

**BRIDGING THE GAP: ADHERENCE TO TREATMENT  
FOR  
CYSTIC FIBROSIS IN ADOLESCENCE**

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**Submitted in part fulfilment for the degree of Doctor of Clinical  
Psychology  
July 1997**

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# **Bridging the Gap: Adherence to Treatment for Cystic Fibrosis in Adolescence**

**Sarah Longley**

## **ABSTRACT**

The purpose of this study was to explore the issue of adherence to treatment for Cystic Fibrosis (CF) in adolescence. Poor adherence may lead to accelerated deterioration, increased frequency of hospital admissions, outpatient appointments and reduced ability to predict the efficacy of treatments. Several studies have shown adherence tends to decrease during adolescence (Gudas et al 1991; Patterson et al 1993). The literature, however, has repeatedly concluded that the influences upon adherence are elusive and highly complex, indicating the need for in depth qualitative study of this issue.

The grounded theory method (Strauss & Corbin 1990) was used to generate a substantive theory from the accounts of eleven young people who suffer from CF, aged between ten and eighteen years.

The theory is described in terms of the sufferer needing to 'bridge the gap' between normality and difference effectively. It is as if the sufferer and his/her family could be visualised as standing with one foot either side of a broken swing bridge. In order to survive they have to keep their balance. Illness symptoms and treatment serve to pull the sufferer to one side or the other at different times. The strategies used to manage the gap can be learned from the familial response to the disease. The beliefs the sufferer develops in relation to the illness and the value of treatment can provide him/her with the tools to maintain a balance in a variety of circumstances.

The theory 'bridging the gap' is discussed in terms of the current literature pertaining to adherence to treatment for CF and speculation regarding its intersection with attachment theory. The potential implications for clinical intervention generated through the relating of the two theories are considered.

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## **INTRODUCTION**

### **Cystic Fibrosis**

Cystic Fibrosis (CF) is one of the most common terminal genetic diseases of white children, affecting approximately one in two thousand five hundred live births (CF Trust 1995). The disease leads to a number of problems, including blockage of the pancreatic ducts and insufficient enzymes for efficient food digestion, which result in malnutrition if untreated. The disease causes dehydration of the mucus found in the respiratory tract making the lungs more prone to infection. It is usually progressive lung disease, due to increased susceptibility to infection, that is ultimately fatal for the affected individual.

Due to advances in treatments for CF the average life expectancy now reaches into the late twenties with some sufferers living well into their thirties or even reaching their forties. The average life expectancy in 1991 was twenty nine years (Cystic Fibrosis Foundation Patient Data Registry 1991) with a predicted average life expectancy for a baby born now of forty years (Duncan-Skingle 1992). Treatment regimes are, however, very rigorous and require much commitment and time, not to mention endurance on behalf of sufferers and their families. As Parcel et al (1994) point out, the requirements of the treatment and consequently the demands made and the skills required by the CF sufferer and his/her family are continually changing.

Treatments are aimed at preventing lung infections, as far as is possible, and alleviating the effects of the disease. Daily management typically involves chest physiotherapy at least twice daily, rigorous exercise, oral and inhaled medication for the respiratory system and oral pancreatic enzymes and vitamins for the control of malabsorption. Sufferers also require a very high calorie diet to ensure adequate nutrition. As the disease progresses they may require dietary supplements typically delivered overnight by nasogastric or gastrostomy tube, overnight oxygen and regular intravenous antibiotics to fight infections.

### **The concept of adherence**

Traditionally the phenomenon of adherence has been referred to as compliance. Haynes, Taylor & Sackett (1979) proposed the definition of compliance as "the extent to which a person's behaviour coincides with medical advice". The term therefore refers to both adherence to preventative self care recommendations and

prescribed therapeutic medical regimens. The phenomenon relates to all kinds of disease or illness in people of all ages, but perhaps presents specific problems to those with chronic illnesses, due to the long term and often complex nature of the associated treatment regimes (Haynes 1976; Blackwell 1992).

Adherence is becoming the preferred term, as compliance is considered by critics to engender too many connotations of authoritarianism. The term compliance is purported to imply that the patient is a passive recipient of medical prescription rather than being actively involved in his/her own self care. This is considered by Donovan (1995) to be a particularly outdated stance within modern health care systems, within which there is a predominance of patients with chronic illness who continually question the opinion and advice of their health care teams. In view of the implicit suggestion of the patient taking an active stance to treatment and of a greater degree of collaboration within the doctor patient relationship, adherence will be the preferred term throughout this thesis.

#### **Adherence to treatment in CF sufferers**

The literature focusing specifically upon adherence to CF treatment is at present relatively sparse and tends to concentrate either on adult patients or upon the parents or family rather than on child and adolescent sufferers. Pownceby (1996) in a study of adolescent and young adult CF sufferers suggests CF patients are a relatively adherent group as compared to other chronic illness sufferers.

