Attitudes and adherence in adults with cystic fibrosis

The multisystem nature of cystic fibrosis makes treatment regimens often quite complex, time-consuming and intrusive for the sufferer. Amanda Plummer and colleagues evaluated the perceived levels of adherence and views about self-management of adults with cystic fibrosis attending the Cystic Fibrosis Unit at Sheffield Teaching Hospitals NHS Trust, and the findings are presented here. In a second study the authors investigated factors that affect adherence and these data are presented in an accompanying article.

Abstract

Objectives. Cystic fibrosis (CF) is the most common life-threatening, recessively inherited disease in the UK. The treatment of CF is complex because of the multi-system involvement. It is often intrusive and may take a large portion of the day leading to difficulties in a predominantly young, mobile population. This study investigated the perceived level of adherence and views on self-management of adults with CF, and the views of the multidisciplinary team.

Design. The study consisted of semi-structured interviews.

Participants. Twenty-five patients and 10 members of the multidisciplinary team were included.

Main outcome measures. Perceived level of adherence and views on self-management.

Results. Patients took nine (3–17) different medicines each day with a frequency of four (2–6) times per day excluding the additional use of pancreatic enzyme supplementation with meals, snacks and feeds. The patients estimated their median percentage adherence to be 95% (10–100%). This value was contradicted by the verbal descriptions of their medicine routine by four participants in specific circumstances. The members of the multidisciplinary team predicted different adherence rates than the participants' estimations, with a median predicted adherence level of 50% (range 25–60%). A lack of awareness of the concept of self-management was described by the participants despite many having personal experience of this.

Conclusions. Patients should be involved in discussions about their treatment in the context of their everyday life including detailed discussions to evaluate more objectively the actual adherence level and achievable objectives to gain this.

Introduction

Cystic fibrosis (CF) is the most common, life-threatening, recessively inherited disease in the UK,[1] caused by a single gene defect on the long arm of chromosome seven. The gene encodes the cystic fibrosis trans-membrane conductance regulator (CFTR), a 1480 amino acid protein. CF is inherited in a Mendelian recessive manner. In the UK it is estimated that one in twenty-five of the population carries the CF gene and approximately one infant in every two thousand five hundred has CF.[2]

Because CFTR is present in the lungs, saliva glands, pancreas, liver, bowel, brain, sweat ducts and reproductive tract, CF is a multi-system disease. Defective CFTR results in abnormal ion transport across the cell membrane resulting in thickened secretions, mucus plugging, obstruction, scarring of tissue and secondary infection.

The treatment of CF is complex. The goals of therapy are to improve nutritional status, to prevent or slow the decline in pulmonary function and to aim for as 'normal a life' as possible with the best quality. The doses of medicines used are generally greater than those used by the non-CF population because of poorer oral bioavailability, enhanced clearance and a larger volume of distribution. The treatment may involve taking tablets, inhaled therapy (nebulisers and inhalers) and intravenous antibiotics, in addition to physiotherapy, oxygen and supplemental feeding. The regimen is often intrusive and treatment may take a large portion of the day leading to difficulties in adherence within a predominantly young, mobile population.

Adherence has been defined as 'the extent to which the patients fulfil the intention of the prescriber in taking medication.'[3] One of the difficulties when defining adherence in CF is the lack of evidence for some of the treatments and the unknown optimum level of adherence for maximum clinical benefit for an individual treatment and dosage schedule. Research suggests that adherence with individual treatments varies.[4,5,6,7,8] However, most of these data are derived from self-report methodology, which is known to over-estimate true adherence. Table 1 details some of the evidence available.

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Lask, in 2001, identified difficulties when considering adherence research,[19] all of which can be applied to the studies in CF as follows:

- There is no universally agreed definition of adherence
- There is poor methodology in empirical research
- There is lack of valid and reliable methods of measurement
- There is a tendency to polarise between adherence and non-adherence
- There may be different adherence rates to each component of treatment.

In 2001 the UK government published the 'expert patient' strategy[20] to involve patients with chronic illness in managing their own health. It was recognised that patient knowledge and experience was an 'untapped resource' and that patients often knew their disease better than health care professionals. The use of self-management was strongly promoted, emphasising the support for concordance and shared decision-making. However, efforts to improve self-management skills through education programmes, as described by Bartholomew and colleagues[20,21] and Parcel and co-workers,[22] have been found to be ineffective. If clinical outcomes are to be improved then interventions will need to include more than just the improvement of knowledge.

