Title page

Journal: Psychology & Health

Authors:
Sophie Dawson[1,2], Karen Rodham[2,3], Jennifer Taylor[2], Jane Dewar[1], Martin Wildman[4]

Affiliations:
[1] Wolfson Adult Cystic Fibrosis Centre, Nottingham University Hospitals NHS Trust, Nottingham, UK.
[2] Staffordshire Centre for Psychological Research, School of Health, Science and Wellbeing, Staffordshire University, Stoke-on-Trent, UK.
[3] Institute of Psychology, Business and Human Sciences, University of Chichester, College Lane, Chichester, UK.
[4] Sheffield Adult Cystic Fibrosis Centre, Sheffield Teaching Hospitals NHS Foundation Trust, Sheffield, UK.

Corresponding author:
Sophie Dawson: sophie.dawson@nuh.nhs.uk

ORCiDs:
Sophie Dawson: 0000-0002-2903-3215
Karen Rodham: 0000-0002-5377-1998
Jennifer Taylor: 0000-0002-7133-2913
Jane Dewar:
Martin Wildman: 0000-0002-1658-881X

Twitter handles:
Sophie Dawson: @sophie_doph
Karen Rodham: @RodhamKaren
Jennifer Taylor: @DrJenTaylor246

"I think most people feel like healthcare professionals tell them to take their treatments and judge them for not taking them": Reflexive thematic analysis of the views of adults with cystic fibrosis on how treatment adherence is discussed in healthcare.

Abstract

Objective

Previous research exploring patient-practitioner communication in relation to adherence in cystic fibrosis (CF) is limited. This UK study explored the views of adults with CF on how treatment adherence (related to all CF treatments) is discussed in routine CF care.

Methods

12 White British adults (ten females; aged 20-37 years; mean 30.1 years) with CF participated in semi-structured interviews.

Results

Three overarching themes were developed through reflexive thematic analysis: 1) 'The power of language'; 2) 'Health professionals do not recognise the importance of context'; and 3) ''Admitting' non-adherence is difficult'.

The way in which adherence is discussed in adult CF care is viewed as paternalistic and infantilising. Participants reported that healthcare professionals do not always consider the desire to balance treatment-taking with living a normal life. Unwelcome responses from healthcare professionals, and the inability to accurately self-report the amount of treatment taken made it difficult to 'admit' non-adherence.

Conclusions

A culture change is needed in CF care such that people who struggle to take their treatments are not labelled as disobedient, wilfully disobeying orders from healthcare professionals in positions of authority. Instead, an open, honest, non-judgemental approach, as recommended by healthcare agencies for over a decade, should be adopted.

Word count: 200 words (max. 200 words)

Keywords: Medication adherence; Communication; Cystic Fibrosis; Language; Healthcare professional behaviour; Qualitative research

Background

Cystic fibrosis (CF) is a chronic, genetic condition usually diagnosed shortly after birth through newborn screening (UK CF Registry, 2022). There are over 10,900 people living with CF in the UK (UK CF Registry, 2022), over 32,000 people in the US (CF Foundation Patient Registry, 2022), and around 90,000 people worldwide (Bell et al., 2020). The median predicted survival age of someone born with CF between 2017-2021 is 53.5 years in the UK (UK CF Registry, 2022), and 53.1 years in the US (CF Foundation Patient Registry, 2022).

People with CF are prone to recurrent chest infections (pulmonary exacerbations) due to the build-up of thick, sticky mucus in their lungs and digestive systems (UK CF Registry, 2022). Although advances in medicine mean that many of the symptoms of CF can be managed effectively, in practice this means that a complex daily regimen of preventative or maintenance treatments is required (Sawicki et al., 2009). These may comprise inhaled (or nebulised) therapies; pancreatic enzymes; vitamin supplements; and daily chest physiotherapy (CF Trust, 2011). In recent years, precision medicines, also known as CF transmembrane regulator (CFTR) modulator treatments, which target the underlying dysfunction of the CFTR protein, have become available and are now suitable for around 90% of people with CF (Burgener & Moss, 2018). These treatments (e.g., elexacaftor-tezacaftor-ivacaftor) appear to have changed the landscape for many people with CF, with rapid and transformative improvements in health observed in clinical trials (Heijerman et al., 2019; Middleton et al., 2019).

Various terms are used to describe patient treatment-taking behaviour. '*Compliance*', for example, is 'the extent to which the patient's behaviour matches the prescriber's recommendations' (Haynes et al., 1979). In contrast, '*adherence*' refers to 'the extent to which the patient's behaviour matches *agreed* recommendations from the prescriber' (Horne et al., 2005, p. 4). This term is considered to be more patient-centred and therefore preferable to '*compliance*' because the recommendations are agreed between rather than given to the patient by the healthcare professional. The term '*concordance*' is more recent and places greater emphasis on negotiation and agreement between patients and clinicians after a shared discussion but is less widely used in practice (Horne et al., 2005). Finally, although '*medication-taking'* has been suggested more recently as an appropriate term to use with patients (Dickinson et al., 2017), 'adherence' is widely used in the literature and has been proposed as the "term of choice" (Horne et al., 2005, p. 13), so this is the term we use throughout this paper.

Low adherence to treatment is a global health problem linked with poorer health outcomes and increased healthcare costs (World Health Organisation; WHO, 2003). Consistent with adherence rates across a range of long-term conditions (WHO, 2003), people with CF often struggle to take their preventative treatments. For adults with CF, objective treatment adherence estimates range from 31-

36% for inhaled therapies (Daniels et al., 2011; Hoo et al., 2021), 43% for pancreatic enzymes, and 34% for vitamins (Modi et al., 2006). Even with the recent introduction of highly effective modulator treatments (oral tablets) in CF, early evidence suggests that adherence may be suboptimal (e.g., 61-85%; Mitchell et al., 2021; Siracusa et al., 2015).

One outcome of low adherence is an increased need for 'rescue' treatment in the form of 10-14 day courses of intravenous (IV) antibiotics (Eakin et al., 2011). This causes significant disruption for the person with CF and their family and has a detrimental impact on patient quality of life (Britto et al., 2002). In addition to the costs for individuals, low adherence has widespread implications for healthcare services, with higher adherence to CF treatments associated with improved outcomes including reduced hospital admissions (e.g., Quittner et al., 2014) and length of inpatient stays (Nasr et al., 2013).

The influence of patient-practitioner communication on adherence is well documented in the research literature, with enhanced clinician communication highly correlated with greater patient adherence (e.g., Haskard-Zolnierek & DiMatteo, 2009). Not surprisingly, being able to communicate openly with clinicians about the barriers to adherence, obtaining key information, feeling supported and involved in care decisions may be an important facilitator of adherence (Haskard-Zolnierek & DiMatteo, 2009). Indeed, the UK National Institute for Health and Care Excellence (NICE; 2009) recommends that healthcare professionals use a non-judgemental approach when discussing adherence with patients to promote open and honest conversations.

Language is an important aspect of patient-practitioner communication and can have a profound, lasting impact on patients living with a chronic condition (Cooper & Swindell, 2021; Cox & Fritz, 2022). Language has the power to have a motivating or negative impact (Cooper & Swindell, 2021). It can influence patient health preferences, expectations, perceptions, and outcomes (e.g., Dickinson et al., 2017; Nickel et al., 2017). In the diabetes field, which is similar to CF in that it is a chronic condition that requires a high level of daily self-management, there has been call for a "language movement" (Dickinson et al., 2017; p. 1790). This involves moving away from language with potentially negative connotations (e.g., 'poorly controlled diabetes'; 'non-compliant patient') and, instead, using neutral, non-judgemental language that is respectful, free from stigma, and fosters collaboration between patients and healthcare professionals (Dickinson et al., 2017).

Healthcare professionals working in the CF field need to understand the amount of treatment that patients are taking in order to inform clinical decision-making (Wildman & Hoo, 2014). However, with the exception of data-logging nebulisers (e.g., PARI eFlow® rapid nebuliser system with an eTrack Controller® (PARI Pharma GmbH, Starnberg, Germany); Philips I-neb Adaptive Aerosol Delivery (AAD) System (Philips Respironics, Chichester, UK)), which record time- and date-stamped usage data, few objective measures of adherence are available for use in routine CF care. Instead, an assessment of adherence typically relies on patient self-report. Self-report is subject to a range of biases (Osterberg & Blaschke, 2005), and evidence has highlighted the discrepancy between self-reported and objectively measured adherence in people with CF (e.g., Daniels et al., 2011; Modi et al., 2006).

