Adherence to Treatment in Adolescents with Cystic Fibrosis: The Role of Illness Perceptions and Treatment Beliefs

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Objectives This study was conducted to explore the relationships between illness perceptions, emotional representations, treatment beliefs and reported adherence in adolescents with cystic fibrosis (CF).

Methods Thirty-eight adolescents completed questionnaires assessing their perceptions of CF, beliefs about prescribed treatments and reported adherence to chest physiotherapy, enzyme supplements, and antibiotics.

Results Reported non-adherence to chest physiotherapy was associated with the way in which patients judged their personal need for treatment relative to their concerns about potential adverse effects. Patients reported strong doubts about the necessity of chest physiotherapy. Reported non-adherence to antibiotics was related to doubts about the necessity of antibiotics, believing that CF is not amenable to treatment control. Despite these beliefs about treatment, participants perceived CF as a chronic condition.

Conclusions The findings provide preliminary support for the self-regulatory model, using the necessity-concerns framework to operationalize treatment beliefs, in explaining adherence to treatment in adolescents with CF.

Key words cystic fibrosis; adherence; illness representations.

Cystic fibrosis (CF) is the most common, fatal, hereditary disease of childhood (Sharp, McNeil, Wales, Cooper, & Dawson, 1994). It affects multiple organ systems, primarily the lungs and pancreas, resulting in premature death, usually from progressive respiratory failure (Miller, Jelalian, & Stark, 1999). Recent advances in the medical management of the disease have dramatically improved survival rates (Elborn, 1998). However, there is currently no cure for CF, and, in order to maintain optimal health, patients are required to follow a lifelong, multi-component treatment regimen. The three main components are as follows: chest physiotherapy (CPT), at least twice daily; pancreatic enzyme supplements (ES) with all meals and snacks; and routine courses of oral or nebulized antibiotics. Treatments differ both in terms of their impact on symptoms and the time and effort they require (Miller et al., 1999). CPT and nebulized antibiotics are typically prescribed prophylactically, and therefore provide very little in the way of symptomatic benefit. Moreover, CPT is both the most time-consuming and tiring aspect of treatment, and may lead to unpleasant symptoms such as coughing. ES taken with food prevent stomach pains, bloating, and bulky stools (Koocher, Gudas, & McGrath, 1992).

Research has shown that adherence to treatment is associated with improved pulmonary status and increased longevity (Elborn & Bell, 1996; Patterson, Goetz, Budd, & Warwick, 1993; Thomas, Cook, & Brooks, 1995). However, non-adherence to treatment, especially CPT, is a significant problem, particularly during adolescence when the responsibility for performing treatment transfers to the patient (Abbott, Dodd, Bilton, & Webb, 1994; Conway, Pond, Hamnett, & Watson, 1996; Hamlett, Murphy, Hayes, & Doershuk, 1996).

Few studies have investigated the factors associated with adherence in adolescents. Research in adult and
pediatric populations has identified a number of possible influences, including knowledge about the disease (Levers et al., 1999), locus of control beliefs (Myers & Myers, 1999), self-efficacy (Czajkowski & Koocher, 1987), perceived illness severity (Gudas, Koocher, & Wypij, 1991), optimism (Czajkowski & Koocher, 1987; Gudas, Koocher, & Wypij, 1991), coping style (Abbott, Dodd, Gee, & Webb, 2001), family factors (Geiss, Hobbs, Hammersley-Maercklein, Kramer, & Henley, 1992), support (Abbott et al., 1994), and characteristics of the treatment regimen (Abbott et al., 1994; Conway et al., 1996). It is difficult to make cross-study comparisons and draw firm conclusions about the relative importance of different factors, however, as research has largely been conducted outside a theoretical framework and without consistent means of assessing key variables.

