ORIGINAL RESEARCH Objective Nebuliser Adherence Data as "Proof" of Adherence in the Management of Cystic

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Fibrosis: A Qualitative Interview Study

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Purpose: Low adherence to medication via nebulisers is linked to poor clinical outcomes for people with Cystic Fibrosis (PWCF). Advances in technology allow electronic monitoring of nebuliser usage and feedback of objective adherence data to PWCF and clinical teams caring for them. CFHealthHub is a new intervention that collects and displays objective adherence data in easy-to-read formats with the aim of improving nebuliser adherence and health. There is little understanding of how objective adherence data is perceived by PWCF and healthcare professionals (HCPs).

Patients and Methods: A qualitative study using semi-structured interviews with 22 PWCF and 31 HCPs who had used the CFHealthHub intervention.

Results: Objective adherence data was welcomed by the majority of PWCF in the sample, and HCP delivering the intervention, because the data allowed PWCF to reflect on patterns of adherence or non-adherence. Ease of use and characteristics of data display were important, particularly the use of a "traffic light" system to allow PWCF to easily see if they were meeting their adherence targets. For PWCF objective adherence data was used as "proof to self", offering reassurance to high adherers, and a wake-up call to those with lower levels of adherence. It could also provide 'proof to others' where PWCF had higher levels of adherence than HCP or family members believed. The data could sometimes change HCP perceptions of PWCF's identities as poor adherers. Where adherence was not high, data was used to facilitate honest discussions between PWCF and HCPs about how to increase adherence. HCPs perceived that it was important to use the data positively to motivate, rather than criticise, PWCF.

Conclusion: Objective nebuliser adherence data in CFHealthHub can offer proof of adherence to PWCF and HCPs. It is important to use it constructively to facilitate discussions on how to improve adherence.

Keywords: nebulised therapy, behaviour change, non-adherence, subjective reports, compliance, electronic capture

Introduction

Cystic fibrosis (CF) is a genetic, life-threatening disease which affects around 10,500 people in the United Kingdom (UK), and around 100,000 people worldwide.¹ Due to a defective gene in people with CF (PWCF), the lungs produce thicker than normal sputum, which affects internal organs, particularly the lungs and digestive systems, resulting in chronic respiratory infections, lung inflammation, and digestive problems. There is currently no cure for CF. However, due to advances in treatments, life expectancy is rising with a UK median survival age of 49.¹

PWCF have a substantial treatment burden, spending an estimated average of two hours daily on treatment related activities.² Daily treatments can include multiple nebuliser-inhaled medications, physiotherapy, exercise and digestive enzymes in tablet form. Non-adherence to inhaled medications is linked to poor health outcomes such as lower lung function, increased pulmonary exacerbations, increased hospital admissions and longer hospital stays.^{3–5} The benefits of pulmonary treatments are dependent on an individual's level of adherence, and as treatments are often time-consuming

and repetitive, poor or non-adherence to CF treatments is commonly reported; objective adherence recordings of inhaled therapies in adults have been shown to be around 31-36%.^{6–8}

For the purpose of this paper the use of the term "adherence" follows the World Health Organisation 2003 definition of the extent to which a person's behaviour (in terms of taking medication, following diets, or executing lifestyle change) coincides with medical or health advice.⁹ The World Health Organisation adopted the use of the term "adherence", rather than "non-compliance", in an effort to emphasise that a patient is free to decide whether or not to adhere to a doctor's recommendations, and to move away from apportioning blame to patients who do not follow their prescribed treatment regimen.¹⁰

Much of the recent literature on adherence in CF has focused on experiences of medication regimens,¹¹ barriers and facilitators to adherence,^{12,13} or ways of improving adherence.^{14–17} Traditionally, measurement of adherence has relied on subjective methods of assessment such as patient self-report, or clinician's subjective assessment, both of which have been found to be inaccurate.^{6,18} Nebulisers that electronically record nebulisation activity provide objective adherence data for nebuliser treatment that has actually been taken. There is little understanding of how objective data, when used as evidence of adherence, is perceived by PWCF and their clinical teams. We explored this for a new intervention, CFHealthHub, which measures and displays objective adherence data.