The discrepancy may at least in part be related to communication issues. Indeed, as we noted above, developing a caring, trusting, and mutually respectful relationship in which patients feel comfortable talking openly about their adherence is an important facilitator of adherence. Unfortunately, a qualitative study conducted by Arden et al. (2019) highlighted that people with CF with low adherence to inhaled therapies reported conflict with health professionals. Whether conflict had arisen because of low adherence, or whether conflict with health professionals resulted in a lack of trust which resulted in lower adherence would need to be investigated in a further, quantitative study. Nevertheless, if conflict is a possible contributory factor or outcome linked to low adherence, then people with CF may not feel able to talk openly with healthcare professionals, and so non-adherence remains “invisible” (e.g., Wildman & Hoo, 2014, p. 16).

To our knowledge, although the importance of good communication has been highlighted in relation to adherence in CF (e.g., Arden et al., 2019; Lask, 1994; Sawicki et al., 2015), no study has specifically explored the role of patient-practitioner communication on treatment adherence from the perspective of those with CF. The aim of this study is therefore to explore the views of adults with CF on how the subject of treatment-taking, or adherence, is discussed in routine CF care. This is pertinent since "improving and sustaining adherence to treatment" was identified as a James Lind Alliance top ten CF research priority (e.g., Calthorpe et al., 2020). The scope of our study was intentionally broad to focus on all aspects of CF care and was not limited to focus on specific treatments or interactions that typically take place in routine CF care (e.g., outpatient clinic appointments or interactions with certain roles of the MDT). A qualitative approach was used to gain rich and detailed insights into the views of people with CF. Understanding these views has the potential to influence CF care and the development of interventions to support treatment-taking in people with CF.

Methods

Participants and recruitment

12 White British adults with CF (10 females) aged between 20-37 years (mean 30.1 years) participated in the study (Table 1; Appendix 1). One participant (Sally) was post-lung transplant. Ethical approval (SU_20_018) was obtained from the first author’s second institution.

An opportunistic sampling strategy was used, with participants recruited through Twitter due to the pausing of new or ongoing non-COVID-related research studies in the NHS during the COVID-19 pandemic. The first author advertised the study on Twitter, with relevant organisations (e.g., national CF charity and NHS CF centres) tagged to aid recruitment. Participants who met the study eligibility criteria (i.e., adults with CF, aged 18 years or over, who self-reported attending a specialist CF centre in the UK) were invited to contact the researcher for further information by email or private message on Twitter. The researchers followed Braun and Clarke's (2019a) guidelines for sample size recommendations and aimed to recruit between 10-20 participants for a medium-sized study.

Interviews

A semi-structured interview schedule was developed (Appendix 2), to provide structure whilst allowing the flexibility for participants to discuss issues that were important to them and that had not been anticipated in advance. The schedule covered three topic areas (language; patient-practitioner interactions; written communication) and was informed by existing literature (e.g., Sawyer & Aroni, 2003; Tilson, 2004), blogs (e.g., Cooper & Swindell, 2022; Corkhill, 2018; Hoffman, 2015; Thomas, 2012), social media, and discussions that the first author had had with people with CF in her clinical role. During the interviews, the researcher asked all the questions in the schedule as well as some unplanned, spontaneous follow-up questions (e.g., "Can you tell me more about that?"). Since the management of CF involves medication along with other treatments (e.g., physiotherapy), the term 'treatment-taking' was used throughout the interviews to encompass all CF treatments. All participants provided written informed consent which they emailed to the researcher before the interview. Verbal consent was also obtained at the start of the interview, and participants were asked to provide some basic demographic details (e.g., sex, age, ethnicity).

Interviews were conducted by the first author, a 30-year-old female who was conducting this research as part of a Professional Doctorate in Health Psychology. She also works in an adult CF centre and has experience supporting people with CF with their adherence. The first author kept a reflective diary throughout data collection and analysis, which allowed her to consider how her prior knowledge of CF may influence interview prompts and to be conscious to allow the participant to direct more of the interview. Interviews were audio-recorded and transcribed verbatim, using pseudonyms to protect the anonymity of participants. Interviews lasted 73 minutes on average (range 39-109 minutes). Data collection took place in November 2020. The decision to stop data collection was a situated, interpretative judgement made by the researchers (Braun & Clarke, 2021), influenced by the richness of the dataset and how that linked with the aims of the study (Braun & Clarke, 2022). All participants who expressed an interest in taking part were interviewed. Participants were invited to enter a prize draw to win a £25 online voucher. Due to the COVID-19 pandemic, all interviews were conducted by video-call (which had the advantage of facilitating participation across a wide geographical area in England).

Analysis

Transcribed interviews were analysed using inductive reflexive thematic analysis, using the guidelines recommended by Braun and Clarke (2019b), so that the analysis was driven by the data as opposed to a predetermined theory or framework. This was conducted in relation to a critical realist epistemological standpoint, which assumes that how we experience reality is shaped by culture, language and political interests (Maxwell, 2012). After reading and re-reading each interview transcript to familiarise herself with the data, the first author (SD) noted down items of potential interest. Following this, each transcript was inductively coded by SD using NVivo^{TM} (QSR International) software to identify aspects of the data relevant to the research question across the entire dataset. In reflexive thematic analysis, coding is a 'process of exploring the diversity and patterning of meaning from the dataset' (Braun & Clarke, 2022, p. 53). A code can be an analytically interesting idea, a concept or meaning associated with segments of the data. Similar codes were then combined on NVivo to create potential patterns, or 'themes', which were then reviewed and revised to ensure the themes fit well with the coded data and that each theme had sufficient supporting data. If this was not the case, themes were either omitted or collapsed to form a sub-theme of another theme. Themes were then renamed and defined to ensure that they described the data. SD conducted the initial analysis. Braun and Clarke (2022) recognise that asking questions of the data and yourself as a researcher is an important part of the analytical process. With that in mind, three transcripts were read and fully analysed independently by an experienced qualitative researcher (KR). Whilst the analysis conducted by KR generated similar themes, the process of discussing the differing interpretations allowed the first author to: reflect on her own analysis; ensure that the themes generated reflected the data; and clarify assumptions she might have been making as a function of her experience in the CF field.

Results

Three overarching themes were developed through reflexive thematic analysis: 1) The power of language; 2) Healthcare professionals do not recognise the importance of context; and 3) 'Admitting' non-adherence is difficult. Themes will be summarised below, with illustrative quotations, accompanied by the participant's pseudonym in parentheses.

Theme 1: The power of language

This theme focuses on participants' views of the terms 'compliance, 'adherence' and 'concordance', and the impact of the language health professionals use in relation to treatment-taking. It was common for participants to view this language as paternalistic and infantilising.

Participants had not heard the term 'concordance' used in relation to treatment-taking and were uncertain about its meaning, e.g., *"I ain't got a scooby* [clue] *what it means"* (Sally). However, most participants were familiar with the terms 'compliance' and 'adherence', and many had heard them used by healthcare professionals. The terms were *"technical"* (Steph), "*medical"* (Sarah), *"scary, clinical"* (Nicola) terms. These words emphasised the power imbalance between the patient and healthcare professional. For example, at times of disagreement, healthcare professionals would sometimes default back to using technical terms and more directive language, reflective of the notion of 'compliance':

> *"They tend to get almost exasperated when I'm sort of saying, 'I'm actually really struggling to take these [treatments]… even though they're only a mouthful… and they tend to get quite exasperated and then terms come out like, you know, 'you need to be more compliant' and things like that"* (Alice).

In contrast, when healthcare professionals used more 'every day', lay language to discuss treatment-taking, participants felt more connected and recognised that the healthcare professionals were making an effort to communicate clearly. For example, Nicola said:

> *"I feel like they always kind of meet people on their level, or at least my CF team is very good at that and will talk in like very much like layman's terms and are very good communicators. And they will just say simply like 'how was taking this treatment or this medication?'"* (Nicola).

'Compliance' was viewed as a paternalistic term (Charlotte: *"Doctor says, patient does"*), in which the person with CF was perceived to have limited agency and a passive role in their CF care. It was associated with authority, and a lack of freedom or control, which created a perceived power imbalance between patients and healthcare professionals:

> *"'Compliant' is like you're being told to do something, it's like almost like a bit of a battle or a struggle to see if you can like squeeze someone into compliance"* (Nicola).

