to attribute the paternity of some changes specifically to any member in the team.

Discussion

Success factors for patient/parent long-term involvement in QIP have been identified. Answers to questions raised by the stakeholders about the feasibility, efficiency and usefulness of P&PI in PHARE-M could be given but new questions arose about the sustainability of continuous quality improvement over time. Perspectives such as an educational framework to develop the skills and behaviors of professionals engaged in collaborative practice with patients and families and large patient experience surveys could be used to capture patients' experience of care in the improvement work.

Key words:
Quality improvement, patient involvement, cystic fibrosis.

Introduction/Background

Patient involvement in quality of care improvement is discussed in various ways depending on the perspective and the point of care delivery.

Regarding self-management of care, strategies have been developed and evaluated to inform, educate, and involve patients in their direct care [119]. A new model of care for persons with chronic diseases has been conceptualized that focuses on their experience and knowledge, and endeavors to shift from paternalism to a care partnership [120 ;121]. Formalized processes such as shared decision making have been developed to support patient engagement in their own options for care [122;123]. In several countries, the movement to empower chronically ill patients has given rise to specific trainings to involve them in mentoring or in peer-to-peer programs in order to support other patients with the disease [124]. Experience with patients as teachers at schools of medicine or interprofessional healthcare programs is ongoing [125;126;127].

Quality of care in hospital settings was defined by the US Institute of Medicine in 2001 as clinical effectiveness, safety and patient centredness [128]. Clinical effectiveness is generally viewed as too technical to accommodate patient contributions and the usefulness of patient surveys in assessing medical quality of care remain debatable [129]. However, it is widely accepted that patients may make significant contributions to non-clinical aspects of care [130]. Many opportunities have been identified for patients to contribute to the safety of the care they receive at the hospital [131]. Moreover, reporting of safety information on medical errors and adverse events through patient interviews or surveys may also aid in identifying failures in every stage of the care process, from diagnosis to medication or clinical services [132]. Therefore, patients are recognized as being capable of contributing substantially to safety in the care by identifying care factors that potentially lead to harm or helping to learn from an incident to avoid it in the future [133]. Beyond matters of safety, the involvement of patients or their representatives in the organization of hospital care is usually associated with activities related to planning services, designing processes or assessing quality management. Groene and Sunol proposed a conceptual framework for patient involvement in quality management

comprising 5 stages: criteria development, process design, quality committees, improvement projects and discussion of quality improvement project results [134]. Their literature review and a cross-sectional survey at hospitals in the DUQuE project [135] reported experiences of patients involvement across these stages [136]: 1) on guideline development to address the needs of chronically ill patients as well as aspects of continuity of care and integration of service; 2) in assessing care preferences and designing process through surveys, focus groups and observations ; 3) in regular formal meetings to ensure quality and safety ; 4) in establishing a partnership with the QI team to plan and deliver a QI intervention in a series of plan-do-study-act (PDSA) cycles ; 5) more rarely in discussing quality improvement project results.

The history of cystic fibrosis (CF) care has been one of continuous improvement, led by the worldwide combined efforts of patient organizations, researchers and clinical teams. Therapeutic advances associated with the implementation of CF specialized care centres have brought about a dramatic increase in life expectancy and quality of life for people with CF. In the new century, Quality Improvement Programs (QIP) have emerged as new strategies to reduce variability in care as well as in patient outcomes across centres facilitating implementation of Best Practices in all centres. In this rare disease, QI is driven by comparisons of patient outcomes between national patient registries at national and centre levels [137]. Since the 2000s, the US CF Foundation and the Dartmouth Institute have developed a CF Learning and Leadership Collaborative (LLC) program to accelerate the improvement of CF care across the US centres [138].

France is a country of major prevalence of this genetic disease with 6,585 patients recorded in the national Registry in 2013, 53.7% of whom were adults. Since newborn screening became generalized in France in 2002, the French CF care network has been organized into specialized CF centres (CFCs). In the frame of the second French National Plan for Rare Diseases two centres of expertise were designated in order to develop French national action plans. The US CF QIP was transposed to France by the Nantes-Roscoff centre of expertise, and the PHARE-M[††] program was launched in September 2011 through a pilot phase involving 14 centres

[††] Programme Hospitalier d'Amélioration des Résultats et de l'Expertise en Mucoviscidose – A hospital-based program for improvement of results and expertise in cystic fibrosis care

volunteer to test and adapt the method to the French CF care organization (Table I) [139]. This QI approach is innovative in France as it installs a quality improvement dynamics and culture among the health care teams focusing on disease specific care practices and patient health outcomes improvement [140] when most QI interventions are framed by the French National Health Authority certification process. PHARE-M intends to involve patients and parents on a long-term collaboration with their care teams (nearly 3 years) to take into account their experience and preferences along the successive PDSA cycles for the redesign of the care process at their centre. The attempt to establish this long-term partnership to improve the care process is part of the innovation of this QI approach in France which needed to be evaluated. Some aspects were particularly questioned from the point of view of the patients/parents and the professionals: how did they perceive the conditions in place to allow the participation of patients and parents in the program? How did the quality team's professionals perceive this participation and what were the feelings of the participating patients and parents? Is the quality of care appreciated in the same way by patients and professionals after three years of joint work? How effective were the quality teams perceived in organizing the QI work and mastering the QI method and tools to which they had been trained? How effective was the participation of all members in the discussions and in decision-making? In the end, was the contribution of patients / parents perceptible in the quality improvement work and on the results on the process of care?

The objective of this article is to report and reflect on patient and parent involvement at the 14 centres engaged in the pilot phase of the PHARE-M program from the perspective of the patients and parents and from the perspective of the professionals on the quality teams. By illustrating Groene's conceptual framework regarding *the partnership between patients and the QI team to plan and deliver a QI intervention in a series of plan-do-study-act (PDSA) cycles*, we intend to contribute to the field with our experience of patient/parent involvement in a learning and leadership quality improvement program within a rare disease network in France.

Method

We present successively the conditions set up for patient and parent involvement in the PHARE-M program then how this involvement has been analyzed, first in the evaluation of the transposition process of the US QIP to France, then in the assessment of the program's effectiveness after three years [141].

Setting: Patient and Parent involvement in the PHARE-M

The PHARE-M was developed and adapted to the French setting by the senior pediatrician director of the centre of expertise, and a parent of an adolescent with CF, an engineer by training. Both attended the quality course in The Dartmouth Institute Microsystem Academy. The parent became the teacher and coach in the QI program.