The research we report here was part of a programme to develop and evaluate a new intervention to increase adherence to nebuliser treatment in adults with Cystic Fibrosis (ACtiF). The CFHealthHub intervention comprised of a web platform and app which displayed graphs and tables of real-time objectively measured nebuliser adherence for each PWCF. Data was gathered from dose counting nebulisers that transmit the data to the CFHealthHub server infrastructure where it is stored securely and used for display in both a web-based interface and a mobile app. It displays individual real-time adherence data and personalised information in a "Toolkit" area. It also included modules of behaviour change techniques designed to increase motivation for adherence, to address capability and opportunity barriers, and to build habits for treatment taking. The CFHealthHub intervention was delivered in a randomised controlled trial that took place in nineteen CF centres around the UK. The intervention arm of the randomised controlled trial and HCPs could access the objective adherence data at any time. Further information about the intervention and the randomised controlled trial is available elsewhere.^{19,20}

Methods

Design

This paper reports findings from a qualitative interview study with PWCF, HCPs delivering the CFHealthHub intervention, and HCPs who were members of multidisciplinary teams (MDTs) offering routine care to PWCF. The qualitative study was part of a process evaluation undertaken alongside the CFHealthHub trial described previously. Individual semi-structured interviews were conducted using topic guides specific to PWCF, interventionists and members of MDTs. Examples of the topic guides used are in <u>Appendix 1</u>.

Ethics

The reported study complies with the Declaration of Helsinki. Ethical approval was obtained from London Brent REC: 17/LO/0035. All participants gave written informed consent to the recorded interview and agreed to the use of anonymised quotes in publications. Identifying data was removed from interview transcripts prior to analysis, and all participants were given identifiers to maintain anonymity.

Sample

For patient interviews the research team purposively selected seven centres in the trial to include a diverse range of large and small, rural and urban CF centres from around the UK. PWCF from these seven centres, who had used the intervention, were then approached for interview.

PWCF were purposively sampled for different levels of objectively measured adherence levels at the start of the trial, and different genders, ages and socioeconomic backgrounds. Eighty-six eligible PWCF were initially contacted via

email, phone or post. Thirty-two PWCF agreed to be interviewed and twenty-two interviews were conducted. Ten PWCF agreed to be interviewed but never provided data; three of these cancelled on the day of interview due to illness, one died before the interview date and six stopped responding when attempting to confirm an interview date.

Each CF centre in the trial had a research lead who was also part of the MDT offering care to all PWCF in that centre. Nine research leads/members of the MDT were approached for interview and five were interviewed; of the four not interviewed one left their NHS post during the recruitment process, one initially agreed to participate but then did not respond to attempts to arrange the interview, one never responded to contact, and one was off work until after the interview study finished.

Some CF centres had more than one interventionist due to sharing of job roles. At least one of the interventionists from every centre was interviewed.

Data Collection

Interviews were conducted by two researchers (EL and SD). They took place between May 2018 and June 2019. All PWCF were interviewed face-to-face in their own homes and interviews lasted between 18–83 (average 42) minutes. Twenty-five interventionists and one multidisciplinary team member interview were conducted in person at the participants' CF centres; the others were conducted by telephone. These HCP interviews lasted between 39–135 (average 88) minutes.

Analysis

All interviews were audiotaped and transcribed verbatim. Transcripts were entered into NVivoTM (QSR International) to facilitate analysis. The framework approach was used for analysis.²¹ This involved reading transcripts for familiarisation; developing a thematic framework based on the transcripts and the different components of the intervention; coding the transcripts to the framework; reading codes and considering who was making different points; and finally considering the relationships between themes and sub-themes. During this process, one of the qualitative researchers (EL) noted the recurrence of the word "proof" within transcripts and its relationship with the objective adherence data. She explored codes in NVivo related to this issue and identified themes and sub-themes related to this. The qualitative research team (EL, SD, AS, AOC) met regularly to discuss findings and interpretations, and also discussed them with the wider research team.

Results

Characteristics of Interviewees

The 22 PWCF interviewed had a mean baseline objective adherence category of very low < 25% (n=3), low 25.1%-50% (n=4), moderate 50.1–79% (n=8) and high >80% (n=7). They were mainly Caucasian (n=21), with one North East Asian. They were evenly distributed across gender and age: male (n=10) and female (n=12); 19–25 years (n=5), 26–30 years (n=3), 31–40 years (n=6), 41–49 years (n=4), 50 years and over (n=4). They came from a mix of social deprivation quintiles where 1 is the most deprived and 5 is the least deprived/most affluent: quintile 1: n=4, quintile 2: n=1, quintile 3: n=5, quintile 4: n=7, and quintile 5: n=5.