Patients who did not 'comply' felt as though they were infantilised and perceived as being disobedient, wilfully ignoring orders from someone in a position of authority, e.g., *"it's possibly got a bit more of like a connotation of like, you know, being at school or following the rules, and that it's sort of quite naughty to not take [treatments]"* (Richard). The term therefore elicited a negative emotional reaction: *"The idea of being compliant sort of rubs me up the wrong way um because I just like to feel like I've got my own autonomy"* (Nicola).

'Adherence' was viewed as having a similar meaning to 'compliance', although 'adherence' was considered slightly less paternalistic, less *"instructional"* (Rosie), and more flexible, e.g., *"['Adherence'] seems slightly less um stigmatising than 'compliance', but neither are nice… I think compliance is the worst, but adherence isn't much better*" (Charlotte). 'Adherence' was generally associated with patients having more of an active role in treatment-taking; it suggested that there had been some agreement between the patient and healthcare professional in terms of the treatment regimen, e.g., *"I think ['adherence'] feels a lot more relaxed, a lot more kind of free and open to collaboration from myself"* (Sarah).

Participants highlighted other terms that they found similarly problematic (e.g., when treatment-taking was discussed in written communication) because they emphasised the power differential between the patient and healthcare professional:

> *"'Disclose'… you 'disclose' something you're ashamed of, and I don't think you should be ashamed of your treatment regimen. And 'reported' is… that is quite a submissive term, so I 'report' things to my manager or whatever, it's not an equal playing field"* (Charlotte).

This language impacted on how participants felt and spoke about their levels of adherence, with many using phrases usually associated with criminal activity. For example: Charlotte: *"I admitted to my consultant a while ago that I wasn't taking my vitamins";* Sally: *"I'd have to come clean").* One participant (Charlotte) described a time when she had *"confessed to [her] sins"* during a clinic appointment and had subsequently been referred to psychology for 'non-compliance'. She remembered this as being undermining, disappointing, and feeling like a punishment: *"It was a crap situation, [. . .] it doesn't seem like a massive endorsement of your character to be referred back to psychology"* (Charlotte).

Theme 2: Healthcare professionals do not recognise the importance of context

Participants believed that healthcare professionals often failed to recognise the importance of context when treatment-taking was discussed. For example, healthcare professionals were viewed as failing to work collaboratively with, or to consider the demands made of the patient by other members of the CF multidisciplinary team (MDT). The overwhelming feeling that can result is summed up by Jenny:

> *"The problem is you've got all these different specialists and they all want to try and help and so you have in one clinic, you can have the physio come in, talk about 'how much are you doing? Are*

> *you doing everything you're supposed to do?... How many times a day are you doing physio? How long do you spend doing it?... Then you have the dietitian come in and the dietitian says, 'How are you managing to eat and how is your digestion, and are you having your insulin? Maybe if you have your insulin an hour earlier, maybe if you have two units, maybe we'll try this and will try that'… And then you have the pharmacist come in and then you have someone else come in… And it's just, it's a constant, you know, each of them have their little targets… But what they don't realise is that you've got five people doing exactly [the same]"* (Jenny).

There was also a sense that healthcare professionals told patients what to do or 'talked at' rather than 'with' participants. The consensus was that healthcare professionals failed to see the 'bigger picture' and understand that "*life is not just about treatments, that there has to be a balance to have a normal life as well"* (Alice). Healthcare professionals were often perceived as being too busy to take the time to get to know the patient. When healthcare professionals did not truly listen to the patient or made assumptions without considering patient life context, it was disheartening for patients, who were unable to implement what were for them, inappropriate suggestions. As a result, when such suggestions were made, participants suggested it was easier to just go along with them, even though they had no intention of trying them, for fear of being viewed as a 'bad patient'. Jenny summed this tension up:

> *"you end up going down a road that you don't wanna go down where it's… it's then suddenly something that they want to help you fix [. . .]. Your entire life is a constant, 'well, maybe if you tried doing this, maybe if you tried doing that...' And none of those things ever really work so all you end up doing is just saying, 'yes, OK'"* (Jenny).

In contrast, on the occasions when patient life context was considered, participants spoke positively about working in partnership with healthcare professionals who wanted to work with them, collaboratively, to see how treatment-taking could be balanced with their other priorities. This helped participants to feel like they had a more active role in treatment-taking. Sarah remembered:

> *"When I had my first year at uni, my doctors were helping me brainstorm ways that I could do my overnight feeds and to get the calories in whilst also being able to go out with friends in the evening and go to 9am lectures and it was that kind of thing that really helped me to have a bit more of a positive relationship with the treatment, but also make me more likely to do it because it felt like, you know, we had come to an agreement that everybody was happy with, that also let me do what I wanted to do"* (Sarah).

However, such truly collaborative interactions were rarely mentioned by participants. Instead, healthcare professionals were perceived as wanting to reduce treatment-taking to a number, which did not take into account the variability and complexity of adherence. Richard found this reductive approach perplexing and confusing:

> *"They sort of want to have it like a score... It's odd… they talk about compliance and then want like an exact figure on it, whereas maybe it's not quite so, you know, scientific as that"* (Richard).

This reductive approach also served to reinforce a belief that healthcare professionals sometimes only focused on the amount of treatment they were taking and did not understand or acknowledge context and the complex range of factors that influence adherence. This was disempowering and upsetting for participants. Charlotte recalls:

> *"When I got re-referred to psychology a few years ago, and it was like oh 'a referral for non-compliance'. Ouch. Just really like, um, really dismissive and as I keep banging on about, no context. No, you know, 'this is what's happening at work', or 'this is what's happening at home', or like, 'we've just increased her treatment regimen by four hours'… No, just 're-referral for non-compliance'… you just feel very small in that, like you're not really in there"* (Charlotte).

Theme 3: 'Admitting' non-adherence is difficult

It was evident that being a 'compliant patient' was important to participants' self-identity: "*I don't want [them] to have a perception of me as being non-compliant*" (Charlotte). Indeed, participants were aware of normative expectations to take treatments: *"you grow up knowing that you're supposed to do all these things, and if you don't do them, then you're, then it's naughty or it's bad"* (Jenny).

Participants also understood the importance of being honest:

> *"I think what the doctors probably want is for you to be honest… so if I come to clinic and I've got a flare up, it's probably more helpful for the doctor to know that I have missed my antibiotics all last week, whereas if I haven't, if I've been taking my antibiotics and I still got a flare up, then I probably need IVs"* (Charlotte).

Participants said they were more inclined to be honest with healthcare professionals if they felt they could have an open conversation, free from judgement, with healthcare professionals who were personable, collaborative, and treated them as an equal partner:

> *"There are a couple of inhalers that I always seem to forget in the mornings… I said this to my physio, and she's great, like she's someone who's super open and chatty and so I felt like I could*

> *really talk to her about it and it's very much her personality that made me feel OK with bringing it up"* (Nicola).

However, few such positive experiences were reported by participants. Instead, participants said that healthcare professionals often responded to adherence difficulties by assuming participants were ill-informed and launching into a lecture highlighting the importance of treatments and the consequences of non-adherence: *"I know I'll get a lecture and I… I don't want it, I don't want a lecture…"* (Sally).

Healthcare professionals were often perceived as being blaming and judgemental, e.g., *"I think most people feel like healthcare professionals tell them to take their treatments and judge them for not taking them"* (Nicola). This resulted in participants not feeling safe to talk openly with healthcare professionals about the amount of treatment they were taking. Jenny explains:

> *"I don't think I've ever been in a circumstance where I've thought that I could be totally honest without any form of judgment. I don't think that's existed… yeah, I don't think I've ever experienced a time where I felt totally confident to say exactly, lay everything out on the table. Never experienced that"* (Jenny).

Some participants even described healthcare professionals who had been angry with them after they shared their adherence difficulties:

> *"The doctor said, you know, 'how is your compliance?', and I said, 'well it's not great'… And he was actually really cross. And he got quite – in fact it's one of the reasons I moved clinic to be honest - he was, you know, 'you need to, you need to be on this, you need to be compliant, you know, you've got to look to the future, the transplant, if you're non-compliant now…', and it was actually quite aggressive and then eventually I got quite upset"* (Alice).

These interactions left participants feeling ashamed and guilty about their adherence difficulties which made it less likely they would talk about their adherence struggles with healthcare professionals in the future:

> *"I don't want them [CF team] to know because I feel guilty, that's what it is… They've put in all this hard work prescribing you what you need and, you know, telling you what's gonna work and what's not [. . .], I don't want 'em to be disappointed in me, 'cause I'm disappointed in myself when I don't take it, so I think for someone else to be disappointed in you and for you to see that look on their face, I think just makes you feel a bit meh"* (Sally).