The PHARE-M is a one year training program that follows a step by step curriculum known as the Dartmouth Microsystem Improvement Ramp [142]. This curriculum consists of multiple steps described in this OJRD supplement [139] including the declaration of a theme for improvement, the identification of leverage factors and the establishment of PDSA cycles to implement changes in the care process. As many changes require two to three years to be fully implemented, post PHARE-M sessions have been organized at the request of the teams, consisting in an on-site benchmarking visit each year, allowing to review methodological points, follow up the CFCs' actions, analyze the results achieved, and prepare publications of QI experiences.

The quality team (QT) formed at each CFC involves 4 to 5 professionals from the multidisciplinary team and is led by a physician. The recruitment of a parent (pediatric program) or a patient (adult program) in the quality team is a prerequisite to engage in PHARE-M. It has been operated by the physician leader following a recruitment procedure including a list of criteria on an application form. The consent form specifies that neither their participation nor their withdrawal would have any impact on their own care or their child's care and that their participation in the QT can cease at any time they wish. One « correspondent » professional is in charge for liaising with the patient or parent to regularly share information, answer their questions and solve practical issues. When recruited, patients and parents are enlisted in the PHARE-M training sessions as QT members. They exercise the method with their

team during the face-to-face-meetings. Patient outcomes as well as key process indicators are transparently shared with them, those regarding their centre as well as those regarding the other centres involved in the training session. Patients or parents are also invited to participate in the PHARE-M web conferences every 4 to 6 weeks. Their travel fees are reimbursed by the national patient organization. They are invited at the local QT meetings which are generally hold every 2 to 3 weeks. If they can't attend these meetings, they are updated on the work done by their correspondent on the QT. All personal health information from patients included in redesigned care processes are anonymized before being discussed at any QT meetings attended by the patient or parent. Ethical rules are established in relation to the information shared at the meetings. When a patient or parent group is active at the centre, rules are defined for communication with the group.

P&PI analysis as part of the transposition process evaluation

An evaluation was requested by the leader of the Centre of Expertise as part of the transposition process of the US CF QI program to France [139]. It was conducted by a sociologist from Mines Nantes School on the PHARE-M pilot session in order to investigate requirements for a successful national roll-out of the PHARE-M, identify the possible technical or cultural barriers and propose possible adjustments to the program to adapt it to the French context.

The assessor participated as an observer in two web meetings and one Face-to-Face meeting. The assessment included becoming familiar with PHARE-M documents, interviews with a panel of professionals and the patients/parents on the QTs, the members of the national PHARE-M team, the American supervisor from the Dartmouth Institute, and visiting one CFC site. All interviews and focus groups were recorded and fully transcribed. The data was managed (coding, categorization), processed (analysis, validity) and interpreted according to the standard thematic content analysis protocol (Miles & Huberman, 2003 [143]). This was followed by manual grouping and counting within a framework for analysis with the following dimensions: process applicability (terminology, formalization, tools, remote coordination); patients and parents involvement (roles, time spent, obstacles); French national and regional coordination (roles, nature of support, mechanisms for incorporation); process adoption (perceived benefits and costs, working atmosphere, engagement, acquisitions); and effects (operation, working practices, cooperation

with partners). Results on the dimension regarding patient and parent involvement during the pilot phase PHARE-M training year are reported in this article.

P&PI analysis as part of PHARE-M effectiveness assessment after 3 years

Since the introduction of PHARE-M in France in 2011, questions were raised by the stakeholders about the effectiveness of this quality improvement program. The first evaluation concluded that effectiveness could not be assessed at the end of the first year, neither on patient outcomes nor on results of changes in the care process, but should be assessed after three years on the basis of the program's measurable effects.

A research project was drawn up by the Centre of Expertise of Nantes-Roscoff to analyze the performance of the PHARE-M program after three years (2015) at the 14 CF centres involved in the pilot phase of the program. This research project was funded by the French ministry of Health (Decision of the Call for project PRePS – Dec 2012). The aims and protocol of the broader project from which the results are drawn are described in the OJRD supplement [144]. In brief, the protocol combines a quasi-experimental evaluation of the effectiveness of the program on patient outcomes evolution over three years with a process evaluation [145]. Following a realistic approach, the latter was designed to understand what works, for whom and under which circumstances (context) [146]. To understand which dimensions of the context were critical for the effectiveness of the programme, a questionnaire was designed assembling existing validated tools when they existed and developing new tools when necessary.

- Development of the questionnaire

The questionnaire was prepared by a panel of experts (professionals and parents/patients), tested with 3 multidisciplinary teams (N=29 respondents including 1 parent and 2 patients) and reviewed by experts in Sorbonne Paris Cité University. The final questionnaire was composed of 7 chapters covering the various aspects of the organization of care and the PHARE-M effectiveness at the centres: quality of the care process, organizational culture, patient centredness, leadership, mastering of the QI process and tools, quality team functioning and patient/parent involvement.

- Studied population

Every professional in the 14 centres, including professionals belonging to the quality teams and the patients and parents involved.

- Variables

The items in five chapters were based on existing instruments validated in previous research [147; 148]. The items characterizing the chapter about quality of care were developed for this research following the 5 dimensions of the Chronic Care Model [149]: existing goals for improvement; multidisciplinary care; self-management support; support in decision making (guidelines); electronic patient records and resources in the patient community. The items of the questionnaire analyzing patient and parent involvement were developed according to the framework proposed by Carman [150] and adapted by Pomey [120] : 1) patient and parent information/activation 2) patient and parent empowerment and 3) patient and parent contribution to the QI work.

- Data collection

The questionnaire was self-administrated during 14 on site investigations conducted by a clinical research assistant. The respondents had no opportunity to discuss their answers amongst themselves. Each topic is covered by a list of assertions requiring a response on a 5 degrees Lickert scale from « completely agree », to « fully disagree » with a neutral response « don't know/no opinion ».

- Data analysis

The responses were managed using SAS and XL and were analyzed, according to the purpose, grouping different categories of respondents: professionals in the quality teams, patients and parents. During restitutions to the centre teams, reports by centres were produced to share the results and discuss new improvement goals for the care process.

To answer the questions from the point of view of the patients/parents and the professionals, the analysis of the responses on all items of the questionnaire was made for the two groups of respondents: the patient/parent group (N=12) and the professional group in the quality teams pooled for all teams and all disciplines (N=64). We first identified the items that achieved a « strong consensus » in the patient/parent group considering unanimous or nearly unanimous responses

(unanimity less one vote or unanimity less two votes; >80%) as either positive (grouping « agree » and « completely agree »), negative (grouping « disagree » and « fully disagree ») or neutral (« don't know » or « no opinion »). We then identified the items that achieved a strong consensus in the professional group (> 80% responses with either positive, negative or neutral answers). We define dissensus or consensus between the patient/parent group and the professional group using Fisher's exact test [151] (Results available on request).