In the present study we investigated the perceived level of adherence and views on self-management of adults with CF. We also explored similar views of the multidisciplinary team. Many of the difficulties experienced by patients with CF could also be applied to other young populations with complex therapies.

Table 1. Reported adherence levels in people with cystic fibrosis

Various reports in the literature show:

- Greater than 50% of patients claimed to be taking more than 80% of their prescribed treatment[4]
- Total adherence has been estimated in different studies to be 75%[9] and 84.1%[10]
- Adherence to medication was found to be the highest, diet intermediate and physiotherapy and exercise the least[11]
- Adherence with physiotherapy was reported at 50% and to exercise, pancreatic enzyme supplementation and diabetic care it was higher at 80%[12]
- Adherence was higher with diabetic medication, followed by pancreatic enzyme supplementation, nebulisers, dietary supplements and then physiotherapy[4]
- 75% reported total adherence to vitamins and 45% total adherence to pancreatic enzyme supplementation[13]
- 85% were adherent to pancreatic enzyme supplementation[5]
- Adherence to pancreatic enzyme supplementation was reported to be 80% with meals and 59% with snacks[14]
- 47% of adult patients were considered adherent to vitamins[5,15]
- 42% of adults reported full adherence to ADEK vitamin supplementation but 56% were not adherent[16]
- 73% adherence to ADEK vitamin supplementation was reported in children less than 12 years of age and and 56% in children greater than 12 years[17]
- Adherence with vitamins and antibiotics was high, greater than 90% had complete adherence and less than 5% had poor adherence[6]
- 80% of patients complied completely with prescribed antibiotics[18]
- 67% adherence to Dornase alpha was reported in children less than 12 years of age and 72% in children greater than 12 years[17]
- Self-reported adherence in 16 adults was shown to vary over time with a general trend to increase over time for an individual[11]

Methods

This qualitative study consisted of 25 semi-structured interviews with patients attending the Adult Cystic Fibrosis Unit at Sheffield Teaching Hospitals NHS Trust and 10 members of the multidisciplinary team between May 2003 and May 2004.

The patients attending the unit were categorised according to their 'band' and gender. Patients are normally placed into five bands each year for funding purposes.[21] For local purposes a sixth band is used for patients who have received a transplant. The banding system revolves largely around the number of courses of intravenous antibiotics the patient receives each year, with a band one patient receiving no intravenous antibiotics and a band five patient receiving the most intensive input.

A non-parametric quota sample was used to ensure patient participants were recruited from all bands, and that there was an equal representation by gender. Patients who had other non CF-related health complications were excluded from the study. For example, anyone following a therapy regimen through prison services or mental health services were excluded. The study was also limited to patients who had been attending the Adult Cystic Fibrosis Unit for a minimum of one year to exclude any issues relating to either the care received at their previous centre or the transition process. Ethics approval for the study was gained from the North Sheffield NHS Local Research Ethics Committee.

Participants were asked to identify their adherence to their medicines using a Likert Scale as 'if taken all the time, sometimes or rarely'. This classification is similar to other published research.[6] Qualitative data were analysed according to the thematic framework method described by Ritchie and Spencer.[23] A proportion of the coding procedures were independently analysed. Quantitative data collected during the interviews were analysed using Statistical Package

for Social Scientists (SPSS) v.11. Statistical data were limited and therefore treated as non-parametric; averages are expressed using the median.

Results

1. Adherence

Patient participants were asked to estimate their 'average adherence' and 'individual adherence for all their medicines' using the Likert Scale. However, the majority of participants offered a percentage figure.