Some participants believed that there was a limited quota of support available from their CF team and that if they were labelled as ‘non-compliant’ they would not be able to access ongoing support (*“Healthcare professionals only have like a certain repertoire of things they can do to help and then you're essentially on your own”*; Nicola). Once labelled as ‘non-compliant’, participants felt they were unable to shake off that label, e.g., “*even if you have [done everything], it's very hard to convince them that you're being compliant if you have a history of non-compliance”* (Charlotte). Furthermore, and potentially more worryingly, since healthcare professionals were seen as gatekeepers, there was a fear that being labelled ‘non-compliant’ could reduce participants’ access to new treatments or lung-transplants. For example, Richard wondered: *“maybe if I’m non-compliant and the hospital knows it, does that like put me down the pecking order when a new drug comes out?”* and Simon speculated: *“[. . .] it’s something that they could maybe use against you towards not putting you on the transplant list”.*

Participants talked about the challenges of being asked to self-report their adherence, which usually involved *“guesstimating”* (Lottie) the amount of treatment being taken (or not taken), e.g., “*you sort of end up feeling like you should say something rather than nothing, but you might not be right… I might be underselling what I do or overselling what I do because I don't always know”* (Jenny). Healthcare professionals were sometimes viewed as pedantic, picking faults with patients or trying to catch them out, e.g., *“some people with CF probably see the whole healthcare team as um combatants, you know, ‘they're always trying to pick me up on this or that’”* (Charlotte). Participants seemed to feel pressurised into saying the ‘right’ thing or reporting the ‘correct’ amount of treatment being taken which, as Jenny summarises, could have an impact on their ability to talk openly: *“It's like a test. Every single time is a test. And so, you know you have to get the answers right. It's about, it's more about getting the answers right than actually saying what you're doing”* (Jenny). This has potentially serious implications; for example, healthcare professionals may be unable to differentiate health deterioration due to low adherence from health deterioration despite high adherence (suggesting there may be other underlying causes of decline). Sally’s experience reflects this:

> *“So you’d drink them [Scandishakes] and then you’d eat and you’d do your best, but this weight just wouldn't go on and they would just nag at you constantly 'cause they say ‘you're not eating enough, you must be missing them’. ‘Well no, I'm not missing them, they're just not working!’, and that used to really wind me up”* (Sally).

Objective measures of adherence (e.g., data-logging nebulisers or pill bottles) were perceived as removing the need for participants to self-report the amount of treatment taken and providing more accurate information. Participants who had tried automated data transfer approaches found they reduced the pressure on them to remember accurately: *“I know I'm terrible at remembering, so it's quite nice to know that it’s something I don't really have to think about anymore”* (Rosie).

Discussion

In this study, we explored the views of adults with CF on the way in which treatment-taking, or adherence, is discussed in routine CF care. Although there is a desired shift in healthcare towards patient-centred care (NICE, 2021; WHO, 2015), in which patients are treated as equal partners who collaborate with their healthcare providers (NEJM Catalyst, 2017), this was not reflected in our participants' experiences.

The language used by healthcare professionals in relation to treatment-taking was considered important (Theme 1), with 'compliance' having negative connotations due to its association with authority and a lack of freedom or control. The word 'compliance' was associated with an imbalance of power as in a parent-child relationship between patients and healthcare professionals. Patients who did not 'comply' were viewed as disobedient. Although it has been suggested that 'adherence' may be a more appropriate term, as it emphasises the need for agreement between the patient and healthcare professional (Horne et al., 2005), both terms were viewed negatively because they made participants feel passive in their own care. The use of shaming, stigmatising language by healthcare professionals in response to non-adherence (e.g., the patient 'disclosed', 'admitted', or was 'non-compliant') reinforces this, even though it has been suggested that non-adherence should be viewed as "the norm rather than the exception" (CF Trust, 2018; 8:25), and as "a problem of humans, rather than simply a problem of 'naughty' patients" (Wildman & Hoo, 2014, p. 16). 'Concordance' is a more recent term, which highlights shared decision-making between the patient and healthcare professional, but no participant in this study was familiar with the term, suggesting it may not be widely used in practice. As highlighted by Brunton (2017), it is possible that "we have simply slapped a new label ("non-adherent") over the old "non-compliant" label, but we are still blaming and shaming our patients" (p.76). Indeed, whilst language is important, the endless cycling of terms which become more negative over time (pejoration) is perhaps a smoke screen for the paradigm shift needed to overcome how the stigmatised behaviour (non-adherence) is seen.

Our study clearly shows the difficulty patients face managing their complex condition alongside the demands of their day-to-day lives. Participants believed that healthcare professionals sometimes failed to see the 'bigger picture' and instead focused on the amount of treatment they were taking without recognising context and the complex range of factors that influence adherence (Theme 2). This is consistent with a finding by Sawicki et al. (2015) in which adolescents with CF in the US wanted recognition from healthcare professionals that they were balancing CF and treatments along with 'normal' life (e.g., university, work, social life). Behaviour change is complex and rarely straightforward (Kelly & Barker, 2016), and healthcare professionals need to understand patient life context in order to make relevant and appropriate recommendations. This may be challenging since many participants suggested that healthcare professionals were 'too busy' and there was often limited time to discuss what the patient wanted to discuss during clinic consultations. In addition, healthcare professionals often lack the confidence, motivation and training to deliver behaviour change interventions (Bull & Dale, 2020), and there have historically been few interventions that increase adherence (Nieuwlaat et al., 2014). This leads to an inevitable clash between patient behaviour/needs and healthcare professional targets/skills, since healthcare professionals have a limited understanding of how to effectively support patient behaviour change.

In addition, participants suggested that healthcare professionals sometimes failed to consider the patient in the wider healthcare context. UK CF teams comprise a range of specialists, including consultants, nurses, physiotherapists and dieticians (CF Trust, 2011). Participants reported receiving mixed messages from members of the MDT in relation to treatment-taking priorities. Participants felt this reflected a disjointed health system that failed to consider the patient as a 'whole person'. This is potentially problematic since, as highlighted by Riekert et al. (2015), "too many cooks in the kitchen" (p. 146) can mean that the patient is left to decide the best course of action in relation to treatment-taking.

It is perhaps not surprising, given Themes 1 ('The power of language') and 2 ('Healthcare professionals do not recognise the importance of context'), that participants found it difficult to 'admit' non-adherence (Theme 3). Consistent with previous research (Drabble et al., 2019), participants in our study showed an awareness of normative expectations to take treatments because it was rational to maintain 'good health'. During the interviews, participants 'othered' 'non-compliant' patients and framed their own non-adherence in ways which allowed them to maintain their identity as a 'compliant' patient. This may have been a strategy aimed at helping them to overcome the 'moral dilemma' of maintaining their identity as a 'good patient' even when they were not adhering to their treatment (Drabble et al., 2019). Our study expands on this previous research by finding that, in addition to normative expectations to take treatments, there were normative expectations among participants that patients 'should' be honest, and that it was in the interests of healthcare professionals to have an accurate understanding of adherence. This therefore creates an even bigger 'moral dilemma' for non-adherent patients; how do they maintain their identity as 'good' patients who take their treatments but who are also honest with healthcare professionals about the amount of treatment (not) being taken?

Participants spoke of previous encounters with healthcare professionals in which they had been judged, lectured, or 'told off' for admitting non-adherence. This is consistent with previous reports of conflict between people with CF and healthcare professionals in relation to low adherence (Arden et al., 2019). Blame and judgement shuts down conversations (Dickinson & Maryniuk, 2017) and may lead to a lack of trust which could influence the extent to which people with CF feel they can talk openly again in the future. This may lead to people with CF making changes to their treatment regimen themselves without seeking the advice of healthcare professionals, which could have clinical implications. Whilst guilt may be a driver of adherence in some individuals (George et al., 2010), lecturing or eliciting fear by confronting patients with the negative consequences of non-adherence in an attempt to increase motivation is a risky strategy and could simply lead to avoidance (Kok et al., 2018). In our study, consistent with Knudsen et al. (2018), many participants spoke about strong feelings of guilt and shame for not taking their treatments. These findings suggest that there is some way to go in meeting NICE (2009) and WHO (2003) recommendations that healthcare professionals use a patient-centred, non-judgemental approach to promote open and honest conversations in relation to adherence.