The results highlighted the following categories: 1) items achieving a consensual position between the two groups of respondents (consensual positions were found always in the same sense in the 2 groups, positive (+), negative (-) or neutral (N)); 2) items achieving consensual position in the patient group only; 3) items achieving consensus in the professional group only; and 4) items achieving no consensus (NC) in either of the two groups.

Presentation of consensus/dissensus between the Patient/Parent and the Professional groups

Items category	Consensus amongst P&P	No consensus (NC) amongst P&P
Consensus amongst Professionals	1) (+,+) or (-,-) or (N,N)	3) (NC,+) or (NC,-) or (NC,N)
No consensus (NC) amongst Professionals	2) (+,NC) or (-,NC) or (N,NC)	4) (NC,NC)

Due to the small sample of patients and parents (N=12) and their affiliation to 12 different centres, variations in their responses regarding local culture, organization, leadership and the performance of the QIP achieving no consensus are mainly to be attributed to "centre effects". We did not set out to compare the responses of the patient/parent to the responses of the professionals by center.

Regulatory authorizations were granted from the Ethics Committee of the Brest University Hospital and by CNIL (DR2015040).

Results:

Results from the observations and interviews conducted as part of the QIP transposition process to France

The opinions and concerns regarding the participation of parents and patients involved in the QTs during the program training year are summarized in Table II. The following themes emerged:

- The place of the patient/parent in the healthcare system

Patient and parent involvement disrupted assigned places, led to readjustments and reinterpretations, and highlighted resilient patient and parent profiles.

- Reasons and barriers expressed by parents for participating

They stressed contributing their testimonial on their experience and sticking to merely conveying their feelings and day-to-day experiences. They were careful not to appear to teach professionals their profession.

- Reasons and barriers expressed by patients for participating:
    - **Wariness/caution** towards the care team and the medical world.
    - **Consent and curiosity** to get to know a CF setting, to better get to know the teams that they visited as their care providers.
    - **Engagement under tension** between on one hand, the desire to understand, be curious, gain autonomy and confidence, remove obstacles, and, on the other hand, the difficulty of pushing oneself to talk in front of others about one's experiences with the care of a disease that one would like to keep at a distance.
- Healthcare providers' vision of patients/parents involved in the quality teams:

Their vision of patients/parents was confronted with real patients and parents. The presence of a patient on the team called into question healthcare providers' preconceived notions and desire. Some healthcare providers recognized that they granted themselves the authority to have a particular vision of patients and parents and to talk about them, about what they believe to be patients' experience and feelings, given their in-depth knowledge of the « ill human being ». The presence and intervention of a real patient or parent in the quality team challenged their representation and some raised the question of the representativeness/validity of the speech of the patient or parent involved.

The patient or parent participation on the QTs and their presence at the PHARE-M Face-to-Face training sessions as well as at many local meetings was perceived as an opportunity for the healthcare providers to reflect on their conceptions of the patients/parents as both patients and healthcare system users. Curiosity about the teams' functioning and comparison between the various center organizations and their outcomes led patients to overcome their initial barriers and grant their consent to participate.

Results from the *PHARE-M effectiveness assessment after 3 years*

Volunteer patients and parents were recruited by all care teams after information given on the QI program and on the importance of their involvement to improve care at their centre [152]. Over the 3 years, three of them stopped their participation. One parent wanted to stop because of health worsening of her child and was replaced by another parent who happened to be a quality engineer in pharmaceuticals. One CFC stopped the program when the physician leader retired. The 3rd CFC chose to work with the parent group (as historically) and collect feedback on change actions at annual patient group meeting.

During on site investigations 140 people from the 14 CFCs completed the questionnaire, either as QT participants or as multidisciplinary team members outside the QTs. The QT respondents totaled **76 people** (54% of all respondents): **12 patients and parents** (6 patients and 6 parents) and **64 professionals**, including 56 healthcare providers and 8 non-healthcare providers (quality engineers and others). Two CFCs were unable to contact the patient or parent to ask them to complete the questionnaire. Forty-six (82%) professionals in the QTs belonged to the CF multidisciplinary "core" team (physician, nurse, physiotherapist). Psychologists and dieticians were heavily engaged in the QTs (9 people).

Quality of care at the centre

Table III presents the items that achieved consensus or dissensus among the patients/parents and the professional groups on items related to Quality of care and organizational features at the centres after three years of joint QI work.

All the items that achieved a strong positive consensus among the patients and parents also achieved a strong positive consensus among the professionals on the QTs. They were related to the following domains of the chronic care model: 1)

GOALS: the existence of improvement goals at the CFC and indicators to monitor them, 2) SELF-MANAGEMENT SUPPORT : the existence of a therapeutic education program and professionals trained to deliver it 3) MULTIDISCIPLINARY CARE: an adequate multidisciplinary team, stable over time and possessing expertise in CF care, as well as KEY PROCESSES OF CARE: an optimized clinic visit process allowing the patient to see all members of the core team and any referral professionals from various disciplines when necessary as well as an optimized process of answering telephone or email messages from patients and families 4) INFORMATION SYSTEM: the existence of an electronic patient record (EPR) system at the centre.

Items detailing patient therapeutic education in practice, as well as items regarding certain information contained in the patient record achieved no consensus neither in the patient/parent group nor in the professional group.

The patients and parents granted unanimous neutral response ("Don't know") to items regarding the use of the EPR by the team during the staff meetings and the existence of a procedure to inform professionals on updates to guidelines when the professionals showed no consensus on these items.

Three items achieved a strong positive consensus among the professionals only. They were related to the following domains of the chronic care model: 3) MULTIDISCIPLINARY CARE: the systematic review of the records of the patients who came to the CFC; 5) DECISION SUPPORT: the availability of care guidelines to all professionals and 6) COMMUNITY NETWORK: the organization of a network of professionals in the patient community for managing care at home.

Organizational features at the centre

Unanimity was achieved for items related to PATIENT CENTREDNESS, taking patient needs and requests into account and analyzing causes of complaints to prevent problems from recurring. However, no consensus was achieved with respect to using data from the patients themselves to improve services. The same results were observed for the responses of the professionals with a rate of agreement of more than 90% on the first items, and a lower rate of agreement (< 70%) on using data from the patients themselves.