The results showed that participants took nine (3–17) different medicines each day with a frequency of four (2–6) times per day excluding the additional use of pancreatic enzyme supplementation with meals, snacks and feeds. Only four patients were taking four medicines or less, with nine taking greater than 10 medicines per day. Overall, the participants estimated their percentage adherence to be 95% (10–100%). Seven participants thought that they took an average of less than 80% of their medicines, whereas seven other participants reported that they took 100% of all their medicines. This latter value was contradicted during the interview by four participants. For example, one participant estimated 95% adherence to flucloxacillin but then described not taking it at lunch, which suggested adherence could only be a maximum of 75%. Another expressed a fear of being perceived as not being 100% adherent, perhaps because of the knowledge that adherence is a factor looked at when considering transplantation.

Adherence rates for individual medicines are listed in Table 2. The median adherence level to pancreatic enzyme supplementation was reported to be 95%, but many participants described not taking this with snacks or not taking it when they were out of the house. Only one participant appeared to have an accurate picture, which supported their described routine.

Members of the multidisciplinary team predicted different adherence rates to those the patients gave. Professionals were asked their views on the average adherence level and the range of adherence of the participants attending the clinic. Adherence was thought to be 50% (range 25% to 60%), clearly less than the participant estimations but with some differences between the team members. For example, the lowest prediction was from the dietitian about dietary products. All professionals interviewed thought that the range of adherence would be from zero to most of their treatment (80–95%).

> It is particularly challenging to reduce the length of a course of medicines in chronic illness [such as CF] and each medicine should be regularly reviewed to check for necessity and adverse effects.

All team members identified that adherence would be individual to the nature of the treatment and half reported that it would also be individual to the patient. For example, two professionals suggested 'problems experienced during adolescence' might influence adherence, and one team member proposed that changes in adherence might occur over time. According to the professionals a number of specific therapies were raised as being likely to produce low adherence; the majority of team members (7) identified vitamins with low adherence. Other suggestions made by individuals included preventive therapies and nebulisers (3), physiotherapy (2) and prophylactic nebulisers, inhalers, oxygen, dietary supplements or PEG feeds and insulin.

Table 2. Adherence to individual treatments by percentage

		Estimated percentage of medicines taken						
Drug	Number of participants expressing a % figure	median	mode	minimum	maximum	25 percentile	75 percentile	one or two values only
Pancreatic enzyme supplements	15	95	100	50	100	80	100	—
Vitamins A&D	13	93	100	25	100	70	100	—
Vitamin E	14	77.5	100	25	100	65	100	—
Long-acting beta-2 inhibitor	7	95	95	70	100	95	100	—
Steroid inhaler	9	95	95	0	100	45	97.50	—
Colistin nebulisers	6	75	100	0	100	15	100	—
TOBI nebulisers	1	—	—	—	—	—	—	100
Dornase alpha nebulisers	2	—	—	—	—	—	—	60, 95
Flucloxacillin	9	93	95	60	100	75	95	—
Ciprofloxacin	3	100	100	95	100	95	100	—
Azithromycin	5	100	100	75	100	87.5	100	—
Proton pump inhibitor	9	100	100	50	100	75	100	—
Insulin	5	100	100	70	100	70	100	—
Prednisolone	4	97.5	100	80	100	83.75	100	—
Tacrolimus	2	—	—	—	—	—	—	85, 100
Risedronate	3	100	100	100	100	100	100	—

2. Self-management

Participants were invited to describe their understanding of self-management and illustrate this with relevant examples. Sixteen participants reported that they understood self-management or gave examples of this, seven reported to have no experience of it and two were unsure. Only 11 participants reported they felt confident with self-management. Five stated that they would want to involve the CF team.

Participants were asked to describe systems they use to keep track of their medicines. Nineteen responded with a variety of systems to aid medicines management the majority of whom stated they used 'routine/habit' and 'memory'. Other systems included 'diaries or a written record', 'compliance aids', 'medicines kept in same place', 'medicines taken together' and by 'following stock of a particular medicine'.

A number of areas of concerns about self-management were raised by patient participants. These included:

- The need for self-management to be checked and supported by staff
- The need to be reliable if the activity is to be successful
- More information or guidelines about self-management
- Knowledge of side-effects and antibiotic sensitivities
- Self-management could not be undertaken with some medicines
- Concern was expressed of the likelihood that they would just take less of the medicines prescribed.

In comparison, health care practitioners were asked to provide a definition of self-management. The majority of practitioners (9) identified the patient taking 'control' or having 'responsibility' for their own medication as a major theme. One team member had not heard of self-management in such terms. A number of factors were reported and are listed below.