Furthermore, some participants believed that CF teams had a limited quota of adherence support that they could access, suggesting that people with CF may find it difficult to reach out for or accept support when needed. Participants described times when healthcare professionals had made assumptions and ‘problem-solved’ for them. Suggesting solutions can disempower or insult the patient as it fails to consider the patient’s life context and assumes both that the healthcare professional knows best, and that the patient has not thought of the ‘solution’ themselves (Robertson, 2005). Patients who do not feel listened to are also less likely to follow healthcare professionals’ advice (Butler et al., 1998). Reasons for non-adherence are complex and a simple, ‘one-size fits all' solution or intervention is unlikely to be effective at increasing adherence (Arden et al., 2019). Our study adds weight to previous research (e.g., Arden et al., 2019; Sawicki et al., 2015), showing that people with CF feel more able to talk openly and ‘admit’ non-adherence to healthcare professionals who are personable, open, non-judgemental, and collaborative.

Clinical implications

This study illustrates the role of healthcare professionals in supporting adherence in adults with CF. This could influence CF care and the development of interventions to support treatment-taking in people with CF. Often the onus is on the patient to ‘be honest’ about the amount of treatment they are taking and if a patient does not ‘disclose’ their non-adherence, they are perceived as “attempting to deceive clinicians” (p. 16) who are doing their best to care for them (Wildman & Hoo, 2014). Our findings do not support the notion that people with CF are motivated by an intention to mislead healthcare professionals. Rather, we suggest that ‘adherence’ is a metric that makes visible the extent to which MDTs are underserving a population with a long-term condition. In line with WHO (2003), we suggest that healthcare provider and system-level factors that contribute to non-adherence ought to be further explored. Just as patients need the capability, opportunity and motivation in order to adhere to their treatments (Jackson et al., 2014), healthcare professionals need the capability (e.g., skills to achieve sustained behaviour change), opportunity (e.g., time), and motivation to effectively support patient adherence (Girling et al., 2022).

As highlighted elsewhere (e.g., Cox & Fritz, 2022), healthcare professionals need to consider the language they use with patients, as this can have an influence on the power dynamic and patient-practitioner relationship. Further research on healthcare professionals' views and experiences of the impact of language on patients is needed. Approaches used in other fields such as the adoption of 'People First Language' in diabetes could be replicated to move away from words that judge or label towards words that are strength-based, person-centred, and empowering (Dickinson et al., 2017). We recommend that this is an approach that could be adopted in CF care, particularly in relation to treatment-taking. This might include replacing judgemental terms like 'good' and 'bad' with more factual terms like 'high' and 'low'; avoiding use of the terms 'compliance', 'compliant' and 'non-compliant', and shaming terms like 'admitted' and 'disclosed'; and minimising use of the term 'adherence' in favour of lay terms like 'treatment-taking'.

Where available, objective measures of adherence could remove some of the issues identified in discussing adherence by providing people with CF (and their clinical teams) with factual information, rather than information based on judgements (Dickinson et al., 2017). It is well known that patient self-report is unreliable (e.g., Daniels et al., 2011; Modi et al., 2006) and is subject to social desirability and recall bias (Osterberg & Blaschke, 2005). Indeed, in our study, participants highlighted the challenges of being asked to accurately recall the amount of treatment being taken. Objective adherence measures could empower people with CF to have more of an active role in treatment-taking, provided that participants have control over their data and can decide whether to share this with the CF team, to overcome concerns over "Big Brother" watching (Campbell et al., 2016, p. 338). Having an accurate measure of adherence can influence clinical decision-making and targeting of resources (WHO, 2003). However, as with all measures of adherence, it is crucial that the data is used to empower and support patient behaviour change rather than as a tool to chasten people with CF, which will only amplify existing power imbalances.

Our recommendations are focused on changing the behaviour of healthcare professionals. This, it is noted, is no less challenging (Potthoff et al., 2022) than changing the behaviour of 'patients'. A culture change is needed (Mannion & Davies, 2018) to change, not only the language used by healthcare professionals (Cox & Fritz, 2022), but also entrenched healthcare professional views and beliefs about patients who do not take their treatments. An approach where healthcare professionals work collaboratively with people with CF to balance treatments along with other life priorities is required. Due to the number of different specialisms involved in delivering CF care (CF Trust, 2011), this needs to be a joined-up MDT approach. Such an approach could be facilitated by the appointment of a 'behaviour change champion', for example a Health Psychologist, or someone who has the knowledge, skills, and training to work holistically in the MDT to support patient (and healthcare professional) behaviour change. In addition, as recommended by the Medical Research Council (MRC), behaviour change is a complex intervention requiring a coherent conceptual framework (Craig et al., 2008). Having a conceptual framework can enable healthcare professionals to consider the barriers to adherence and to assess how an intervention might act on these barriers to produce and then maintain adherence (e.g., through habit formation; Arden et al., 2021). One such conceptual framework (Arden et al., 2021) has already been developed and tested in CF, in a 19-centre, 607-patient randomised-controlled trial which found a significant increase in adherence to inhaled therapies among adults with CF alongside a decrease in perceived treatment burden (Wildman et al., 2021).

Lastly, it is important to note that our study was conducted shortly after elexacaftor-tezacaftor-ivacaftor was commissioned for use in the UK. The availability of CFTR modulator treatments appears to have changed the landscape for many people with CF. Healthcare professionals may therefore need to be even more mindful when supporting treatment-taking in a cohort of people on modulator treatments who may be feeling more well and leading a more ‘normal’ life, compared to previous generations of people with CF whose outlooks were perhaps somewhat different. It is plausible that these changes could even lead to a lack of treatment-taking for some treatments outside of modulator treatments (Keyte et al., 2022), with questions such as ‘how much other treatment is enough?’ becoming pertinent in the CFTR-modulator era (Sandler et al., 2023). In addition, since elexacaftor-tezacaftor-ivacaftor is a high-cost drug, and the annual cost to the NHS is thought to be in the region of £100,000 per patient (BBC, 2022), it is crucial that people with CF feel empowered to talk openly about any adherence challenges, to ensure they can access appropriate support as required.

Strengths and limitations

Our study expands on previous research (e.g., Arden et al., 2019; Sawicki et al., 2015) by exploring in more detail the communication of healthcare professionals and the potential impact this can have on the adherence of adults with CF. To our knowledge, this is the first study that explores the views of adults with CF on language use in relation to treatment-taking.

However, the study also had limitations. Firstly, since participants were recruited through Twitter due to the COVID-19 pandemic, it was a self-selected sample and the views of those who volunteered to participate may be different to other adults with CF. Future research should explore the views of ‘under-served’ people with CF. Secondly, although the purpose of our study was to explore participant views about *the way* in which treatment-taking is discussed in routine CF care, rather than to explore views about any specific CF treatments in particular, it may have been helpful to collect data on the type and number of treatments participants were taking. It is possible, for example, that those on

CFTR modulator treatments or prescribed a greater number of treatments may have slightly different views to other adults with CF. Thirdly, most participants described having high adherence, with some saying they rarely missed a treatment. Whilst self-reported adherence is notoriously inaccurate (e.g., Daniels et al., 2011; Modi et al., 2006), and all participants described times when they had missed treatments, it would have been helpful to have an objective measure of patient adherence to supplement the interviews (as per Arden et al.'s (2019) study). Although the focus in this study was not on one specific treatment type (e.g., inhaled therapies), and objective adherence data is not routinely available for many treatments, having a measure of adherence may have enabled the identification of differences between participants with low and high adherence, which could be targeted in the development of future interventions.

Additionally, despite CF being equally common in both sexes (CF Trust, n.d.), the sample of participants in this study was predominantly female (n=10; 83%). It is possible that our findings relate more to women than to men, although the themes that were developed were apparent in the data from the small number of males in the study, as well as the females. Additionally, most participants were adults in their 30s (n=8; 67%), so it would be interesting to explore the views of people of other ages (e.g., younger people who have recently transitioned from paediatrics to adults, which is often highlighted as a challenging time in CF (Nazareth & Walshaw, 2013)).

Conclusion

This study highlights that a culture change is needed in CF care such that people who struggle to take their treatments or meet the high demands placed on them are not labelled as 'disobedient children', wilfully disobeying orders from healthcare professionals in positions of authority. Instead, an open, honest, non-judgemental approach should be adopted, where healthcare professionals work collaboratively with people with CF to balance treatments along with other life priorities. Although using this approach in healthcare has been recommended by NICE (2009) for over a decade, our findings highlight how this may not always be used in practice. Adherence is a skillset that can be built, and healthcare professionals themselves need the skills to know how best to support adherence and patient behaviour change. CF is an archetypal, multi-morbid, long-term condition, and the findings from this study could have implications for patient-practitioner communication in other chronic, complex health conditions.

