Institution:
University of Central Lancashire

Unit of Assessment:
4 Psychology, Psychiatry and Neuroscience

Title of case study:

Changing mental health care to improve wellbeing in people with cystic fibrosis and their caregivers

Period when the underpinning research was undertaken:
2001-2015

Details of staff conducting the underpinning research from the submitting unit:

Name(s):	**Role(s) (e.g. job title):**	**Period(s) employed by submitting HEI:**
Janice Abbott	Professor of Health Psychology	1st February 1992 – present date

Period when the claimed impact occurred:
1st August 2013 - 31st December 2020

Is this case study continued from a case study submitted in 2014? Y/N
N

1. Summary of the impact (indicative maximum 100 words)

In developed countries, the life expectancy of people with cystic fibrosis (CF) is 35-40 years. Our vision was to change the culture of mental health care and the way care is delivered, so that mental health would become a vital, integrated aspect of CF care. We have changed international policy by developing and publishing CF Mental Health Guidelines for health professionals; created jobs when the US CF Foundation allocated approximately USD20,000,000 to fund 138 Mental Health Coordinators and changed stakeholder engagement through the creation of US and European Mental Health Working Groups. Furthermore, a change in international clinical practice occurred when annual mental health screening and appropriate treatment referrals for patients and caregivers was implemented. Finally, and most significantly, people with CF and their caregivers, have reported globally, an improvement in their mental wellbeing.

2. Underpinning research (indicative maximum 500 words)

Initial research to demonstrate the magnitude of the problem

Cystic fibrosis is a genetic, life-limiting condition with an immense treatment burden. Due to medical advancement, survival has dramatically increased to 37 years in the UK. However, for over two decades, the negative psychological impact of living with CF has been documented in the work led by Abbott. This has robustly demonstrated that psychological distress in people with cystic fibrosis has been associated with worse health outcomes such as decreased survival, inadequate self-management of the condition and poor quality of life [1, 2, 3]. To evaluate the prevalence and predictors of depression and anxiety in people with cystic fibrosis and their caregivers, a large international study, the *Cystic Fibrosis International Depression and Anxiety Epidemiological Study* (CF TIDES) was undertaken. This international endeavour involved eight European countries and the US. The UK arm of the work was co-led by Abbott and included over 2,700 participants from 39 UK Cystic Fibrosis Centres. As part of the UK work, predictors of disordered mood were identified and a novel UK gender-specific CF scoring system was developed to help the clinician determine when to refer a patient for specialist psychological assessment: a significant step in the management of mood disorders in CF [4]. The International work recruited over 10,000 patients aged 12 and over, and their parent caregivers, across 154 CF Centres. Elevated symptoms of depression that required further assessment, were found in 10% of adolescents, 19% of adults, 37% of mothers and 31% of fathers. Elevated symptoms of anxiety were reported by 22% of adolescents, 32% of adults, 48% of mothers and 36% of fathers. Of 1,122 parent-teen dyads, if either parent screened positive, the adolescent was twice as likely to report elevated depression or anxiety. Additionally, disordered mood was associated with poorer quality of life and poorer physical health outcomes. Recognition of this work as world-leading by the Journal Editor led to the award of 'Editor's Choice' with free 'open access' (*Thorax* 2014; IF 8.27; 250+ citations) [5]. The results highlighted the importance of measuring and treating mental health issues in patients and families coping with CF and sparked an aspiration, within the CF community, to improve mental health. Specifically, the European CF Society and the US CF Foundation formed a joint expert, multidisciplinary, *International Committee in Mental Health* to develop mental health guidelines for CF health professionals, with the aim of improving mental well-being in patients and their caregivers.

Research undertaken during development of CF International Mental Health Guidelines

During the creation of the Mental Health Guidelines a total of 21,756 references were retrieved and 344 articles reviewed to inform the development of fifteen recommendations for improving mental health care in CF [A]. To inform the committee's deliberations concerning mental health care delivery, the views of the international CF clinical community were sought via a web survey. Abbott led the evaluation of the survey (1,454 responses from 48 countries), presented the results to clinicians in symposia at International Conferences, and published the work. The *Survey of Mental Health Care Delivery* highlighted that care was inadequate and highly variable across CF centres and countries. Insufficient resources and a lack of training, competency and standardisation, such as the use of 48 different depression and anxiety screening tools, generated barriers to mental health provision [6]. These data informed the first draft of the CF Mental Health Guidelines and the subsequent establishment of separate US and European Mental Health Committee/Working Groups.