Leventhal’s Self-Regulatory Model (SRM; Leventhal, 1993; Leventhal, Diefenbach, & Leventhal, 1992; Leventhal, Zimmerman, & Gutmann, 1984) postulates that an individual’s personal model of an illness \(^1\) is a proximal determinant of both their emotional and behavioral response to a health threat. This approach differs in at least three ways from other models that concern the role of patient beliefs and attitudes in determining health behaviors. First, personal models are an extension of schema theory from cognitive social psychology. Thus, unlike other social cognition models, personal models are grounded in a general theory of cognition that accounts for the merging of incoming information with past experience. Second, unlike some theories, which have been developed from theoretical reasoning with little empirical induction, the core concepts in Leventhal’s model were derived from qualitative, anthropological, and psychological research. That is, the core constructs assessed were derived from patients’ accounts of illness and reflect those concepts that patients themselves believe to be central to their experience of illness and its management. Third, personal models include the representation of emotional responses to disease and treatment, which is lacking in the other health belief models. Leventhal argues that individuals have two interacting challenges when encountering an illness: the management of the objective aspects of the threat to health and the management of the emotional responses that occur as a result of the diagnosis, treatment, and management of the condition. These two aspects of illness management are seen as separate, but interacting pathways, and as a result it is argued that Leventhal’s model provides a comprehensive system for conceptualizing the individual’s response to living with an illness or condition.

Research using these underlying principles has identified five components to personal models of an illness: illness identity and associated symptoms, that is the individual’s perception of what is wrong; its cause, what factors or factors have interacted to lead the individual to develop the condition; the consequences of the illness, how the condition will affect their physical, mental, and social functioning; the individual’s understanding of the illness duration, that is whether it is a condition they will have the rest of their life, will be cured with treatment, or may come and go in cycles; and treatment beliefs, that is beliefs about the efficacy, necessity of, and possible negative aspects of treatment (Lau, Bernard, & Hartman, 1989; Leventhal, Nerenz, & Steele, 1984; Meyer, Leventhal, & Gutmann, 1985).

Personal models of illness perceptions have been found to relate to treatment adherence in patients with hypertension (Meyer et al., 1985), adolescents (11–18 years) (Skinner, John, & Hampson, 2000; Skinner et al., 2003) and young adults with diabetes (Griva, Myers, & Newman, 2000; Skinner, Hampson, & Fife-Schaw, 2002), and asthma (Horne & Weinman, 2002) and to predict attendance at cardiac rehabilitation classes (Cooper, Lloyd, Weinman, & Jackson, 1999; Petrie, Weinman, Sharpe, & Buckley, 1996). The majority of studies examining treatment beliefs have focused on assessment of treatment efficacy. However, Horne and colleagues (e.g., Horne, Weinman, & Hankins, 1999) have highlighted the benefit of including measures of medication-related beliefs, (specifically with perceptions of the necessity of the medication and concerns about the medication’s potential, adverse effects) as part of the treatment beliefs dimension. In a meta-analysis of the work using this model, Hager and Orbell (2003) report that the more an individual believes that their treatment is effective, the more problem focused coping reported, the more support they seek and the better their disease state. By contrast, perceptions of more severe illness consequences and a stronger illness identity were associated with more negative reports of functioning but not objective measures of health status.

As seen from the above literature and meta-analysis, there is a significant body of work exploring the utility of the SRM in a number of chronic illnesses, however, to our

\(^1\)The term ‘personal model of illness’ is used throughout this article, in line with previous papers in this journal (Skinner, John & Hampson 2000; Edgar & Skinner 2003). However, the wider literature also uses the terms ‘illness representation’, ‘illness perception’, and ‘explanatory model’ to describe these core dimensions of a person’s illness beliefs.
knowledge, it has not been tested in relation to CF. Therefore, the aim of this study was to explore whether the personal model of illness, held by adolescents with CF, is associated with their use of the three principal forms of treatment: CPT, ES, and antibiotics. Based on studies in other illness populations, it was anticipated that use of CPT, ES, and antibiotics would be associated with perceptions of the efficacy, necessity, and concerns about the potential adverse effects of treatment.