References

Arden, M. A., Drabble, S., O'Cathain, A., Hutchings, M., & Wildman, M. (2019). Adherence to medication in adults with Cystic Fibrosis: An investigation using objective adherence data and the Theoretical Domains Framework. *British Journal of Health Psychology, 24*(2), 357-380. https://doi.org/10.1111/bjhp.12357

Arden, M. A., Hutchings, M., Whelan, P., Drabble, S. J., Beever, D., Bradley, J. M., Hind, D., Ainsworth, J., Maguire, C., Cantrill, H., O'Cathain, A., & Wildman, M. (2021). Development of an intervention to increase adherence to nebuliser treatment in adults with cystic fibrosis: CFHealthHub. *Pilot and Feasibility Studies, 7*(1), 1. https://doi.org/10.1186/s40814-020-00739-2

BBC (2022). NHS spending: How much money does it need? Retrieved from https://www.bbc.co.uk/news/health-59865822

Bell, S. C., Mall, M. A., Gutierrez, H., Macek, M., Madge, S., Davies, J. C., Burgel, P. R., Tullis, E., Castaños, C., Castellani, C., Byrnes, C. A., Cathcart, F., Chotirmall, S. H., Cosgriff, R., Eichler, I., Fajac, I., Goss, C. H., Drevinek, P., Farrell, P. M., Gravelle, A. M., … Ratjen, F. (2020). The future of cystic fibrosis care: a global perspective. *The Lancet Respiratory Medicine, 8*(1), 65-124. https://doi.org/10.1016/S2213-2600(19)30337-6

Braun, V. & Clarke, V. (2019a). *Answers to frequently asked questions about thematic analysis.* https://cdn.auckland.ac.nz/assets/psych/about/our-research/documents/Answers%20to%20frequently%20asked%20questions%20about%20thematic%20analysis%20April%202019.pdf

Braun, V., & Clarke, V. (2019b). Reflecting on reflexive thematic analysis. *Qualitative Research in Sport, Exercise and Health, 11*(4), 589-597. https://doi.org/10.1080/2159676X.2019.1628806

Braun, V., & Clarke, V. (2021). To saturate or not to saturate? Questioning data saturation as a useful concept for thematic analysis and sample-size rationales. *Qualitative Research in Sport, Exercise and Health, 13*(2), 201-216. https://doi.org/10.1080/2159676X.2019.1704846

Braun, V., & Clarke, V. (2022). Thematic Analysis: A practical guide. London: Sage

Britto, M. T., Kotagal, U. R., Hornung, R. W., Atherton, H. D., Tsevat, J., & Wilmott, R. W. (2002). Impact of recent pulmonary exacerbations on quality of life in patients with cystic fibrosis. *Chest, 121*(1), 64-72. https://doi.org/10.1378/chest.121.1.64

Brunton, S. (2017). I have never liked the term “compliance”. *Clinical Diabetes, 35*(2): 76-77. https://doi.org/10.2337/cd17-0010

Bull, E. R., & Dale, H. (2021). Improving community health and social care practitioners’ confidence, perceived competence and intention to use behaviour change techniques in health behaviour change conversations. *Health and Social Care in the Community, 29*, 270-283. https://doi.org/10.1111/hsc.13090

Burgener, E. B., & Moss, R. B. (2018). Cystic fibrosis transmembrane conductance regulator modulators: precision medicine in cystic fibrosis. *Current Opinion in Pediatrics, 30*(3), 372–377. https://doi.org/10.1097/MOP.0000000000000627

Butler, C. C., Pill, R., & Stott., N. C. H. (1998). Qualitative study of patients’ perceptions of doctors’ advice to quit smoking: implications for opportunistic health promotion. *BMJ, 316*, 1878-1881. https://doi.org/10.1136/bmj.316.7148.1878

Calthorpe, R. J., Smith, S. J., Rowbotham, N. J., Leighton, P. A., Davies, G., Daniels, T., Gathercole, K., Allen, L., Elliott, Z. C., & Smyth, A. R., (2020). What effective ways of motivation, support and technologies help people with cystic fibrosis improve and sustain adherence to treatment? *BMJ Open Respiratory Research, 7*: e000601. http://dx.doi.org/10.1136/bmjresp-2020-000601

Campbell, J. I., Eyal, N., Musiimenta, A., & Haberer, J. E. (2016). Ethical questions in medical electronic adherence monitoring. *Journal of General Internal Medicine, 31*, 338–342. https://doi.org/10.1007%2Fs11606-015-3502-4

CF Foundation Patient Registry. (2022, September). *2021 Annual Data Report.* https://www.cff.org/media/23476/download

CF Trust. (2011, December). *Standards for the Clinical Care of Children and Adults with Cystic Fibrosis in the UK.* CF Trust. https://www.cysticfibrosis.org.uk/sites/default/files/2020-12/Cystic%20Fibrosis%20Trust%20Standards%20of%20care.pdf

CF Trust. (2018, September 18). CF Trust. *How do we ensure the person is at the centre of their care?* [Video]. https://www.cysticfibrosis.org.uk/video/nojs/4361

CF Trust. (n.d.). *Cystic fibrosis FAQs.* CF Trust. https://www.cysticfibrosis.org.uk/what-is-cystic-fibrosis/faqs#How%20common%20is%20cystic%20fibrosis

Cooper, A., & Swindell, B. (2021, June 18). *The impact of language on people living with long-term conditions: having the rug pulled out from underneath you.* Evidently Cochrane. https://www.evidentlycochrane.net/the-impact-of-language-on-people-living-with-long-term-conditions/?mc_cid=2fd4b1b382&mc_eid=b28ec065be

Corkhill, B. (2018). *Pain signals and other bad language!* https://www.betsan.org/wp-content/uploads/2018/07/Pain-signals-and-other-bad-language.pdf

Cox, C., & Fritz, Z. (2022). Presenting complaint: use of language that disempowers patients. *BMJ, 377*, e066720. https://doi.org/10.1136/bmj-2021-066720

Craig, P., Dieppe, P., Macintyre, S., Michie, S., Nazareth, I., Petticrew, M., & Medical Research Council Guidance (2008). Developing and evaluating complex interventions: the new Medical Research Council guidance. *BMJ, 337*, a1655. https://doi.org/10.1136/bmj.a1655

Daniels, T., Goodacre, L., Sutton, C., Pollard, K., Conway, S., & Peckham, D. (2011). Accurate assessment of adherence: Self-report and clinician report vs electronic monitoring of nebulizers. *Chest, 140*, 425–432. https://doi.org/10.1378/chest.09-3074

Dickinson, J. K., Guzman, S. J., Maryniuk, M. D., O'Brian, C. A., Kadohiro, J. K., Jackson, R. A., D'Hondt, N., Montgomery, B., Close, K. L., & Funnell, M. M. (2017). The use of language in diabetes care and education. *Diabetes Care, 40*, 1790-1799. https://doi.org/10.2337/dci17-0041

Dickinson, J. K., & Maryniuk, M. D. (2017). Building therapeutic relationships: Choosing words that put people first. *Clinical Diabetes, 35*(1), 51-54. https://doi.org/10.2337%2Fcd16-0014

Drabble, S., O'Cathain, A., Arden, M., Hutchings, M., Beever, D., & Wildman, M. (2019). When is forgetting not forgetting? A discursive analysis of differences in forgetting talk between adults with cystic fibrosis with different levels of adherence to nebulizer treatments. *Qualitative Health Research, 29*(14), 2119-2131. https://doi.org/10.1177/1049732319856580

Eakin, M. N., Bilderback, A., Boyle, M. P., Mogayzel, P. J., & Riekert, K. A. (2011). Longitudinal association between medication adherence and lung health in people with cystic fibrosis. *Journal of Cystic Fibrosis, 10*, 258-264. https://doi.org/10.1016/j.jcf.2011.03.005

George, M., Rand-Giovannetti, D., Eakin, M. N., Borrelli, B., Zettler, M., & Riekert, K. A. (2010). Perceptions of barriers and facilitators: Self-management decisions by older adolescents and adults with CF. *Journal of Cystic Fibrosis, 9*(6), 425-432.

https://doi.org/10.1016/j.jcf.2010.08.016

Girling, C., Packham, A., Robinson, L., Arden, M. A., Hind, D., & Wildman, M. (2022). Implementing the use of objective medication adherence data in routine clinical practice via the digital CFHealthHub platform: situation analysis and strategy development using the theoretical domains framework. *Implementation Science Communications*, *3*, 12. https://doi.org/10.1186/s43058-022-00263-9

Haskard-Zolnierek, K. B. & DiMatteo, M. R. (2009). Physician communication and patient adherence to treatment: A meta-analysis. *Medical Care*, *47*(8), 826-834. https://doi.org/10.1097/mlr.0b013e31819a5acc

Haynes, R.B., Sackett, D.L., & Taylor, D.W. (Eds.). (1979). *Compliance in health care.* Baltimore, MD: Johns Hopkins University Press.