A consensus was achieved both in the patient/parent and in the professional group in perceiving LEADERSHIP as driving the organization to meet patient needs and ensure safety of care. Other aspects of leadership related to the multidisciplinary team management were mostly answered by patients/parents with "Don't know". The responses of the professionals by centres, displayed along the 5 axes of "radar" graphics, also show different types of leadership across the centres.

Table III: Consensus and dissensus between the P&P and the Professional groups on Quality of care and Organizational features at the centres

Categories: Quality of care, Patient centredness, Leadership	Consensus amongst P&P	No consensus amongst P&P
Consensus amongst Professionals	**Quality of Care:** **(++)** Existence of improvement goals at the CFC and indicators to monitor them **(++)** Existence of a therapeutic education program and professionals trained to deliver it **(++)** Adequate multidisciplinary team, stable over time and possessing expertise in CF care **(++)** Optimized clinic visit process allowing the patient to see all members of the core team and any referral professionals from various disciplines when necessary **(++)** Optimized process of answering phone or email messages from patients and families **(++)** Existence of an electronic patient record system at the centre **Patient Centredness:** **(++)** Taking patient needs and requests into account **(++)** Analyzing causes of complaints to prevent problems from recurring **Leadership:** **(++)** Driving the organization to meet patient needs and ensure safety of care	**Quality of Care:** **(NC,+)** Periodic review of the records of the patients who came to the CFC, during the multidisciplinary staff meetings **(NC,+)** Availability of care guidelines to all professionals **(NC,+)** Organization of care providers in the patient community
No consensus amongst Professionals	**Quality of Care** **(N,NC)** Use of the EPR by the team during the staff meetings **(N,NC)** Existence of a procedure to inform professionals on updates to guidelines	**Quality of Care:** **(NC,NC)** Patient therapeutic education meeting patients' needs **(NC,NC)** Biology or Imaging Information contained in the EPR **Patient centredness:** Using data from the patients themselves to improve services

PHARE-M performance and QT effectiveness

Table IV presents the items that achieved consensus or dissensus among the patients/parents and the professional groups on items related to the program's performance and the QTs' effectiveness.

The perceived performance of the PHARE-M was expressed with items focusing on the experience of the respondents as members of the QTs. A strong positive consensus was achieved amongst both patients/parents and professionals regarding their satisfaction as a member of the QT and their wish to remain on a similar team working on QI. Moreover, their perception of the usefulness of the work of the team in improving care and meeting the organization's needs was unanimously positive. All stated that an ongoing quality improvement process had to be maintained to continuously improve care at the centre.

The performance of PHARE-M as a "training-action" program on this QI approach was appreciated by the respondents with items characterizing their mastery of the quality methods and tools. There was a strong positive consensus in the two groups that the PHARE-M led to a clear vision of the area on which to focus the efforts for improvement at the centre, provided a guide for organizing QI work, and enabled the team to change its way of working and analyze data to ensure that these changes represented an improvement. Both groups agreed that a specific data collection had to be established for the QI work. The others topics related to the availability of data at their centre, by the end of the program, to allow to analyze and identify problems as well as to follow the implementation of changes achieved no consensus neither in the patient/parent group nor in the professional group.

Regarding the techniques to lead changes, no consensus was achieved in both groups on PDSA cycles monitoring to implement changes through tests and evaluations before extension. The support for changes implementation from the other departments in hospital achieved no consensus among the two groups.

Table IV: Consensus and dissensus between the P&P and the Professional groups on PHARE-M perceived performance and QT effectiveness

Categories: PHARE-M performance QT effectiveness	Consensus amongst P&P	No consensus amongst P&P
Consensus amongst Professionals	**Experience on the QT:** **(++)** Satisfied with my experience as a member of the QT **(++)** Wish to remain on a similar team working on QI **QI work done by the QT:** **(++)** Usefulness of the work done by the quality team in improving care **(++)** QI work meets the organization's needs **(++)** An ongoing quality improvement process has to be maintained to continuously improve care at the centre **Mastery of PHARE-M method and tools:** **(++)** A clear vision of the area to focus the improvement efforts on **(++)** A guide for organizing the QI work **(++)** Ability to implement changes **(++)** Ability to analyze data to ensure changes were improvements **(++)** Need to set up a specific data collection for QI work	
No consensus amongst Professionals		**Mastery of PHARE-M method and tools:** **(NC,NC)** Ability of the QT to analyze variations in processes over a period of time **(NC,NC)** Availability in routine of data to analyze and identify problems **(NC,NC)** Availability of routine data collection to follow the implementation of the new processes of care **Change Management (PDSA cycles):** **(NC,NC)** Ability to conduct tests of changes with PDSA cycles and learn from the results **(NC,NC)** Support from the other hospital departments to conduct changes

QT functioning

Table V presents the items that achieved consensus or dissensus among the patients/parents and the professional groups on items related to the QT's functioning. Those items address successively QTs process strategies, decision-making in the QTs, normative management, and internal or external collaborations [148].

A strong positive consensus was achieved on the items describing **QT process strategies**: the leader's behavior reflecting the importance he/she placed on the quality team functioning well, the team receiving all information required to plan and organize its work and, the availability of enough resources and skills on the team to work properly. The process of **shared decision making** on the team was rated as highly positive with attention being paid to the contributions of each member of the team, most team members participating in decision-making, and ease for all members in suggesting ideas for change. The **normative regulation** on the QTs was rated high regarding the agreement on and achievement of the objectives of the QI project. Though consensus was achieved on the professionals group on all members focusing on achieving the same goals, there was no consensus among the patient/parent group on this item. Last, internal **collaborations** in the QTs were rated high in the two groups but no consensus was achieved on external cooperations with the other departments of the hospital.

Table V: Consensus and dissensus between the P&P and the Professional groups on QT functioning

Categories: QT functioning	Consensus amongst P&P	No consensus amongst P&P
Consensus amongst Professionals	**Process strategies:** (**++**) Leader's behavior reflecting the importance he/she placed on the quality team functioning well **(++)** Members of the team came from different backgrounds, experiences and skills **(++)** Availability of enough resources and skills on the team to work properly **(++)** Team receiving all information required to plan and organize its work **Decision Making**: **(++)** Attention being paid to the contributions of each member of the team **(++)** Most team members participating in decision-making	**Process strategies:** **(NC+)** The leader also asked the opinions of the other members of the team **Decision Making**: **(NC+)** We appreciated and built with our differences **Normative:** **(NC+)** The team members were all focused on achieving the same goals.