Therefore, we hypothesized that greater belief in the efficacy of treatment, in personal control over their CF, stronger beliefs in the necessity for treatment and fewer concerns about treatment would be predictive of better self-care. As the predicted relationship between time-line perceptions and adherence would vary depending on the condition, we hypothesized that affirming a chronic model of CF would predict greater adherence based on our supposition that affirming a chronic time-line is evidence of a more adaptive attitude towards CF. We also anticipated that identity, consequences, and emotion representations would not be associated with adherence, but would be correlated with each other, in line with Hageer and Orbell’s (2003) meta-analysis. Finally, in line with other research with adolescents with diabetes (Skinner et al., 2000), we anticipated that personal models of CF would also mediate the association between demographic variables and adherence. This study had ethics committee approval from the Local NHS Research Ethics Committee.

Method
Design
A cross-sectional design was used to investigate the relationships between personal models of CF and reported adherence in adolescents with CF.

Participants
Participants aged between 11 and 17 years were recruited from the pediatric outpatient departments of five hospitals in the South of England. Potential participants were excluded if they were receiving in-patient care during the period of data collection as this has been shown to confound adherence rates (Abbott & Gee, 1998). Fifty-two adolescents were approached to participate in the study. One was excluded and 12 declined. Of the remaining 39, one died before data had been collected; 38 participants (73%) completed the study.

The sample consisted of 20 females (age M 13.1 years, SD 2.3) and 18 males (age M 14.1 years, SD 1.7). This gender split is comparable to the CF population as a whole (Cloutier, 1997). There were no statistical differences between the participants (N = 38) and those who refused consent or were unable to take part (N = 14) in terms of gender, $\chi^2(1) = 0.39$, $p > 0.05$, or age, $t(50) = 0.36$, $p > 0.05$.

Materials
The Revised Illness Perception Questionnaire (IPQ-R); Moss-Morris et al. (2002), comprises several subscales assessing Illness identity (participants’ ratings of the presence or absence of 14 general symptoms and 5 CF-related symptoms, which were added specifically for the purposes of this study); Time-line (10 items assessing participants’ views about the chronic and/or episodic nature of CF); Consequences (six items assessing beliefs about the impact of CF); Personal and Treatment control (11 items assessing views about how amenable CF is to personal actions and treatment); Cause (18 items assessing attributions about possible causes for CF); and Emotional representations (six items assessing respondents’ emotional responses to their illness). With the exception of Illness identity, all subscales are rated on a standard five-point scale from strongly disagree to strongly agree. Responses to the Cause subscale were subsequently re-coded according to whether or not each participant endorsed an item as a cause of their CF. This instrument has been shown to have good reliability, construct, content, and predictive validity in a number of chronic conditions with the Identity, Time-line, and Consequences scales having previously been used in four studies of 11–18-year-olds with type 1 diabetes (Skinner et al., 2003). Although there are no previous studies with CF patients to provide psychometric data, in this sample internal consistency (Cronbach’s alpha) was good for all scales (Identity = 0.73; Time-line = 0.81; Consequences = 0.66; Personal control = 0.66; Treatment control = 0.73; Emotional representation = 0.84), except the Time-line-cyclical scale, with an alpha of 0.18, which was dropped from further analysis.