Heijerman, H. G. M., McKone, E. F., Downey, D. G., Van Braeckel, E., Rowe, S. M., Tullis, E., Mall, M. A., Welter, J. J., Ramsey, B. W., McKee, C. M., Marigowda, G., Moskowitz, S. M., Waltz, D., Sosnay, P. R., Simard, C., Ahluwalia, N., Xuan, F., Zhang, Y., Taylor-Cousar, J. L., McCoy, K. S., … VX17-445-103 Trial Group (2019). Efficacy and safety of the elexacaftor plus tezacaftor plus ivacaftor combination regimen in people with cystic fibrosis homozygous for the F508del mutation: a double-blind, randomised, phase 3 trial. *Lancet, 394*(10212), 1940–1948. https://doi.org/10.1016/S0140-6736(19)32597-8

Hoffman, K. (2015, June 11). *“Compliance?” “Adherence?” The Language of Medication Taking.* Medivizor. https://medivizor.com/blog/2015/06/11/language-of-medication-taking/

Hoo, Z. H., Totton, N., Waterhouse, S., Lewis, J., Girling, C., Bradburn, M., Arden, M. A., Whelan, P., Ainsworth, J., Dawson, S., Millward, S., Barnett, K., Dewar, J., Barr, H. L., Saini, G., Shepherd, E., Carroll, M., Allenby, M. I., Daniels, T. V., Nightingale, J. A., … Wildman, M. J. (2021). Real-world adherence among adults with cystic fibrosis is low: a retrospective analysis of the CFHealthHub digital learning health system. *Chest, 160*(6), 2061-2065. *https://doi.org/10.1016/j.chest.2021.06.039*

Horne, R., Weinman, J., Barber, N., Elliott, R., Morgan, M., Cribb, A., et al. (2005). *Concordance, adherence and compliance in medicine taking: report for the National Co-ordinating Centre for NHS Service Delivery and Organisation R&D (NCCSDO).*

Jackson, C., Eliasson, L., Barber, N., & Weinman, J. (2014). Applying COM-B to medication adherence. *European Health Psychologist, 16*, 7-17.

Kelly, M., & Barker, M. (2016). Why is changing health-related behaviour so difficult? *Public Health, 136*, 109-116. https://doi.org/10.1016%2Fj.puhe.2016.03.030

Keyte, R., Kauser, S., Mantzios, M., & Egan, H. (2022). The psychological implications and health risks of cystic fibrosis pre- and post- CFTR modulator therapy. *Chronic Illness*, 17423953221099042. https://doi.org/10.1177/17423953221099042

Knudsen, K. B., Boisen, K. A., Katzenstein, T. L., Mortensen, L. H., Pressler, T., Skov, M., & Jarden, M. (2018). Living with cystic fibrosis - a qualitative study of a life coaching intervention. *Patient Preference and Adherence, 12*, 585–594. https://doi.org/10.2147/ppa.s159306

Kok, G., Peters, G. Y., Kessels, L., Ten Hoor, G. A., & Ruiter, R. (2018). Ignoring theory and misinterpreting evidence: the false belief in fear appeals. *Health Psychology Review, 12*(2), 111–

125. https://doi.org/10.1080/17437199.2017.1415767

Lask, B. (1994). Non-adherence to treatment in cystic fibrosis. *Journal of the Royal Society of Medicine, 21*(87), 25-27.

Mannion, R., & Davies, H. (2018). Understanding organisational culture for healthcare quality improvement. *BMJ, 363*, k4907. https://doi.org/10.1136/bmj.k4907

Maxwell, J. A. (2012). *A Realist Approach to Qualitative Research.* London: Sage.

Middleton, P. G., Mall, M. A., Dřevínek, P., Lands, L. C., McKone, E. F., Polineni, D., Ramsey, B. W., Taylor-Cousar, J. L., Tullis, E., Vermeulen, F., Marigowda, G., McKee, C. M., Moskowitz, S. M., Nair, N., Savage, J., Simard, C., Tian, S., Waltz, D., Xuan, F., Rowe, S. M., … VX17-445-102 Study Group (2019). Elexacaftor-Tezacaftor-Ivacaftor for Cystic Fibrosis with a Single Phe508del Allele. *The New England Journal of Medicine, 381*(19), 1809–1819. https://doi.org/10.1056/NEJMoa1908639

Mitchell, R. M., Jones, A. M., Stocking, K., Foden, P., & Barry, P. J. (2021). Longitudinal effects of ivacaftor and medicine possession ratio in people with the Gly551Asp mutation: a 5-year study. *Thorax, 76*(9), 874–879. https://doi.org/10.1136/thoraxjnl-2020-215556

Modi, A. C., Lim, C. S., Yu, N., Geller, D., Wagner, M. H., & Quittner, A. L. (2006). A multi-method assessment of treatment adherence for children with cystic fibrosis. *Journal of Cystic Fibrosis, 5*, 177-185. https://doi.org/10.1016/j.jcf.2006.03.002

Nasr, S. Z., Chou, W., Villa, K. F., Chang, E., & Broder, M. S. (2013). Adherence to dornase alfa treatment among commercially insured patients with cystic fibrosis. *Journal of Medical Economics, 16*(6), 1-8. https://doi.org/10.3111/13696998.2013.787427

Nazareth, D., & Walshaw, M. (2013). Coming of age in cystic fibrosis – transition from paediatric to adult care. *Clinical Medicine, 13*(5), 482-486. https://doi.org/10.7861%2Fclinmedicine.13-5-482

NEJM Catalyst. (2017, January 1). *What is patient-centred care?* NEJM Catalyst. https://catalyst.nejm.org/doi/full/10.1056/CAT.17.0559

National Institute for Health and Care Excellence (NICE). (2009, January 28). *Medicines adherence: involving patients in decisions about prescribed medicines and supporting adherence.* NICE. https://www.nice.org.uk/guidance/cg76

NICE (2021). *Patient experience in adult NHS services: improving the experience of care for people using adult NHS services.* NICE. https://www.nice.org.uk/guidance/cg138/chapter/1-guidance#tailoring-healthcare-services-for-each-patient

Nickel, B., Barratt, A., Copp, T., Moynihan, R., & McCaffery, K. (2017). Words do matter: a systematic review on how different terminology for the same condition influences management preferences. *BMJ Open, 7*, e014129. https://doi.org/10.1136/bmjopen-2016-014129

Nieuwlaat, R., Wilczynski, N., Navarro, T., Hobson, N., Jeffery, R., Keepanasseril, A., Agoritsas, T., Mistry, N., Iorio, A., Jack, S., Sivaramalingam, B., Iserman, E., Mustafa, R. A., Jedraszewski, D., Cotoi, C., & Haynes, R. B. (2014). Interventions for enhancing medication adherence. *Cochrane Database of Systematic Reviews*, 11, CD000011. https://doi.org/10.1002/14651858.CD000011.pub4

Osterberg, L., & Blaschke, T. (2005). Adherence to medication. *The New England Journal of Medicine, 353*, 487-497. https://doi.org/10.1056/nejmra050100

Potthoff, S., Kwasnicka, D., Avery, L., Finch, T., Gardner, B., Hankonen, N., Johnston, D., Johnston, M., Kok, G., Lally, P., Maniatopoulos, G., Marques, M. M., McCleary, N., Presseau, J., Rapley, T., Sanders, T., Ten Hoor, G., Vale, L., Verplanken, B., & Grimshaw, J. M. (2022). Changing healthcare professionals' non-reflective processes to improve the quality of care. *Social Science & Medicine (1982), 298*, 114840. https://doi.org/10.1016/j.socscimed.2022.114840

Quittner, A. L., Zhang, J., Marynchenko, M., Chopra, P. A., Signorovitch, J., Yushkina, Y., & Riekert, K. A. (2014). Pulmonary medication adherence and health-care use in cystic fibrosis. *Chest, 146*, 142-151. https://doi.org/10.1378/chest.13-1926

Riekert, K. A., Eakin, M. N. Bilderback, A., Ridge, A. K., & Marshall, B. C. (2015). Opportunities for cystic fibrosis care teams to support treatment adherence. *Journal of Cystic Fibrosis, 14*(1), 142-148. https://doi.org/10.1016/j.jcf.2014.10.003

Robertson, K. (2005). Active listening: more than just paying attention. *Australian Family Physician, 34*(12), 1053-5.