	(++) Ease for all members in suggesting ideas for change **Normative:** **(++)** Team members agreed on the project's objectives **(++)** The achievement of the objectives guided the activities of the members of the team. **Internal/external collaborations:** **(++)** The people I've worked with are comfortable suggesting changes and improvements	
No consensus amongst Professionals		**Normative:** **(NC,NC)** The team members did what was expected of them. **Internal/external collaborations:** **(NC,NC)** There was a lot of cooperation between the departments of the hospital.

Patients and Parents involvement in the PHARE-M

Table VI presents the items that achieved consensus or dissensus among the patients/parents and the professional groups on items related to Patient and Parent Involvement in the PHARE-M.

The first series of items concerned the selection and activation of the patient/parent recruited. There was a consensus that the presence of a patient or parent on the quality team was “a given and an asset” despite a possible lack of education or their personal problems. A strong consensus was found to recruit a patient or parent well informed regarding the QI program goals and the need for a good relationship between the team and the patient/parent involved. The development of coping skills (*knowing how to manage emotions and stress; solving problems, making decisions, and making choices; knowing how to communicate and being at ease in relationships with others; and knowing how to put oneself in the place of others)* was by consensus a requirement for the patients and parents to be recruited to the QT. These items also achieved a strong consensus among the professionals, who had a higher rate of agreement on the “required qualities” for the patient or parent to join the team. Those qualities were not explicitly stated in the questionnaire.

Three items achieved a consensus among the patients and parents regarding their empowerment for participation: the reimbursement of their travel fees, their high motivation to improve care for all – achieving a weaker consensus to improve care for themselves, and the fact that their role on the QT was conveyed to the other patients or parents followed up at the centre. Only 8 out of 12 patients/parents

agreed on the need to be knowledgeable about the disease and its management beyond the requirements of their own care – while professionals had no consensus on that need. The professionals had a higher rate of agreement on the importance of the patients and parents taking a step back and drawing general lessons from their own experience. No consensus was achieved in both groups on the need for the patient or parent involved to understand the general functioning of the hospital. Finally, the patients and parents unanimously indicated that the organization of the PHARE-M throughout France promoted their membership on QTs.

Regarding their contribution to the QI work, the two groups agreed that patients and parents could make significant contribution to the work of the quality team and that their ideas and proposals were generally taken into account. Both groups agreed that patients and parents had to participate in the local QT meetings – rather than in the national meetings, to make these contributions. No consensus was achieved in both groups on the assertion that certain decisions made by the quality teams were inspired by the patient/parent.

Table VI: Consensus and dissensus between the P&P and the Professional groups on Patient and Parent Involvement

Categories: P&PI	Consensus amongst P&P	No consensus amongst P&P
Consensus amongst Professionals	**Activation/Recruitment:** (**++**) The presence of a patient or parent on the quality team is "a given and an asset" **(++)** Importance of the information provided to the patient or parent regarding the QI program goals **(++)** Need for a good relationship between the care team and the patient/parent involved **Empowerment:** **(++)** P&P role on the QT has to be conveyed to the other patients or parents followed up at the centre **(++)** The patient or parent is motivated to improve care for all **(++)** The organization of the PHARE-M throughout France created good conditions for their membership on QTs **Contribution:** **(++)** The patient or parent participates in and contributes significantly to the work of the QT. **(++)** Their ideas and proposals were generally taken into account **(++)** The patient or parent's regular participation at team meetings at the	**Activation/Recruitment:** **(NC,+)** The patients and parents are informed regularly (annually or more often) by the team about general subjects concerning cystic fibrosis care and research. **(NC,+)** P&P must have "required qualities" to join the team **Empowerment:** **(NC,+)** P&P have taken a step back and drawn general lessons from their own experience (**NC,+)** The patient or parent is also motivated to improve his or her own management by participating in the program.

	CFC is indispensable.	
No consensus amongst Professionals	**Activation/Recruitment:** **(+NC)** Patients/parents should have developed copying skills (with the disease) **Empowerment:** **(+NC)** Reimbursement of P&P travel fees	**Activation/Recruitment:** **(NC,NC)** The patients and parents are rather familiar with general cystic fibrosis information: research, progress made, and Registry data **Empowerment:** **(NC,NC)** The participation of a patient or parent should be facilitated by the reimbursement of other expenses: child-care, lost working hours, etc. **(NC,NC)** P&P need to be knowledgeable about the disease and its management beyond the requirements of their own care **(NC,NC)** The participating patient or parent does not represent all patients **(NC,NC)** It would be necessary to include several patients or parents to ensure that more different points of view are represented **(NC,NC)** P&P need to understand the general functioning of the hospital **Contribution:** **(NC,NC)** The participation of a patient or parent on the team at French national training and information meetings is indispensable. **(NC,NC)** The patient or parent participated and contributed as much as the professionals during the French national meetings **(NC,NC)** The atmosphere of work at the QT meetings is better and more productive when the P&P is present. **(NC,NC)** The pace of work is slower when the patient or parent is present at the QT meetings. **(NC,NC)** Certain decisions made by the QT are inspired by the patient/parent.

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Discussion

Following the results of the investigations conducted with the care providers and patients/parents, we review the highlights on the instrumentality of the method to involve patients and parents in PHARE-M QIP. We then discuss the initial questions raised about this partnership during the PHARE-M program in France and propose a list of success factors which seem essential to long term patient/parent involvement in QI work in Table VII.

Highlights on the method to involve patients and parents in PHARE-M

PHARE-M quality improvement program was innovative in France in 2012 as it intends to install a culture of quality improvement in the CF care teams, focusing on patient outcomes improvement and process of care redesign. Patients and parents were involved on a long time period with the care teams at their centre to work together on quality improvement of care.

- Conditions for patient and parent recruitment

The essential selection criteria underlined by both patients/parents and professionals were a good relationship with the team, a desire to improve care for all patients and a willingness to take a step back and draw general lessons from their experience with the disease. Training on the general functioning of the hospital or the management of the disease have not been offered at recruitment and didn't appear to be a pre-requisite for participating. The professionals contributed their in-depth knowledge of the disease and its treatments to the discussions. This was made easier by the stability, expertise and experience of the team members. Extensive information on the program provided to the other patients or parents of patients followed up at the centre as well as to the hospital administration was indispensable to legitimize the participation of the patients and parents. Nevertheless, three parents stopped their participation at the end of the first year for reasons related either to the physician at the centre or to a worsening in the patient's health status. This illustrates the impact of the medical leadership on patients/parents' long-term involvement and confirms that a stable health condition on the part of the patient is a prerequisite to engage or stay in such a program [131].