The Beliefs about Medicines Questionnaire-Specific (BMQ-Specific; Horne et al., 1999). This assesses respondents’ beliefs about the Necessity of a specific treatment and their Concerns about its potential adverse consequences. As with the IPQ-R, items are added or adapted for each treatment. In this study, adaptations assessed beliefs about CPT, ES, and antibiotics. For CPT, two items from the Concerns scale were changed and one was added: ‘I sometimes worry about the long-term effects of my ‘treatment’ and ‘I sometimes worry about becoming
too dependent on my ‘treatment’ were replaced with ‘My CPT is a hassle’ and ‘My CPT is difficult because I need help from others to do it properly’. For ES, one item was added: ‘My ES/antibiotics give me unpleasant side effects’. These alterations were made after the discussion with one of the pediatric teams involved in the study and approved by R. Horne (principal author of the BMQ). The modified questionnaire was titled the Beliefs about Treatment Questionnaire (BTQ) for the purposes of this study. The same five-point scale from 1 strongly disagree to 5 strongly agree described above was used. For the Necessity scales, internal consistency was good (Cronbach’s alpha physiotherapy = 0.90; ES = 0.81; antibiotics = 0.81), but for the Concerns scale, it was good for ES (alpha = 0.76) but poor for both physiotherapy and antibiotic use (alpha = 0.55 both scales).

Adherence was assessed using the Cystic Fibrosis Treatment Questionnaire (CFTQ), which was adapted from the Living with Cystic Fibrosis Questionnaire (Myers & Horn, 2006), which measures health perceptions and adherence behavior in CF patients. Patients are asked to rate their level of adherence to CPT, ES, and antibiotics according to: (a) how often they carry out the treatment as prescribed; (b) whether they see the treatment as part of their daily routine; and (c) how often they miss the treatment, using a five-point response format ranging from never to always. The CFTQ attempts to reduce over-reporting of adherence by normalizing the process of modifying treatment to suit lifestyle, with all scales evidencing good internal consistency (Cronbach’s alpha Physiotherapy = 0.91; ES = 0.85; Antibiotics = 0.81).

Non-adherence was assessed using the Medication Adherence Report Scale (MARS; Horne & Weinman, 2002). The MARS was designed to diminish the social pressure on individuals to over-estimate adherence by normalizing the process of adapting treatment to fit their lifestyles (Rand & Wise, 1994). The questionnaire is preceded by a statement assuring respondents that many people follow treatment in a way that suits them and that this may differ from recommendations given by health professionals. Respondents’ rate how frequently they engage in a non-adherent behavior (e.g., forgetting to take their treatment, or avoiding it) on a five-point scale from always to never. This focus on non-adherence challenges participants to recall acts that obstruct their self-care, in contrast with the CFTQ. Studies in non-CF samples indicate good reliability and internal consistency; a finding replicated in this study for the Physiotherapy and Antibiotic scales (Cronbach’s alpha = 0.88 and 0.75, respectively) but not for the ES scale, alpha = 0.51.

**Procedure**

Potential participants and their families were identified by a member of the pediatric team, and given or sent an information letter inviting them to participate. Those who agreed were encouraged to ask questions and discuss the study before completing a consent form. Once written consent had been obtained from the participant (and from parents, for those aged below 16), a home visit to administer the measures was arranged. In all cases, participants were assured that the study was independent of the hospital and that their responses were confidential and anonymous. It was hoped that this would encourage the adolescents to give their own views rather than those they considered to be socially desirable (Rand & Wise, 1994). The measures were administered in the following order: CFTQ, IPQ-R, BTQ, and MARS. The researcher (KH) sat with participants whilst they completed the questionnaires in order to answer any queries and reduce the risk of missing data.

**Data Analysis**

Data were analyzed using SPSS for Windows release 15.0.0 (2006). Subscale scores were calculated by taking the mean item score for each participant. Given that the ES subscale from the MARS had problematic internal consistency, and so as to avoid the problem of shared method variance, combined adherence scales were generated. Scores on each question were converted into z-scores. Those of the MARS (a measure of non-adherence) were inverted (multiplied by –1) and then the z-scores for the items in each subscale on both scales summed to produce three scores. The resultant Cronbach’s alphas for each scale were CPT, \( r = 0.94 \), ES, \( r = 0.84 \), and Antibiotics, \( r = 0.84 \). Subsequent analyses employed these combined adherence subscales.