The 26 HCPs delivering the intervention who were interviewed were from a mix of clinical disciplines: CF nurse (n=6), CF physiotherapist (n=13), psychologist (n=3), non-CF physiotherapist (n=1), paediatric physiotherapist (n=1), CF pharmacist (n=1), and dietician (n=1). Interventionists were usually existing members of the CF team within that CF centre who were employed to deliver the intervention during the RCT. Five members of the multidisciplinary teams were interviewed, all of whom were doctors.

Overview of Themes

This analysis focused on experiences and views of using objective adherence data, specifically on perceptions of the data, the value of the data, and how the data was used in practice. The analysts identified the following themes: objective data was generally welcomed; ease of access to and presentation of data; data as "motivation and reward"; data as "proof to

self"; data as "proof to others"; changing identity of "poor adherers"; facilitating honest and challenging discussions; and using the data responsibly. These themes are described below with supporting quotes. Quotes from PWCF are labelled with a unique number for each interviewee, the CF centre number, and the level of adherence when joining the randomised controlled trial. Quotes from the interventionists, and multidisciplinary team members, have less identifying information to protect anonymity. The descriptor "HCP" is used regardless of whether they were an interventionist or multidisciplinary team member, and a unique number was allocated to each person.

Objective Adherence Data Generally Welcomed

PWCF receiving the intervention in the randomised controlled trial, and the HCPs, welcomed having objective adherence data available to them. The majority of PWCF valued viewing their data regardless of their baseline adherence level. They found it useful to see when they tended not to use their nebuliser or to spot that their adherence had dropped over the previous days and needed to be brought back up to prevent negative impacts on their health. HCPs described it as: "the strongest part of the intervention" (HCP 26 - interventionist) because it allowed them to focus discussion on what PWCF were actually doing with their medications. The intervention also provided information that historically had not been available.

"But actually seeing [the data] you know, it's the data. We like data!"

(PWCF 3, centre 1, moderate adherer)

"It's a piece of the jigsaw that previously was missing, so I think it's something that we have found is a really critical part of what we're doing now" (HCP 30 - multidisciplinary team (MDT))

Although the response to using objective adherence data was mainly positive from both PWCF and interventionists in this sample, some negative issues were raised. If objective data showed that adherence was lower than PWCF had expected, in some cases this could be disheartening and even distressing. A number of HCPs discussed PWCF who were moderate or high adherers, with no major concerns about their adherence from the HCP perspective, yet the PWCF expressed anxiety, fear, or in some cases anger at themselves, when the data showed any missed nebulisers. HCPs reflected on how the discussions about their data had provided insight into the anxiety and fear that was driving some PWCF to try and maintain their adherence rates, something that they had not anticipated.

"I asked him 'what's your reason for doing treatments'. He said 'because I'll die if I don't'. And I think he really, really believed that. And he's a very well lad, and he's almost hanging on to every day thinking 'I might die tomorrow' when he's got a lung function of 110% so actually you need to, you're going to have a lovely long life ahead of you. There's things you can go off and do but he's sort of, very frightened, very frightened about everything, and I think having the data was almost heightening his fear of, he went on to quite a lot of psychological support after seeing [CFHealthHub] actually, because he was so, it just heightened his sense of fear about his life. So that was very unexpected." (HCP 17 - interventionist)

Ease of Access to, and Presentation of, Data

PWCF valued both the different types of graphs and the range of time periods they could view. They were able to select the data display that best suited their preferences and needs. HCPs delivering the intervention noted the importance of having different kinds of data, presented in a variety of ways, to PWCF. They acknowledged that this was one of the reasons that the data could appeal to many different people.

"I think having the different views of the data is really important. There's different ways of looking at it, bar graph, line graph, weekly graphs, daily graphs all that kind of thing. I think the fact that there's so many different options means it fits, it works for everyone."

(HCP 27 - interventionist)

Ease of viewing adherence data was really important to PWCF, both in terms of accessing the data and being able to digest the information. Most PWCF interviewed tended to use the phone app to view their data because this was quick

and easy, even though the phone app did not offer the same range of data displays the website did. The use of a traffic light system of red (no adherence), amber (some adherence) and green (100% of target adherence) on graphs enabled PWCF to see at a glance if they were achieving their targets.