Sandler, R. D., Hoo, Z. H., & Wildman, M. J. (2023). P209 Personalised data-Linkage Understanding Treatment Optimisation (PLUTO) in the CFHealthHub Learning Health System: understanding how much is enough for normal life expectancy in the post-modulator era. *Journal of Cystic Fibrosis, 22*(S2), S128-S129.

Sawicki, G. S., Seller, D. E., & Robinson, W. M. (2009). High treatment burden in adults with cystic fibrosis: Challenges to disease self-management. *Journal of Cystic Fibrosis, 8*, 91-96. https://doi.org/10.1016/j.jcf.2008.09.007

Sawicki, G. S., Heller, K. S., Demars, N., & Robinson, W. M. (2015). Motivating adherence among adolescents with cystic fibrosis: Youth and parent perspectives. *Pediatric Pulmonology, 50*(2), 127-136. https://doi.org/10.1002/ppul.23017

Sawyer, S. M., & Aroni, R. A. (2003). Sticky issue of adherence. *Journal of Paediatrics and Child Health, 39*(1), 2–5. https://doi.org/10.1046/j.1440-1754.2003.00081.x

Siracusa, C.M., Ryan, J., Burns, L., Wang, Y., Zhang, N., Clancy, J. P., Drotar, D. (2015). Electronic monitoring reveals highly variable adherence patterns in patients prescribed ivacaftor. *Journal of Cystic Fibrosis, 14*(5), 621-626. https://doi.org/10.1016/j.jcf.2015.05.009

Thomas, C. (2012, December 13). *Why don’t patients take their meds as prescribed?* Heart Sisters. https://myheartsisters.org/2012/12/13/why-dont-patients-take-their-meds-as-prescribed/

Tilson H. H. (2004). Adherence or compliance? Changes in terminology. *The Annals of Pharmacotherapy, 38*(1), 161–162. https://doi.org/10.1345/aph.1D207

UK CF Registry. (2022). *UK Cystic Fibrosis Registry: 2021 Annual Data Report.* CF Trust. https://www.cysticfibrosis.org.uk/sites/default/files/2022-10/CFT_2021-Annual-Data-Report-WEB.pdf

Wildman, M. J., O'Cathain, A., Maguire, C., Arden, M. A., Hutchings, M., Bradley, J., Walters, S. J., Whelan, P., Ainsworth, J., Buchan, I., Mandefield, L., Sutton, L., Tappenden, P., Elliott, R. A., Hoo, Z. H., Drabble, S. J., Beever, D., & CFHealthHub Study Team (2022). Self-management intervention to reduce pulmonary exacerbations by supporting treatment adherence in adults with cystic fibrosis: a randomised controlled trial. *Thorax, 77*(5), 461–469. https://doi.org/10.1136/thoraxjnl-2021-217594

Wildman, M. J., & Hoo, Z. H. (2014). Moving cystic fibrosis care from rescue to prevention by embedding adherence measurement in routine care. *Paediatric Respiratory Reviews, 15*(S1), 16-18. https://doi.org/10.1016/j.prrv.2014.04.007

World Health Organisation (WHO) (2003). *Adherence to long-term therapies: evidence for action.* WHO. https://www.who.int/chp/knowledge/publications/adherence_full_report.pdf

WHO (2015). *WHO global strategy on people-centred and integrated health services: interim report.* WHO. https://apps.who.int/iris/handle/10665/155002

Word count (including references): 10,001

Acknowledgements

We thank those who participated in the study.

Disclosure statement

The authors report there are no competing interests to declare.

Data availability

The data for this study are available from the corresponding author, upon reasonable request. The data are not publicly available due to containing information that could compromise the privacy of research participants.

Funder information

No funding was received to support the delivery of this project.

Appendices

Appendix 1 – Details of study participants (Table 1)

Table 1. Overview of study participant demographic details*.

ID	Pseudonym	Sex	Age (years)
ID1	Charlotte	Female	33
ID2	Sarah	Female	20
ID3	Nicola	Female	30
ID4	Lottie	Female	32
ID5	Simon	Male	32
ID6	Steph	Female	24
ID7	Rosie	Female	27
ID8	Chloe	Female	27
ID9	Alice	Female	30
ID10**	Sally	Female	36
ID11	Jenny	Female	37
ID12	Richard	Male	33

* All participants were of White British ethnicity.

**ID10 was post-lung transplant.

Appendix 2 – Semi-structured interview schedule

Section A: The first few questions focus on how treatment-taking is talked about. I am especially interested in the language and the terms or labels that people use.

1. What terms have you used, or heard used, in relation to the subject of treatment-taking in healthcare (e.g., by healthcare professionals or members of your CF team)?
    a. What do you think about each of these terms? (e.g., what do you like/ dislike?)

If not already discussed:

2. Have you heard the term 'compliance' used?
    a. What does this term mean to you or make you feel?
    b. What do you like/ dislike about this term?

3. Have you heard the term 'adherence' used?
    a. What does this term mean to you or make you feel?
    b. What do you like/ dislike about this term?

4. Have you heard the term 'concordance' used?
    a. What does this term mean to you or make you feel?
    b. What do you like/ dislike about this term?

5. Of those three terms (compliance, adherence, concordance) which do you like the best and which the least? Can you tell me why?

6. Are there other terms or labels that you think could or should be used when talking about treatment-taking in healthcare?

Section B: The next few questions focus on how treatment-taking is talked about when patients meet with health professionals.

7. What role do you believe healthcare professionals have in supporting people with CF to take their treatments?
   a. Are there things that healthcare professionals can do to help or support people to take their treatments?
   b. Are there things that healthcare professionals do that is unhelpful or makes it more difficult for people to take their treatments?
   c. Do you think that people with CF feel that healthcare professionals support them to take their treatments?

8. When do you think is the best time to discuss treatment-taking with members of the CF Team (e.g., during clinic appointments, inpatient admissions, home visits, any other time)?
   a. Are there times you feel it is more or less appropriate for this to be discussed? If so, when and why?
   b. What would make this a positive/ negative experience?

9. When you are asked by a healthcare professional, "how many doses of X have you taken in the last X weeks?"), to what extent do you feel you can be honest about the amount of treatment taken?
   a. Are there things that help you to self-report the amount of treatment taken?
   b. Are there things that make it difficult for you to self-report the amount of treatment you've taken?

10. What are your views about treatment-taking approaches that, instead of asking you to say how many doses you have taken, measure the amount of treatment taken (e.g., chipped nebulisers that record the time and date that you take your treatments; chipped medication boxes that record the frequency with which you open a box to take a tablet)?
    a. Are there any advantages/ disadvantages of using this kind of approach?

Section C: This last set of questions focuses on how treatment-taking is written about (e.g., clinic letters).

11. What are your views about treatment-taking being mentioned/ discussed in things like clinic letters to people with CF?

    a. Can you think of a time when you received a clinic letter where treatment-taking was discussed/ approached in a positive way?

        i. What made it positive?

        ii. What did you like about it? Were there any words/ phrases in particular that you liked? If so, why?

    b. Can you think of a time when you received a clinic letter where treatment-taking was discussed/approached in a negative way?

        i. What made it negative?

        ii. What did you dislike? Were there any words/ phrases in particular that you disliked? If so, why?

12. For this question, I'm going to read a series of statements, and I'd like you to say what you think about each statement (e.g., what do you like/ dislike?).

    a. "You are taking X/X treatments on average per week."
    b. "You are managing to take X/X treatments a week."
    c. "You have taken X/X treatments over the past X weeks."
    d. "You have X% adherence/ compliance/ concordance to your treatments."
    e. "You are X% adherent/ compliant/ concordant to your treatments."
    f. "Your adherence/ compliance/ concordance is X%."

13. What, if any, is your preference out of the above phrases? Why is this?

14. Are there other phrases you would prefer to see used in clinic letters?

15. Do you have any other comments on the way in which the subject of treatment-taking is discussed in healthcare, or anything else you'd like to add that hasn't already been discussed?

That completes this interview. Thank you for taking the time to share your views and experiences with me.