- Staff identified that patients have variable capabilities (5), are dependent on the CF team (2) or could be scared (1)
- Four professionals thought that education and adequate knowledge would be required for successful self-management
- Three expressed the potential for self-management to be structured with a treatment plan to adjust doses, start treatment courses or seek help
- Two thought that self-management happens anyway in the adult field because patients are empowered or take what they feel like
- Other individual comments included:
  a. The patient would be able to vary treatment if they wished to do so
  b. The patient would be able to recognise symptoms and respond

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  c. The team may feel out of control but that this may be good
  d. There would need to be good patient selection and a check on what the patients are doing.

Four practitioners reported evidence of self-management in the form of patient adjustments to their pancreatic enzyme supplementation, insulin and the use of oral antibiotics.

Discussion

This study was based on a self-report methodology, which is known to overestimate adherence estimations. The high level of adherence described by the patient participants concurred with those found in similar studies in CF[4,9,10] despite the large number of treatments that were taken throughout the day. Eighteen participants described their adherence as 80% or greater compared to 50% as described by Conway.[4] Perhaps this was because the information was being given directly rather than through a questionnaire. Because only seven participants estimated their adherence to be less than 80%, and the seven lower values were in four different bands and four age groups, it was not possible to correlate low adherence with age or band.

The conflict between the estimated adherence level and the description of the medicine routine suggests that a more detailed discussion is required with those patients whose clinical response does not match their suggested adherence level. Doctors and medical students have also been shown to overestimate adherence.[22,24–26] However, this contrasts with the results from this study.

The literature for adherence rates for individual treatments in CF varies,[4,5,13,15–17] but a general principal of higher adherence to pancreatic enzyme supplementation and lower adherence to vitamins and nebulisers was seen in this study. Treatments that were perceived as being effective or that reduced symptoms appeared more likely to be taken.

We recognise a limitation of our study, which was based on a self-report methodology, was subject to the possibility of overestimations of adherence. Some of the data could have been more consistently attained by completion of a questionnaire by the participants and use of percentage figures rather than Likert scales.

Conflicting views were also expressed on the use of self-management in this patient participant group despite a high number being required to apply the principles to their everyday treatment. A variety of systems were described by participants to manage their medicines on a daily basis. A discussion of this process with the pharmacist may help to identify appropriate interventions that could improve adherence.

Future studies

Further research should address the changes in individual adherence rates over time and the role that health care professionals have within this. More objective measures of assessing adherence within clinical practice are required to overcome the conflict between patients' descriptions and their estimates of adherence. However, these should only be used in a positive way to facilitate discussions of individual adherence and views of medicines.

Conclusions

The adherence rates described by the patient participants were generally very high and often conflicted both with their described routines and the expected values from the multi-disciplinary team. The purpose of discussions of adherence with patients is to develop a mutual understanding and promote discussion of factors relating to medicine use. The use of more detailed descriptions will help to guide these discussions and allow the patients to explore their true level of adherence and factors that influence this.

The varied understanding of self-management and the reliance on the CF team conflicts with what the patients do in reality. A more formal education programme, taking into account infection control limitations, and written guidance might help to support patients with self-management when appropriate and when they feel confident.

Tailoring the regimen is difficult for most people with CF because of the complexity of the treatment required. It is particularly challenging to reduce the length of a course of medicines in chronic illness and each medicine should be regularly reviewed to check for necessity and adverse effects.

Recommendations for practice

1. Individual treatments should be discussed with each patient in the context of their everyday life.
2. The ability and desire of a patient to manage to adhere to enough medication to ensure therapeutic efficacy should be a key step in the prescribing process.
3. The adherence rate described by the patient should be investigated in the context of their daily routine.
4. Self-management as a concept should be promoted within this patient population for those who wish to be involved.
5. The individual's system to manage his/her medicines should be discussed and the opportunity to positively influence adherence encouraged or treatments reviewed as appropriate.
6. Achievable targets for adherence should be discussed with the patients and linked with their daily routines and priorities.

Our second original research article describes the factors that we found to affect adherence in these patients. ✣

Declaration of competing interests

The authors declare that they have no competing interests.

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