3. References to the research (indicative maximum of six references)

[1] Abbott J, Morton AM, Hurley MA, Conway SP. 'Longitudinal impact of demographic and clinical variables on Health-Related Quality of Life in Cystic Fibrosis.' *BMJ Open* 2015; 5: e007418doi:10.1136/bmjopen-2014-07418. https://bmjopen.bmj.com/content/5/5/e007418

[2] Abbott J, Hart A, Morton AM, Dey P, Conway SP, Webb AK. 'Can health-related quality of life predict survival in adults with cystic fibrosis?' *American Journal of Respiratory and Critical Care Medicine* 2009; 179:54-58. https://doi.org/10.1164/rccm.200802-220OC

[3] Abbott J, Dodd M, Gee L, Webb AK. 'Ways of coping with cystic fibrosis: implications for treatment adherence.' *Disability and Rehabilitation* 2001; 23:315-324.

[4] Duff AJA, Abbott J, Cowperthwaite C, Sumner C, Hurley M, Quittner AL (on behalf of the TIDES-UK Group). 'Anxiety and depression in adolescents and adults with cystic fibrosis in the UK.' *Journal of Cystic Fibrosis* 2014; 13:745-53. https://www.cysticfibrosisjournal.com/article/S1569-1993(14)00050-2/pdf

[5] Quittner AL, Goldbeck L, Abbott J, Duff A, Lambrecht P, Solé A, Tibosch MM, Bergsten Brucefors A , Yüksel H, Catastini P, Blackwell L, Barker D. 'Prevalence of depression and anxiety in patients with cystic fibrosis and parent caregivers: results of The International Depression Epidemiological Study across nine countries.' *Thorax* 2014; 69:1090-7. https://thorax.bmj.com/content/thoraxjnl/69/12/1090.full.pdf

[6] Abbott J, Elborn JS, Georgiopoulos AM, Goldbeck L, Marshall BC, Sabadosa KA, Smith BA, Quittner AL. 'Cystic Fibrosis Foundation and European Cystic Fibrosis Society Survey of Cystic Fibrosis Mental Health Care Delivery. *Journal of Cystic Fibrosis* 2015; 14:5339. https://www.sciencedirect.com/science/article/pii/S1569199314003129

4. Details of the impact (indicative maximum 750 words)

Several major impacts have emanated from the research, starting with policy development with stakeholders and culminating in improved mental well-being through the implementation of mental health screening in people with cystic fibrosis and their caregivers. Our vision has challenged conventional wisdom, stimulated debate and has changed hearts and minds regarding mental health care in cystic fibrosis around the world.

Changes in international policy development for mental health screening and treatment

The ambition was to change international clinical practice by providing clinicians with clear guidance on mental health care. The *International Committee in Mental Health* consisted of 22 members: a core group directed aspects of guideline development, implementation and wrote the documentation. This included 3 from the US, 1 from Germany and Abbott from the UK, who directed the workgroup on implementation and future research. We collected feedback on the preliminary guidelines following distribution to the CF clinical community, patient and parent groups in Europe and the US. This feedback informed the final guidelines which included fifteen recommendations concerning prevention, screening, clinical assessment, psychological and pharmacological treatments. The guidelines were published in October 2015 [A]. They proposed that just as measuring lung function and weight are part of CF care, screening and treating depression and anxiety should become a routine part of CF care too. Annual mental health screening was recommended, using specific screening questionnaires, for all patients over 12 years old and their primary caregivers. The resulting guidelines publication highlighted the results of *The International Depression and Anxiety Epidemiology Study* and the *Survey of Mental Health Care Delivery* as the catalysts that led to their development [A].

A step by step guide for clinicians, regarding the systematic screening, diagnosis and management of depression and anxiety was also developed and published [B]. Publication of the guidelines corresponded with a US plenary conference presentation 'There Is No Health Without Mental Health' in Phoenix in 2015, attended by over 4,000 delegates from 42 countries. During this plenary session, the US CF Foundation pledged to fund mental health care. The Director of Clinical Communications commented **"I left the plenary energized by the idea of including emotional wellness in the care for people with cystic fibrosis and their parent caregivers".**

Creating Mental Health Coordinator Jobs in the US

The US typically did not have clinical psychologists available to Cystic Fibrosis teams as they did in many European countries. However, as a result of our research, in 2015, the US Cystic Fibrosis Foundation provided initial funding for Mental Health Coordinator appointments in 84 CF Centres at a cost of USD8,600,000. Thirty-six further appointments were made in August 2016 and 18 in April 2018 [C2]. Subsequently, in 2020, maintenance funding was provided with a total investment of approximately USD20,000,000 [C]. The Mental Health Coordinator's role was to undertake annual screening and patient follow-up, coordinate evidence-based treatment for depression and anxiety, develop and maintain a referral network of community-based mental health practitioners, serving as educator and liaison for mental health care in the CF centres. These appointments were imperative to improve the lives of people with CF and their caregivers.