"so it brings up the bar graph and it's got your target line and if it's on the target it's green, if it's below, like mine is 3 treatments if I am on 75% which is 2 out of 3 it's orange, and if I have done one or less it's red. That's the one I use most, because I can open it, see green or see orange or see red and I do not even have to look at the numbers to see"

(PWCF 2, centre 2, moderate adherer)

Data as "Motivation and Reward"

The presentation features such as the traffic light system acted as a motivating factor because PWCF wanted to "see more green" on their graphs, or obtain a particular pattern. For example, one HCP captured this well when recalling a PWCF who wanted to see a more constant pattern which she described as a hedge rather than the inconsistent pattern that looked like trees. PWCF in the sample expressed a sense of satisfaction when they achieved green or the desired pattern, and sometimes described this as a reward. HCP described PWCF who were motivated by competing with themselves, aiming for a 100% adherence rate as a way of winning.

"He was struggling [with adherence]. But for him it was like a competition with the app where he could see whether he'd done it or not done it, and he was wanting to hit that 100% as much as he could."

(HCP 22 - interventionist)

Data as "Proof to Self"

Both PWCF and HCPs described how the objective data provided proof to PWCF of their adherence. This had different meanings depending on the baseline level of adherence when entering the randomised controlled trial. For some high adherers, the data provided reassurance that they were achieving the adherence level they believed they were achieving. They valued this reassurance, even when they were confident in their nebuliser routines.

"Yeah I'm not being smug but I know that [the graph] is going to be filled in right the way across" (PWCF3, Centre 3, high adherer)

For PWCF who had lower baseline adherence on entering the randomised controlled trial, the data could challenge their perceptions by demonstrating conclusively that they were taking less medication than they thought they were.

"because what they believe they were doing is so much higher than what they were actually doing, or they knew they weren't doing very much but to see it was the only thing that made them go 'Mmm that's actually how little I'm doing'"

(HCP 8 - interventionist)

The disjuncture between perception and reality could be an unpleasant surprise. Even if data aligned with perceptions, seeing low levels of adherence could cause them to reflect on their adherence behaviours. The data as "proof to self" could offer motivation to maintain their level for many higher adherers and motivation to increase adherence for some lower adherers.

Data as "Proof to Others"

Some PWCF perceived that their clinical team did not always believe their subjective reports of adherence and were frustrated when they were not believed, regardless of their baseline adherence levels. Having the objective data as "proof" now ensured they were believed.

"they didn't believe me that I did it all the time and they were quite impressed that I did it because obviously I was telling the truth."

(PWCF 2, Centre 4, high adherer)

"When I do have my clinic appointments, they know I'm not lying, that I am doing my treatments and not just saying I am! Because I am sure sometimes they do not believe you when you say 'yeah I'm doing all my [treatment]".

(PWCF 4, Centre 5, Moderate adherer)

HCPs were aware of a dynamic of lack of trust between some multidisciplinary team members and some PWCF concerning adherence to treatment. The HCPs discussed how they were able to use the objective data as proof of patient adherence when sharing it widely with members of the multidisciplinary team. The HCPs interviewed described how sharing of data could improve relationships between multidisciplinary teams and some PWCF by allowing clinical teams to acknowledge the work that PWCF were putting into adherence. Some PWCF also used the data to show family members, who were concerned about their adherence, that they were taking their medication. That is, the data could act as "proof to family" as well as "proof to clinician".

Changing the Identity of "Poor Adherers"

There was a perception amongst some PWCF that due to previous non-adherent behaviour, often as a child or teenager, that they were considered to be low adherers and it was difficult to change this identity, even amongst family, if they were now adhering to treatment.

Although not the best reason it is you know really nice being able to just show my mum and say 'look it's been done for the week' she's like 'oh ok' and that's her off my back you know ... (PWCF 4, Centre 5, Moderate adherer)

HCPs delivering the intervention were also aware of this labelling process. When reflecting on their own practice many of the HCPs in the sample acknowledged that despite their best intentions, they had preconceptions of who they thought did, or did not, adhere to nebuliser treatment.