- Participation at the quality improvement national training meetings

The participation of patients/parents in the national training meetings about the QI method and tools was an integral part of the program. The reimbursement of their

travel fees appeared to be mandatory to enable them to participate at these training meetings. Such participation gave all team members an equal opportunity to be trained in the quality improvement method. Given that none of the « students » had any prior knowledge of this particular quality approach, despite their different professional expertise and background, they all engaged in discussions effectively. The transparency of the outcomes from all centres involved at these meetings was another aspect of the method [139]. It provided results from the patient registry report by centre comparing patient health outcomes to identify potential best practices at some centres. Although this transparency was novel within the French CF care network, it was well accepted by the professionals and well received by the patients and parents, as it led to the choice of a theme for improvement at the centre. Condition for effective partnership between professionals and patients in QI work involved transparency of the results and the commitment to improve them [137]. Given that the goals were clear and shared from that time forward, the patients, parents and professionals were equally committed to achieving them during the program [153]. Moreover, the collaborative aspect of the program created a community of centres willing to continue sharing their work on quality improvement and their results as part of an open process of « benchmarking of practices » [154].

- Contributions made by patients and parents

The contributions made by patients and parents obviously depended on their frequent participation in the QT meetings at their centre. The experience of the patients and parents was brought to the discussions using questionnaires during the clinic visits or phone calls as well as patient shadowing during clinic visits and observation of multidisciplinary staff meetings. The joint work on these processes resulted after three years in the shared opinion of having implemented optimized processes. The patients and parents sometimes also contributed their own expertise (quality, IT, communication etc…) by « specific tasks » assigned to them depending on their wishes, availability and own expertise. Some examples were cited in the comments: a multi-purpose notebook was created to communicate with the care team about events at home, treatments prescribed and educational material ; internet surveys were developed and the results were analyzed for the QT ; a dashboard of indicators in the form of a smiley face was develop for the children to assess their care at the end of the visit; a « gazette » about the QI program was issued by

parents and adolescents; a bulletin board was created to display information about the QI project in the CFC. These contributions seem to have accelerated the QI work of the team and facilitated communication with the other parents/patients. Most often, it was ultimately difficult to attribute certain changes in the centre organization and process of care specifically to any specific team member – patient, parent or professional.

Questions raised by this partnership during PHARE-M in France

The following questions were raised by the stakeholders of the PHARE-M program, including the professionals' and the patients/parents' representatives, on the feasibility, efficiency and utility of this partnership during the program.

- **How were perceived the conditions in place to allow the participation of patients and parents in the program?**

The patients/parents as well as the professionals agreed that the organization of the PHARE-M throughout France created good conditions for their membership on QTs. All the respondents were satisfied with their experience, mostly favorable to further participation on a similar quality team and agreed with the necessity of an ongoing quality improvement process to continuously improve care at the centre. These opinions reinforce the French national PHARE-M team's belief that the program enhances the involvement of patients/parents along with their care teams to improve care at their centre. It also indicates that the participation in the program does not cause deleterious effects to the patients/parents involved, which could have come from the vision of the "defects" seen in the management of care at their centre.

Some items remained not consensual: they may be addressed through further experimentations during the next sessions of the program. They concern "the participation of a patient/parent should be facilitated by the reimbursement of other expenses such as child-care, lost working hours..."; "the necessity to include several patients or parents to ensure that more points of view are represented" and, "the need for patients/parents to understand the general functioning of the hospital". At the beginning of the program, questions about « representativeness » of the patients/parents involved were evoked. Should those involved be individuals recruited by the care teams according to the mentioned criteria or national patient organization or local patient group representatives, when they exist? Is the experience of patients/parents involved sufficient to inform QI work? Should the experience of other patients and parents be captured to complement their own?

These questions raise matters of legitimacy, democracy and responsibility. In the frame of our QI project, the legitimacy of the patient and parent involved appeared to be granted by the care team and not by a patient organization or patient group. It happened in some settings that the parent was a member of the CF local patient group but their involvement was decided upon by the care team and not requested by the patient group. Their position in the quality team did not change the rules for communication between the quality team and the patient group. It was clear that the patient or parent involved spoke to their own experience and not to that of a group of patients/parents. These questions are important and should be clarified at the meso- and macro-system level to facilitate and foster patient involvement in the quality improvement work with their care team, as it has been done for patient representation in hospital committees. Financial aspects related to the participation of the patient/parent in meetings with the care team, in particular travel fees or other allowances, could be part of this clarification.

- **How did the quality team's professionals perceive this participation and what were the feelings of the participating patients and parents?**

At the introduction of the program, barriers from professionals as well as from patients and parents had to be overcome. In the interviews, the switching of roles in parents (I come as a parent to the consultation, and in the quality group I commit myself as a user/ a designer of the process) and in patients (I come as a patient to the consultation, and I commit myself in the quality group as a user/improver) creates a tension between those positions of the patients/parents. The potential for tension arose when they didn't feel satisfied with their experience of the care delivered by the team or with the quality of communication with certain members of the team, and when they had not coped with a previous painful circumstance such as the diagnosis of CF for their child or the management of a complication of the disease. The attenuation of this tension is critical to gradually increase the involvement of parents and patients during the QIP. This attenuation was observed in the results of the investigations after three years, which lets us hypothetize that the QIP might have acted as a process of resilience for patients, parents and professionals.

A shift in the representation of care by professionals and patients/parents was observed in the course of the program towards a co-produced service which co-production is based on a mutual understanding of roles and competences, mutual

participation in communication and actions and respective responsibilities in delivering care. French teams that had previously developed a culture of patient therapeutic education and were used to partnering with patients/parents for their own care, were more favorable to patient and parent involvement in care QI work than the teams that had not. This observation, and whether the other teams have overcome their initial reluctance, will have to be further analyzed in the results by centre, as there was a high consensus after three years that "the presence of a patient or parent on the quality team is a given and an asset". Our experience confirms that the more the professionals and the patients collaborated to plan and develop services, the more this collaboration was accepted among both the professionals and the patients [155].