Changes in stakeholder engagement

Building international capacity for working with different healthcare systems

Our goal was to implement mental health screening by using a unique structure of accredited and recognised CF Centres in Europe and the US. The US has different needs to Europe and different European countries have different needs based on their own healthcare systems. To facilitate far reaching international impact and ensure dissemination and implementation of the guidelines, in 2016, the US CF Foundation established a *Mental Health Advisory Committee* involving Abbott as the European representative. Similarly, the European CF Society approved a proposal written by Abbott to establish *a Mental Health Working Group*. The aim was to work with the whole European CF community including health professionals, patients and caregivers, and to translate the mental health guidelines and resources into numerous European languages. Abbott was Co-Lead on the project which ensured that the community disseminated and implemented the guidelines whilst also evaluating their effectiveness. Clinical psychologists were predominantly appointed as Country Captains for 29 European countries, and Australia, to assist with language translation and dissemination via national networks.

Resources for clinicians, people with CF and caregivers

The European Mental Health Working Group activities are hosted on the European Cystic Fibrosis Society (ECFS) website which was accessed by 3,944 people between October 2017 and October 2018 [D]. It provides easy access to resources developed by both the US and European Committees [E]. There is a 'toolkit' to facilitate implementation of annual mental health screening which includes the guidelines, the recommended anxiety, depression and suicide screening tools. There is also information for patients and caregivers and all these materials are available in at least 17 different languages. We have also helped to develop a range of training opportunities and professional networks. In the US a 'drop box' resource system has been developed and includes a clinician 'Tracking Log' for observing mental health changes. A valuable email peer support and information network for over 400 worldwide mental health professionals is also in operation.

Change in international clinical practice

Annual mental health screening and appropriate treatment referrals for patients and caregivers have been implemented in CF Centres across the US, Canada, South America, Europe and Australia. In work driven by Abbott, the *European Mental Health Working Group* and the *US*

Mental Health Advisory Committee surveyed CF mental health professionals to evaluate the implementation of the guidelines. The responses came from Mental Health Coordinators in the US [F], and from CF Centres across 29 European countries [G] and provided early indicators of effectiveness from the health professional's perspective. By 2019, approximately 25,000 patients and carers were in screening programmes, approximately 10,000 individuals had been referred for treatment with almost 1,000 urgent referrals for suicide ideation. Clearly, these are underestimates as the data are based on a small timeframe and only those responding to surveys, but they highlight rapid commitment to the recommendations provided in the guidelines. With clear, standardised guidelines, it is notable that the prevalence of depression and anxiety is much greater than estimated in the original research. Clinicians reported the benefits of mental health screening to be (1) easier to initiate mental health conversations, (2) greater awareness of mental health among the CF multidisciplinary team, patients and caregivers, (3) quicker recognition of symptoms and (4) mental health becoming destigmatised [F,G]. Subsequently, mental health screening has been added to the US Adult Care Checklist. This encourages partnership between clinicians and people with CF and tracks clinical assessment and procedures recommended by the CF Foundation.

Training and mentoring to enable screening and follow-up care

Feedback from the *Survey to evaluate implementation of the guidelines* [G] also highlighted that mental health was viewed 'as everyone's business' and not just the domain of mental health professionals. Consequently, training courses were provided for the entire multidisciplinary team including doctors, nurses, physiotherapists, pharmacists, dieticians and social workers. Numerous US and European courses have trained over 1000 health professionals in mental health screening, psychological and pharmacological treatments, with an Australian roadshow across six cities providing training to clinicians and sessions for patients and caregivers. To ensure sustained, quality mental health care, in 2018, the *US Mental Health Advisory Committee* set up mentoring programs for new Mental Health Coordinators and Peer Consultation Groups of clinicians who meet each month to discuss best practices in mental health screening, education and treatment.

Survey feedback from clinicians [F,G], people with CF and caregivers [H] have highlighted barriers to appropriate treatments. Specialist CF mental health practitioners are not widely available in the community and the risk of cross-infection often isolates people with CF. In the US, a pilot programme to assist patients and caregivers to use telehealth was successful and the CF Foundation has developed a comprehensive 'how to' guide of CF telehealth care delivery [I]. This has been invaluable during the Covid-19 pandemic.

Improvement in mental wellbeing reported on a global scale

Video testimonials produced by the US CF Foundation from patients, caregivers and health professionals highlight the powerful impact that mental health screening and appropriate care has had on their lives. As this adult with CF describes: **"Depression, it's always there, it's kind of hibernating. Going to therapy is a part of fighting this disease, it's hard to fight this disease every day"**. A parent caregiver states that: **"Mental health screening should start right from the beginning. There are key points within the CF life that absolutely rip you apart… if you are not healthy you cannot take care of your child"** [J].

Individual CF Centre evaluations confirm changes in wellbeing

Screening results are being tracked before and after treatment in numerous CF Centres globally to evaluate the effectiveness of mental health screening and care pathways. Some of these early results have been presented at international conferences and typically show that after five to eight sessions of psychological therapy, patients and caregivers reported significant decreases in anxiety and depression symptoms and an improvement in their wellbeing [K].