"it's difficult when you have got a chronic group of patients. People within the team sometimes develop a real preconception of them, based on something they did five years ago, and sometimes it's hard when you say 'well that's not really true anymore, they've changed". (HCP 18 - interventionist)

The proof offered by the objective adherence data was particularly helpful if PWCF became ill, or if their lung function deteriorated. The data gave PWCF confidence that they could not be blamed for not taking their medication, as might have happened previously. Some PWCF in the sample described how having this proof resulted in the multi-disciplinary team looking for reasons other than non-adherence to medication for their decline in health.

"I think it helps them trouble shoot what's going on because they are like 'you've done everything, we can tell you've done everything. This makes no sense and it must be something else.""

(PWCF 3, Centre 1, moderate adherer)

Facilitating Honest and Challenging Discussions

HCPs discussed how it could be "incredibly difficult" (HCP 32 (MDT)) to initiate conversations about adherence with PWCF, and how having the objective data now facilitated this dialogue. They suggested that having the data gave them a more accurate picture, and therefore could change the tone of any discussions around adherent behaviour from how it might have been if they were less informed. The objective data could show that PWCF had low or moderate adherence levels that they needed to increase, and equally could demonstrate which PWCF had been adhering. Both PWCF and HCPs described how having the data as "proof to self" and "proof to others" could facilitate more open and honest discussions around their adherence behaviour.

"So again it might just prompt a conversation with them about their treatments and trying to make them speak honestly and openly about whether they're doing it or not."

(HCP 4 - interventionist)

Some PWCF acknowledged that previously they had not always been honest about their adherence because there was no proof to challenge their subjective reports.

"There were times when I was honest and there were times when I weren't. This very much played into my own mental state at the timeBut actually coming out of that and being more honest and actually realising that it's not such a good thing so I actually found that I have been more honest now".

(PWCF 2, Centre 6, very low adherer)

Having objective adherence data meant that PWCF and HCPs could also explore sometimes complex reasons for nonadherence. They acknowledged that the need to be honest led to more meaningful conversations with HCPs around why they may not be adhering. This in turn helped with finding ways of increasing adherence.

Using the Data Responsibly

HCPs described the importance of using the objective data positively, and suggested strategies to prevent it from being used negatively. In the initial training of the interventionists the importance of maintaining a non-judgemental approach to conversation was emphasised. The HCPs interviewed were clear that the adherence rates should not be used to chastise or criticise PWCF but rather as a route to open discussion about how to improve adherence. HCPs also highlighted that for some PWCF the adherence targets could add pressure, rather than acting as a motivator, so it was necessary to keep adherence discussions positive rather than focusing solely on non-adherence.

"my only slight worry about this is some of them who sort of really beat themselves up about missing things or sort of having it really in their faces: 'your compliance is down' or 'your adherence is down'. I think some of them find it quite tricky and they have then sort of had to justify, it felt like they had to justify it, but I would say actually you are doing pretty well, I'm not sure I would get too worried about this or that, if you look at your overall adherence" (HCP 28 - MDT)

In the small number of cases in the sample where PWCF experienced demoralisation or fear on seeing their objective data, HCPs described employing strategies that had been covered in their initial training to address this. For example, when faced with demoralised PWCF they tried to frame discussions positively, such as concentrating on what treatments had been done, or showing a longer period on the graphs so that more green – denoting adherence - was visible. After discussion with the multidisciplinary team some HCPs reduced adherence targets temporarily as a way of showing PWCF that they could achieve a set target, with the ultimate aim of increasing the target over time. It was felt that it was better to try and help PWCF to do "some" treatment, rather than set too high a target and have no medications taken. PWCF also described how having a lower initial target helped their sense of achievement, and acted as a spur to increase adherence.

Discussion

Summary of Findings

Objective adherence data was welcomed by the majority of both PWCF and healthcare professionals delivering the intervention in our sample. The data allowed PWCF to reflect on patterns of adherence or non-adherence. Ease of use and characteristics of data display were important, particularly the use of a "traffic light" system to allow PWCF to see at a glance if they were meeting their adherence targets. These graphs could offer motivation and reward to PWCF as they aimed to turn graphs green – a sign of high adherence. For PWCF objective adherence data was used as proof to self, offering reassurance to high adherers and a wake-up call to those with lower levels of adherence than they had been believed. It could also be proof to others when PWCF were adhering more than clinical teams or family members believed. The data could sometimes change HCPs perceptions of PWCF's historic identities as poor adherers and facilitate discussions about alternative reasons for ongoing health problems. Where adherence was not high, data could be used to facilitate honest discussions between PWCF and HCPs about how to increase adherence. HCPs wanted the data to be used positively to motivate rather than criticise PWCF.