Shapiro–Wilk tests revealed that eight subscales deviated significantly from the normal distribution: IPQ-R identity, time-line chronicity, and treatment control; BTQ CPT necessity and ES necessity and all of the combined adherence subscales. Transformation did not produce normally distributed data. Thus, for abnormally distributed variables, median and interquartile range values are reported. Given the large number of subscales with distributional difficulties, Spearman’s rank correlations were used to explore the relationships between respondents’ scores on the measures of self-reported adherence, personal model of CF, and age. Mann–Whitney U tests were used to investigate the relationship between gender and adherence. Finally, hierarchical multiple linear regression analyses were performed to identify the variables that best
accounted for reported adherence. Multiple regression is a relatively robust method and can be used if the residuals rather than the variables themselves are normally distributed (Howell, 1997). The distributions of standardized and unstandardized residuals were explored for all regression analyses. Regression analyses are reported where residuals were normally distributed. In order to avoid the problem of collinearity amongst variables (Tabachnick & Fidell, 1996), only those variables found to correlate significantly at a univariate level with reported adherence were included. As is the convention in multiple regression, effect sizes in the form of squared multiple correlations have been reported. One-tailed tests, alpha 0.05, were used throughout (Howell, 1997).

**Results**

**Descriptive Statistics**

Descriptive statistics for the IPQ-R, BTQ, and combined adherence scales are given in Table I. Those measures that had an internal consistency of less than 0.7 were deemed to be psychometrically unsound, so no further analysis of these scales (IPQ-R Time-line cyclical, Consequences, Personal control, BTQ Physiotherapy concerns, and Antibiotic concerns) is reported. Age was correlated with perceptions of Time-line chronicity, Emotional representation, Concerns about ES, and Necessity for physiotherapy (Table II) such that older participants were more likely to agree that they would have CF for the rest of their life, had fewer negative emotions about their illness, were less likely to think physiotherapy was necessary, and had fewer concerns about ES.

<table>
<thead>
<tr>
<th>Beliefs about treatment</th>
<th>Chest physiotherapy</th>
<th>Enzyme supplements</th>
<th>Antibiotics</th>
</tr>
</thead>
<tbody>
<tr>
<td>Necessity</td>
<td>3.7</td>
<td>4.0</td>
<td>3.9</td>
</tr>
<tr>
<td>Concerns</td>
<td>2.6</td>
<td>1.9</td>
<td>2.2</td>
</tr>
<tr>
<td>Combined adherence scale</td>
<td>0.2</td>
<td>0.3</td>
<td>0.2</td>
</tr>
</tbody>
</table>

IPQ-R: Revised Illness Perception Questionnaire; Combined adherence scale: total of individual z-scores for each item from the MARS (measures non-adherence, inverted) and the Cystic Fibrosis Treatment Scale (measures adherence). *Median and interquartile range reported.*

**Correlates of Reported Adherence**

There were no significant gender differences on the combined adherence measure. However, older participants were less likely to follow physiotherapy treatment recommendations and less likely to follow recommendations for antibiotic use, but there was no relationship with ES used (Table II). Time-line perceptions were associated with reported antibiotic adherence with longer time-line perceptions being associated with more adherence (Table II). Treatment control perceptions were significantly associated with antibiotic use, but not with CPT or enzyme use. Necessity beliefs were associated with all measures of CPT and antibiotic use. Consistent with our hypotheses, Emotional representation was not significantly correlated with any of the treatment measures.

**Exploratory Multiple Linear Regression Analyses**

To identify the variables that best accounted for adherence to CPT and antibiotics, hierarchical multiple linear regression analyses were conducted. To control for the effect of age, this was entered first. Personal models of CF were entered in Block 2. For CPT, predictor variables were age and BTQ Necessity. For antibiotics, predictor variables were IPQ-R Treatment control, BTQ Necessity, and IPQ-R Time-line chronicity.