Upstream conditions could be created to support the participation of patients/parents in the health system, especially in quality of care improvement programs along with their care team. In Canada, a framework for interprofessional education and collaborative practice was developed to address the needs in terms of skills and behaviors for professionals engaged in collaborative practice with healthcare practitioners, patients, families and communities [156]. Six domains were identified: interprofessional communication; patient and family centered care; role clarification; team functioning; collaborative leadership; and interprofessional conflict resolution. Several assumptions underpin this framework one of them being that interprofessional practice is not innate but requires a consistent culture of learning and practice. Further reflection would be needed to refine such a framework to the French system of health continuing education and thus foster the necessary shift towards patient involvement in quality of care improvement programs [157].

- **Is the quality of care at the centre appreciated the same way by patients and professionals after three years of joint work?**

All agreed that the care team was patient centred and eager to meet patient needs and insure safety of care. After three years of joint work, the awareness of the patients and parents on care organization and processes at their centre was high – similar to that of the professionals – concerning matters relevant to them: multidisciplinary care, patient education, the clinic visit process… But their awareness on some aspects of the organization such as the information system (patient electronic record) and the management of care guidelines, remained low.

Even so, these aspects are not to remain fatally out of their attention for quality of care improvement: the impact of educating parent in care guidelines on clinician adhering to them has been demonstrated in a pediatric CF program [158] and patient-led training in medical education has had an impact on the application of safety guidelines by clinical teams [159]. In Sweden, patient electronic records have been opened to allow patients access to their health record and provide input such as the schedule of the next visit, results on health outcomes followed at home and various mailings [160]. When these matters are explicitly shared with them as part of their care, patients and parents will probably be able to contribute to improve these fields by reporting their experience and needs.

- **How effective were perceived the quality teams in organizing the QI work and mastering the QI method and tools to which they had been trained?**

The work of the teams was fostered by leadership intending to achieve high quality of team functioning as well as by a shared decision-making process and clear shared goals, and its efficacy was supported by a good command of the quality tools including the ability to measure the results – despite a more difficult appropriation of PDSA cycles as a change management tool. The absence of consensus on items regarding availability of data in routine to follow and standardize the new processes and lack of support from other departments in the hospital raise doubts about the sustainability of continuous improvement of care at the CF centre after the 3 years. In the centres where the risk is high, a new session of the PHARE-M QIP is proposed on a new theme of improvement to sustain changes over time. The recognition of the PHARE-M program as a Professional Continuous Development program by the hospital continuing education department and the associated credits facilitates the CF teams' participation.

- **Was the participation of all QT members in the discussions and in decision making effective?**

All members felt that they could participate in decision-making, that attention was paid to their contributions and were at ease in suggesting ideas for change. The goals were clear and shared, which probably channelled the discussions amongst the members of the QTs who came from different backgrounds, experiences and skills. Normative characteristics were not dominant except the emphasis on the

goals. The patients / parents' contribution was highly appreciated but changes in the organization or process of care were not specifically attributable to them.

Reflections for further experimentations and research on involving patients' views in quality of care improvement programs

Our experience of patient/parent involvement in the PHARE-M QIP raise matters in relation to the nature and extent of the patient experience incorporated in the QI work. In 2005, Bate et al defined the concept of experience-based design (EBD) as a new way of co-designing health services with the patient in a context where they are no longer a « passive recipient of a product or service » but are « integral to the improvement and innovation process » [161]. Like other design sciences – such as architecture, healthcare is associated with the three aspects of functionality (*how well it does the job and fit its purpose - performance*), safety (*how safe and reliable it is - engineering*) and usability (*how the user interaction with the product or service is experienced*). According to Bate, *EBD is a user-focused design process with the goal of making user experience accessible to the designers, to allow them to conceive of designing experiences rather than designing services*. Which consequences such a vision has on QI work in healthcare? First, patients are incorporated for their experience of care, not necessarily for any prior expertise they may offer. Second, words are used to translate events (adverse or positive events) into experiences which may then be presented in the form of storytelling, sometimes played by actors. Third, experience amounts to more than views, complaints or satisfaction; it features *almost everything that is required to understand strengths and weaknesses and what needs to be redesigned in the care process*. For all these reasons, the acquisition and use of patient experiences in care improvement is a specialized activity which needs to be learned and practiced. It represents one valuable way to incorporate the patient experiences into care improvement. [162].

To address the question of patients' experience incorporated into QI work, specific « patient experience surveys » have been drawn up in some countries [163 ;164]. These surveys intend to collect information on the care pathway and on the characteristics of the care delivered to the patient in the previous months. They are designed to reflect the care that the patient should have received according to the standards of care for the disease. If they are administrated in ways that insure a good response rate from patients and parents, they enable the preparation of a center

report of Patient Reported Outcomes in terms of quality of care [165]. They may provide information about the variability of care across geographic or socioeconomic dimensions and avenues for quality of care improvement. These instruments help fill the gap between individual experiences of care and the general features of the care delivered to most patients.

We cannot conclude without comparing the commitment of patients and parents who accept or sometimes claim to be involved in QI programs to the activism defined by Rabeharisoa [166]. This commitment actually takes up the main features characterizing patient activism:

1) Include and shape the experiential knowledge of patients and parents;
2) Articulate it with credential knowledge in clinical, organizational and quality fields;
3) Reframe what is at stake, that is co-redesign the process of care;
4) Defend the cause: “the best possible care here and now for all patients”; and
5) Organize a network of expertise with credentialed experts in quality, patient therapeutic education, and academic instances.

Limitations of the study

Our research has some limitations. First, the sample of centres as well as patients/parents, all of which volunteered to engage in the PHARE-M QIP sessions and test the program before its roll-out throughout France, may not reflect general opinion at all CF centres in France from 2011 to 2015. Second, the appearance of numerous publications and mediated interventions in favor of taking patients' voices into account in healthcare services has triggered a beginning of a cultural shift in the last years in France. A movement called « Démocratie en Santé » emerged in France in 2015 building on this trend. In the latest PHARE-M sessions, it becomes more obvious to professionals as well as to patients and parents that the latter should be systematically involved in the QI work at the centre, and sometimes more than one at a centre. Their recruitment becomes also easier. It is hoped that arrangements will be made to facilitate patient participation in quality improvement of care, which will in turn have to be evaluated.