Patient and caregiver survey responses

Recently, the views and experiences of mental health screening and treatment were sought from people with CF and their caregivers. Results were posted on the ECFS Mental Health website in July 2020 as feedback to the CF community [H]. There was universal agreement from 474 patients and caregivers that mental health is an important aspect of their CF health; 97% believed they should be asked about mental health symptoms at their clinic appointments and 90% valued the conversation that took place immediately following screening. The voices of people with CF and their caregivers are poignant: An adult with CF said: **"Following the death of two CF friends I kept thinking why I am still here. I was so down, so depressed. I didn't feel worthy of being alive, but I was too much of a pansy to end my life suddenly. At my outpatient appointment I filled in the questionnaires asking me how I was mentally. My results were probably alarming because I then had a long chat with the psychologist about my suicidal thoughts. She was amazing and after seeing her for three months I have my mojo back. I feel life is worth living whatever CF throws at me."** [H] A parent caregiver said: **"Since my son was born 18 years ago, I have lived in fear that his life will be cut short. It is an ongoing nightmare and I feel frustrated and angry when he refuses to do his physio. He tells me not to worry but I can't help it. I am so thankful for the support of the CF Centre who asked me about my mental health. These things really matter and help to make life easier".**

An International Award

It has been an amazing collaborative journey from the initial *CF International Depression and Anxiety Epidemiological Study* to global improvements in mental health care and patient and caregiver wellbeing. In November 2019, the US CF Foundation honoured Abbott with the *Richard and Carolyn Mattingly Award for Leadership in Mental Health* Care in the first year the award was open to researchers and clinicians outside the US.

5. Sources to corroborate the impact (indicative maximum of 10 references)

[A]. CF Mental Health Guidelines (220+ citations). Quittner AL, Abbott J, Elborn JS, et al. Cystic Fibrosis Foundation and European Cystic Fibrosis Society Consensus Statements for Screening and Treating Depression and Anxiety. Thorax doi:10.1136/thoraxjnl-2015-20748
http://thorax.bmj.com/content/early/2015/10/09/thoraxjnl-2015-207488.full

[B]. Depression, anxiety and cystic fibrosis, Step by step guide for clinicians
https://www.cff.org/For-Caregivers/Clinical-Care-Guidelines/Depression-Anxiety-and-CysticFibrosis-Guide-for-CF-Clinicians.pdf

[C]. [C1]: US Mental Health Coordinator Funding Award documentation
https://www.cff.org/Our-Research/For-Researchers/Career-Development-Awards/Mental-Health-Coordinator-Award-Policies-and-Guidelines.pdf

[C2]: CF Mental Health Change Package (provides timeline and funding information). Poster submission to the Institute for Healthcare Improvement Forum, 2018.

[D]. ECFS Mental Health Working Group Website: hosting screening resources, training opportunities and information for patients and families https://www.ecfs.eu/mental-health-in_CF

[E]. Resources developed for parents/caregivers by US and European MH Committees
https://www.cff.org/Living-with-CF/Emotional-Wellness/Depression-Anxiety-and-Cystic-FibrosisWhat-the-Guidelines-Mean-for-You.pdf

[F]. [F1] US screening Implementation. Quittner AL, Abbott J, Hussain S. *et al*. Integration of mental health screening and treatment into cystic fibrosis clinics.
https://www.onlinelibrary.wiley.com/doi/epdf/10.1002/ppul.24949

[F2] Dissemination & Implementation of the Mental Health Guidelines in the US: Results of Implementation in Year 2 @120 CF Centers, Quittner AL, Abbott, J. et al, Poster

[G]. European screening Implementation. Abbott J, Havermans T, Jarvholm S, et al. Mental health screening in Cystic Fibrosis Centres across Europe. Journal of Cystic Fibrosis 18 (2019) 298–302. https://www.cysticfibrosisjournal.com/article/S1569-1993(18)30801-4/pdf

[H] Cystic Fibrosis Patient and Caregiver Mental Health Survey Results, Posted on ECFS website July 2020

[I]. [I1] Telehealth pilot and CF telehealth mode of care delivery formal documentation.

[I2] Sustaining Mental Health Care: Technology Closes the Distance, CF Foundation, May 2019

[J]. Video testimonies from stakeholders and beneficiaries:
https://www.youtube.com/watch?feature=player_embedded&v=387uwRzlpPE
https://www.youtube.com/watch?v=sxFST8W6MZA
https://www.youtube.com/watch?v=z9_f1xkPVDw

[K]. Early results from individual CF Centres demonstrating improved wellbeing following MH screening and treatment. Conference poster examples from CF Centres in the US and Europe.