Age was a significant predictor of CPT. However, when BTQ Necessity was added to the regression, age was no longer predictive of CPT (Table III). Given the significant correlations between age and BTQ Necessity and between BTQ Necessity and CPT adherence, this means that according to Baron and Kenny’s (1986) criteria, Necessity mediated the association between age and CPT adherence.
For antibiotic use, in Block 2, Treatment control and Time-line were predictive of both measures, with Antibiotic necessity also predictive of adherence (Table II). Furthermore, although age was predictive of antibiotic use, once Time-line entered the regression, age was no longer predictive of adherence. Given that age was correlated with Time-line perceptions, this indicated that Time-line perceptions were also mediating the association between age and antibiotic adherence.

The results of the regression analyses were the same if the individual subscales of the CFTQ and MARS adherence/non-adherence measures (except ES for MARS) were used independently.

Table II. Correlations Between Illness Perceptions, Treatment Beliefs and Reported Adherence (N = 38)

<table>
<thead>
<tr>
<th>Variable</th>
<th>Combined adherence scale</th>
<th>CPT</th>
<th>ES</th>
<th>Antibiotics</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age</td>
<td></td>
<td>-0.31</td>
<td>0.15</td>
<td>-0.41†</td>
</tr>
<tr>
<td>IPQ-R</td>
<td>Identity</td>
<td>0.08</td>
<td>0.15</td>
<td>0.06</td>
</tr>
<tr>
<td></td>
<td>Time-line—chronicity</td>
<td>0.32†</td>
<td>-0.19</td>
<td>0.03</td>
</tr>
<tr>
<td></td>
<td>Treatment control</td>
<td>-0.04</td>
<td>0.20</td>
<td>0.20</td>
</tr>
<tr>
<td></td>
<td>Emotional representations</td>
<td>-0.33†</td>
<td>0.04</td>
<td>-0.18</td>
</tr>
<tr>
<td>BTQ</td>
<td>Necessity</td>
<td>-0.31†; -0.12; -0.15</td>
<td>0.71*</td>
<td>0.03</td>
</tr>
<tr>
<td></td>
<td>ES Concerns</td>
<td>-0.47#</td>
<td>-1.7</td>
<td>-1.17</td>
</tr>
</tbody>
</table>

IPQ-R: Revised Illness Perception Questionnaire; BTQ: Beliefs about Treatment Questionnaire; Combined adherence scale: total of individual z-scores for each item from the MARS (measures non-adherence, inverted) and the Cystic Fibrosis Treatment Scale (measures adherence).

*Correlations given for associations between Age and Necessity for CPT, ES and antibiotics, respectively.

†p < .05; †p < .01; *p < .005; *p < .001 (one-tailed).

Table III. Hierarchical Multiple Linear Regression Models of Predictors of Reported Adherence to CPT and Antibiotics as Measured on the Combined Adherence Scale (MARS plus CFTQ).

<table>
<thead>
<tr>
<th>Variable</th>
<th>Model 1a: chest physiotherapy</th>
<th>Model 2a: antibiotics</th>
</tr>
</thead>
<tbody>
<tr>
<td>Block 1</td>
<td>Age</td>
<td>Age</td>
</tr>
<tr>
<td></td>
<td>-0.18†</td>
<td>-0.13†</td>
</tr>
<tr>
<td>Block 2</td>
<td>Age</td>
<td>IPQ-R treatment control</td>
</tr>
<tr>
<td></td>
<td>BTQ necessity</td>
<td>0.09†</td>
</tr>
<tr>
<td></td>
<td>Treatment control</td>
<td>0.03</td>
</tr>
<tr>
<td></td>
<td>Time-line</td>
<td>-0.06†</td>
</tr>
<tr>
<td></td>
<td>BTQ necessity</td>
<td>0.35†</td>
</tr>
</tbody>
</table>