703 **Table I: Number of Patients at the CFC engaged in PHARE-M by year**

CF Program	Year PHARE-M	# Patients Data 2014	Pilot PHASE 2011-2013
PEDIATRIC			
Angers	2013	122	122
Bordeaux	2016	148	
Clermont-Fd	2013	103	103
Créteil	2015	109	
Dunkerque	2015	71	
Grenoble	2013	122	122
Lille	2015	181	
Lyon	2012	290	290
Nancy	2016	113	
Nantes	2012	104	104
Paris R Debré	2012	168	168
Rennes	2013	131	131
Roscoff	2012	75	75
Tours	2016	116	
Vannes-Lorient	2013	81	81
Versailles	2012	65	65
ADULT			
Lyon	2012	313	313
Nantes	2013	203	203
Rennes	2013	101	101
Montpellier	2015	197	
Reims	2012	131	131
Roscoff	2013	75	75
TOTAL Patients in PHARE-M Group		3019	**2084**
% Patients recorded in Registry		47%	**33%**

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706 **Table II -** Opinions, concerns, and illustrative quotes regarding P&PI

Opinion	Concern	Quote
Patients/parents involvement in the Quality Teams		
The place of the patient/parent in the health system	This involvement upset assigned places, led to readjustments and reinterpretations, and highlighted resilient P&P profiles.	**Physician**: "Certain physicians are not ready to accept that there is a patient at the medical staff meeting, or a meeting like the ones that we have, who gets up and disagrees, who bursts in as a consultant who gives his or her opinion." **Parent1**: "I can see that parents who are often negative or react badly to certain situations are parents who are suffering. Sometimes I feel that I stand out from other people, because I am very optimistic by nature and I have a fighting spirit. This may be why I always go a little bit beyond."
Reason for participation by **Parents**	They affirmed contributing their testimonial on their experience and sticking to merely conveying their feelings and day-to-day experiences.	**Parent2**: "I do not aim to teach anyone in a medical setting their profession — one day a physician told me that I was not going to teach him his profession. In participating, I contribute my testimonial as a parent, and that is all. More than anything else, I want to contribute my positive energy and fighting spirit." **Parent3**: "My motivation in participating in the meeting with the pediatric team is being able to give my position as a parent. So I am going to tell them my feelings regarding some of their actions. Sometimes, when I tell them my feelings, they are surprised and tell me that they had not seen things in that way."

Reasons for **Patient** involvement from their perspective	**Wariness**: patients were waried of a medicalized world. **Consent and curiosity**: to get to know a setting, to better get to know the teams that they visited as their care providers. **Engagement under tension between**: *on the one hand*, the desire to understand, be curious, gain autonomy and confidence, and remove obstacles, and, *on the other hand*, the difficulty of pushing oneself to talk in front of others about one's experiences with an invasive disease that one would like to keep at a distance.	**Patient1**: "The idea of meeting with the physicians stressed me out a bit. I wondered what I was going to do, what I should say, how it was going to go." **Patient2**: "The differences that there could be between different hospitals were quite astonishing. For example, the outcomes in FEV1% were quite impressive compared to the outcomes we had. You saw that there were distinctly better figures than what we had, indeed... So that was a bit striking to me. It was also interesting to see how other hospitals functioned and provided care, and what could be done to improve quality for patients, basically." **Patient3**: "I gave my opinion on the feasibility of things. It is all well and good to say, 'We have to do X drainages, X treatments, X thingies, etc.,' but in the end, there is real life which is different from hospital life."
Projection of healthcare providers on patients in QT	The presence of a patient on the team questions healthcare providers' professional ideas and desire. It is tempting for healthcare providers to authorize themselves to have a particular conception of patients and parents and then to talk about them, about what they believe to be their experience, in the name of healthcare providers' experience and in-depth knowledge of the person — his or her journey and record.	**Nurse**: "It would also be necessary to critique healthcare providers. Healthcare providers need to create the patient's needs. That is what they do and they are proud of it. Nevertheless, it assumes having a patient who is completely ideal, compliant, etc. Such a patient does not exist. We do not know such a patient. We have never seen one before. These healthcare providers' pushes always make me very afraid, because I do not lose sight of the fact that they are about the ideal of healthcare providers." **Nurse**: "Sometimes, saying that people do not know their disease suits us well in the end, because we will be able to have an effect on them, to explain and re-explain to them. These people understand very well and live with their disease on a day-to-day basis better than us. I do not think that we have the slightest idea of what they are really going through. They know very well what this disease is about, that the final outcome is death. When these patients relax their efforts, we should respect this and not necessarily go and add things."

Table VII: Success factors sustaining long term patient and parent involvement in QI projects

Factors related to patients and parents: • Good relationship with the care team • Coping with the disease, its complications and the effects of treatments • Stable health condition of the patient or the child of the parent • Stable socio economical family situation • Motivation to improve care for all (beyond improving care for oneself) • Possibility of involving more than one patient or parent in the team to insure the presence of one of them at each meeting and to bring diverse experiences to the discussions (for instance parents of children of various ranges of age or transplanted and non transplanted patients…) • Ability to give time to the project, participating to the trainings and local meetings, and availability of communication tools (internet) at home
Factors related to the care team: • Mature relationship with the patient/parent: readiness to a partnership for care, being at ease with shared decision making and/or patient education • Leadership wishing to involve patients/parents on a long-term basis, « playing the rule » of transparency and effectively taking the responsibility for the project and for the change actions implemented • One professional being the correspondent of the patient/parent for the QI project solving practical issues • Awareness to the guidelines and consensus for care and ability to discuss/share them with the patient/parent • Attention paid to psychosocial difficulties encountered by the patient potentially contradictory with their involvement
Factors related to the QI method • Present the involvement of a patient/parent as a pre-requisite to engage in QI work, based on literature and a « safe » framework to recruit them • Take the financial charge of patient and parent involvement at the program level (thanks to an agreement with the patient organizations if possible) • Offer an appropriate set of communication tools towards the patients/parents followed at the center, including the patient group if any, as well as towards the hospital administration • Provide the same training on the quality methods and tools to the professionals and the

patients/parents involved

- Install resources for the QI work at the centre and manage the regular participation of the patient/parent or his update on the project
- Secure the framework with ethical rules allowing full participation of all members, recalling roles and responsibilities
- Start from where the teams are in terms of patient outcomes, professionals, processes and patterns
- Challenge the teams so that they fix their problems and choose a shared realistic goal to be achieved at the deadline of the project
- Offer new perspectives, facilitate benchmarking with other practices, provide access to guidelines and consensus for care to the whole team
- Provide an on-site Coaching to support the team in analyzing their processes of care from the point of view of the patient/parent (shadowing a patient) and reinsuring the place of the patient/parent involved
- Proceed by PDSA cycles, measuring the results of the test and adjusting if necessary, and share the results with the whole team
- Consider that the results achieved are attributable to the whole quality team and beyond, to the multidisciplinary team who implement the new process of care, and not to one member in particular, be it a patient/parent or a professional

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