Comparison with Other Literature

The benefits of objective adherence data in CF has been shown elsewhere. A 2015 study to identify best practice in CF adherence assessment and counselling in the US advocated that objective data should be meaningful and easily incorporated into clinical appointments.¹³ This study also proposed further research to identify the best way to display objective data for both PWCF and clinicians in order to support understanding and impact on non-adherent behaviour. This resonates with our work where both PWCF and HCPs valued the range of ways of presenting the data so that it was quick and easy to personalise and use.

A systematic review of the experiences of adults with CF in adhering to medication regimens suggested that PWCF wanted to be trusted that they were doing their best, even if this did not meet with the expectations of others.¹¹ In a study of young adults with CF, it was suggested that PWCF wanted to be trusted with self-management.²² However, it has been argued that trust is viewed differently by HCPs and PWCF, with PWCF reporting that they trusted HCPs and were open about their non-adherence, but found that HCPs did not trust their self-reported adherence.²³ Our work builds on this by showing that relying on subjective accounts of adherence requires trust between HCPs and PWCF which can be problematic. The objective data replaces the need for reliance on PWCF's subjective accounts. It is important that trust between HCPs and PWCF is maintained as it is integral to a constructive patient-clinician relationship.

Our study found that how objective data is used is important, with the HCPs we interviewed recommending that clinical team members should try to instigate a discussion about low adherence rather than castigate PWCF. This is supported by other literature. Riekert¹³ and colleagues argued that whilst there were clear benefits to using objective adherence data in an effort to improve treatment adherence, care should be taken to ensure that the data is not used in a pUnitive manner; other studies echo this with the suggestion that reprimanding PWCF is not helpful, even though knowing a patient is non-adherent can be frustrating for clinicians.^{17,24} A recent review examining the use of digital technology in home monitoring, adherence and self-management in CF²⁵ described the use of electronic monitoring of adherence via data logging nebulisers as "an area of promise". However, the authors acknowledged that monitoring alone may not address barriers to adherence and may need to be combined with a more individualised intervention aimed at improving adherence. The intervention explored in our study (CFHealthHub) combined both an individualised intervention aimed at improving adherence data. Our exploration of mechanisms of action of the intervention revealed that multiple mechanisms were at play, including the relationships built between the HCPs delivering the intervention and PWCF receiving the intervention.²⁶

Strengths and Limitations

The sample was drawn from a wide geographical area across the UK, and included a diverse sample of PWCF including those with low and very low adherence levels. A key issue to bear in mind is that the interviewees were PWCF who had agreed to participate in a randomised controlled trial of intervening to improve adherence to nebuliser treatment, and HCPs who had sought employment to deliver the intervention. They may have been more positive about monitoring adherence using objective data than the wider population of PWCF and HCPs.

This paper has utilised the Consolidated Criteria for Reporting Qualitative Studies (COREQ)²⁷ checklist in order to enhance the reporting of the study, demonstrating the comprehensiveness and quality of methods and approach. The completed checklist can been seen in <u>Appendix 2</u>.

Implications for Practice

Historically, objective adherence data has not been widely available in CF centres in the UK²⁸ or US.¹³ This study has highlighted that objective adherence data may be welcomed by some PWCF and HCPs. However, it is not simply about having this data available but about how it is presented and used. The CFHealthHub intervention offered an accessible and acceptable way of displaying objective data on adherence to nebulisers in PWCF. The data could be used as a way of offering motivation and reward to PWCF to encourage them to improve or maintain adherence, and facilitate honest discussions about adherence. The intervention, now called CFDigicare, is currently being implemented in more than 60% of adult CF centres in England (ISRCTN14464661), with almost 2000 patients now using it (https://www.cfhealthhub.com/).

Conclusion

Objective nebuliser adherence data can offer proof of adherence to PWCF and HCPs. It is important to use it constructively to facilitate discussions on how to improve adherence.

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Disclosure

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