Discussion

This study set out to explore whether the personal models of CF held by adolescents with CF are predictive of adherence to CF treatment. Consistent with previous research, adherence to CPT was the poorest in older adolescents (Foster et al., 2001). As hypothesized and replicating results from previous studies and a meta-analysis, the treatment beliefs component of the adolescents’ personal model were predictive of health behavior, with personal models of CF accounting for around 50% of the variance in self-reported adherence behavior. As hypothesized, for CPT and antibiotic use, treatment beliefs were significant predictors of adherence and, along with beliefs about the chronicity of CF, these
beliefs mediated the association between age and adherence. Finally, as predicted, emotional representations were not associated with adherence. These findings are consistent with research carried out in other illness groups, which highlights the importance of perceived control and time-line beliefs in shaping adherence behavior (Cardiac disease: Cooper et al., 1999; Petrie et al., 1996. Diabetes: Griva et al., 2000. Hypertension: Meyer et al., 1985. Meta analysis: Hageer & Orbell, 2002).

The finding that perceptions of illness time-line were predictive of adherence behavior may be somewhat surprising. This suggests that there is variability in young people’s perceptions of how long they will have CF, with the response of 13% of this sample indicating that they do not agree with all the items that indicate their CF is a permanent condition they will have for the rest of their life. This percentage is strikingly consistent with responses of similarly aged participants with type 1 diabetes, where 12%, 14%, and 13% of participants did not agree their diabetes was a permanent condition they would have for the rest of their life (Skinner et al., 2003). The fact that older participants in studies 3 and 4 were more likely to agree that their diabetes was permanent suggests that this may reflect the maturity of responders, but this was not associated with illness duration. However, the fact that older participants both in this study and in the diabetes studies were more likely to agree that their condition was permanent suggests that this may reflect the maturity of responders or greater acceptance as the participants grow older. Clearly, this is an issue that warrants further investigation.

However, none of the components of the adolescents’ personal models were associated with the use of ES. Failing to take ES with food typically results in stomach pains, bloating, and bulky stools (Koocher et al., 1992). Thus, taking ES produces symptom relief that patients can clearly relate to the treatment. This strong link between the immediate symptoms and ES use may suggest that other factors contribute to non-adherence to this aspect of treatment. Item analysis indicates that only low levels of non-adherence to ES were reported and this is largely due to forgetting, consistent with previous research (Conway et al., 1996). Thus, participants may not be making rational decisions to neglect this aspect of treatment, but simply forget to follow their regimen from time to time. As such, their personal beliefs about CF and ES may not influence their adherence behavior.

The present study provides important, preliminary information about the relationships between personal models and reported adherence in adolescents with CF. However, the results of this study must be interpreted with caution. First, the primary outcome measure of adherence was assessed by two self-report methods. As noted in a review of literature on adherence in CF (Kettler, Sawyer, Winefield, & Greville, 2002), no measure of adherence is completely reliable and valid. Even with blood assay methods, pharmacokinetic variation may affect results, and electronic monitoring devices, despite being regarded as the most objective measure available, do not guarantee that a tablet is taken even when a bottle is opened. Therefore, given the preliminary nature of this study, and the resource implications of assay or electronic methods, it was felt appropriate to rely on self-report data. However, as there can be bias in the reporting of adherence behavior, it was also decided to obtain self-reports of non-adherence. There is evidence to suggest that when individuals report non-adherence, they usually tell the truth (Epstein & Cluss, 1982; Horne, 2000). Indeed, some authors argue that, whilst reported adherence may be over-rated, reported non-adherence is usually more accurate (Abott & Gee, 1998).

Although combining the measures of adherence/non-adherence deals with the potential problem of shared method variance, neither measure was objective. Clearly, before developing clinical interventions focused on these identified beliefs, the study needs to be replicated with at least one independent report of adherence behavior. Ideally, this should be undertaken using more direct measures of the individuals’ behavior; this being easiest for taking antibiotics and ES, where pill bottle lids with implanted microchips can be used, with electronic physiotherapy vests being another option (Kettler et al., 2002).

A second problem is the assessment of the personal models of CF, with one of the scales used lacking sufficient internal consistency to warrant its use in subsequent inferential statistics. The preliminary nature of this study means that there were few measures available previously validated in CF populations, resulting in amendments to some items and addition of others, in an attempt to address relevant, specific features of CF treatment and care. Although, this approach seems to have been successful for many of the scales of interest, there is clearly a need to develop the Concerns scales further before completing replication studies, and to improve the internal consistency of the Personal control and Consequences scales of the IPQ-R. Similar research in adolescents with type 1 diabetes suggests that a combination of generic and disease specific scales is likely to be the most efficacious way of assessing personal models in more complex conditions such as CF and diabetes (Skinner et al., 2003).
Due to the cross-sectional nature of the study, it is not possible to infer causality. Thus, in future, longitudinal research is required to clarify the direction of the relationships between variables. Moreover, using prospective designs would enable researchers to investigate changes in personal illness models and adherence behavior over time. The SRM posits that personal models of illness and decisions about whether or not to adhere to treatment are constantly being reviewed as patients appraise the effectiveness of their behavior in terms of symptom relief (e.g., did adhering to treatment work?). Thus, full validation of the model requires longitudinal studies designed to assess how the relationships between variables change in response to symptom fluctuations; although longitudinal studies in other chronic conditions have supported the SRM (e.g., Aikens, Nease, Nau, Klinkman, & Schwenk, 2005; Horne, Cooper, Gellaitry, Leake-Date, & Fisher, 2007; Gonzalez et al., 2007).

Due to the relatively low prevalence of CF and the severity of its impact on patients’ lives, it can be difficult to recruit large numbers of people to CF research (Anthony, Paxton, Bines, & Phelan, 1999). In this context, a sample of 38 adolescents represents successful recruitment. Furthermore, the response rate was high (73%) and there were no differences in age or gender between those who did and did not take part. Thus, we would argue that the sample is likely to be representative of the population of adolescents with CF. Nonetheless, corroborating evidence with additional samples is required. Furthermore, with larger sample sizes more sophisticated statistical techniques, such as structural equation modeling, could be used to explore the interactions between variables (Tabachnick & Fidell, 1996). This would enable the proposed mediating role of treatment necessity beliefs to be investigated. However, the low numbers must be borne in mind, so that the multiple regression analyses need to be treated with some caution, although a variable to case ratio of 1:9.5 or 1:10 has been maintained.

This study makes a new contribution to the CF adherence literature and has some important implications for clinical practice. The results, supporting all hypothesized associations, suggest that the SRM may be a useful framework from which to explore adherence in adolescents with CF. Moreover, if the findings can be replicated in prospective studies, they suggest that assessing adolescents’ perceptions of CF and its treatment may enable clinicians to identify patients at risk of non-adherence, and subsequently, develop appropriate strategies to enhance adherence in this group. For example, an intervention, which demonstrates that CPT has benefits (perhaps by measuring and discussing pre- and post-treatment lung function with teenagers) may improve adherence. In sum, this study has provided evidence regarding the nature of the personal model of CF held by adolescents and about the contribution of these variables to explaining reported adherence to CPT, ES, and antibiotics. Longitudinal studies using larger sample sizes are now required to confirm the findings. These may help clinicians to develop strategies to enhance adherence, and subsequently, improve survival rates for adolescents with CF.

Acknowledgments

We would like to thank the following for all their help in identifying participants: Keith Foote and Beverley Murray at the Royal Hampshire County Hospital; Gary Connett and Judy Madison at Southampton General Hospital; Sheila Peters and her colleagues at St Mary’s Hospital, Portsmouth; and Anne Dewer at Poole General Hospital. Finally, and most importantly, we would like to thank the young people who gave their time to take part in this research.

Conflicts of interest: None declared.

Received June 13, 2008; revisions received November 18, 2008; accepted November 25, 2008

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