As Abbott et al (1993) point out, there is no conclusive link between poor adherence to treatment regimens and progressive disease in CF. Nevertheless, adherence is an area highlighted by medics as important for study in the light of increasing patient involvement in self care. Poor adherence may lead to accelerated deterioration, irreversible organ damage, more outpatient appointments, increased frequency of hospital admissions and reduced ability to predict the efficacy of available treatments.

Studies of adherence in chronic illness initially suggested that adherence is related to such factors as the likely negative effects of stopping treatment, conflicts in medical opinion, number of different therapeutic goals and the patient's perception of the severity of the illness. (Rapoff & Christophersen 1982 cited in Koocher, McGrath & Gudas 1990).

Developments in the literature have expanded the ideas as to factors likely to have a bearing on adherence, either through direct study or via inferences made indirectly, to include more subtle factors such as psychosocial and family functioning (Blair, Cull & Freeman 1994; Sawyer 1992), education and knowledge regarding the disease (Henley & Hill 1990), coping styles (Bombardier, D'Amico & Jordan 1990) psychological and psychosocial adjustment (Aspin 1991; Mullins et al 1991) and health beliefs (Brown, Rowley & Helms 1994).

The major themes raised by studies such as these will be expanded and discussed with reference to similar literature relating to treatment adherence. The main focus will be placed upon those studies concerned specifically with CF sufferers. Occasionally literature relating to other chronic illnesses will be drawn upon, where this is considered to be particularly relevant to the focus of this study. This reflects the view that many of the issues discussed are applicable to many different populations of chronic illness sufferers although, clearly, there are some characteristics which are pertinent to specific disease types.

### **Severity of illness**

Several studies have investigated links between adherence and severity of disease with different conclusions. Czajowski & Koocher (1987) found that adherence to physiotherapy was reduced with increased severity of illness. However, they suggest this may be a function of the increased treatment requirements rather than a response to severity per se. In a more recent study, Abbott et al (1995) concluded that there is no association between adherence and severity. They note that this may be a representation of the difference in patients' perceptions of severity, ratings of close companions, and medical opinions. It seems, in the light of these mixed findings, the relationship is unlikely to be a simple one and will be subject to individual variation.

In a study of adult CF sufferers, Abbott et al (1994) found that adherence tended to be treatment specific with more patients adhering to exercise and enzyme routines than with physiotherapy and vitamin treatments. They found no relationship between demographic variables such as age, sex, employment status, frequency of clinic visits or with disease severity. Those patients who felt immediate benefits from a treatment or therapy were more likely to adhere.

## **Doctor-patient relationship**

The doctor patient relationship has been cited frequently as a factor influencing adherence to treatment regimens prescribed for a wide variety of complaints. Poor communication has repeatedly been shown to relate to poor adherence (Kessler 1991; Proos et al 1992; cited in Donovan 1995). It has been suggested for many years that if patients perceive the physician to at least be attempting to see the situation from their perspective, and treats them with warmth and respect, this leads to a better alliance and the likelihood of greater adherence (Korsch et al 1971).

Donovan (1995) states that patients make their own decisions about treatment based on their beliefs, circumstances and the information they have available. Koocher et al (1990) suggest that this decision making process should be a significant feature of the doctor patient relationship so that the costs and benefits of treatment can be explored collaboratively. Pownceby (1996) found that unrealistic expectations and authoritarian attitudes on the part of physicians lead to lower levels of adherence in adolescent and young adult CF sufferers. Conversely, she suggests that flexibility and openness in the doctor patient relationship may positively influence adherence.

In order to try to enhance the understanding of the nature of adherence and possible reasons for poor adherence, Koocher, McGrath & Gudas (1990) conducted a critical incident analysis from descriptions of crises obtained from CF patients of all ages and their families. They identified three main types of non adherence namely, inadequate knowledge, psychosocial resistance and educated non adherence. In instances where educated non-adherence is clearly based on reasoned and well thought out decisions they suggest intervention by the physicians concerned is not beneficial. They conclude that interventions need necessarily to be based on issues specific to each individual patient, if they are to stand a chance of being successful, due to the complexity of factors likely to be influencing adherence.

Clearly the dynamics of the doctor patient relationship and the influence of these upon adherence is an area worthy of further investigation. The links between patients' relationships with the health care providers and other significant relationships, such as family and friends may be a particularly worthwhile area for further exploration.

## **Role of the family**

Many studies have focused upon the role of the family in influencing adherence to treatment regimes. In a relatively early study, Patterson (1985) demonstrated that family resources and coping played a significant role in the level of adherence to treatment in CF sufferers aged three months to twenty-eight years. Adherence was higher in those families who were more expressive, where the mother was not working, and parents demonstrated a joint approach to coping to help create an atmosphere of integration, optimism and co-operation. Patterson (1985) therefore suggested the need for clinicians to attend to the needs of the family, as well as those of the CF patient, in order to optimise the CF patient's health.

Patterson et al (1993) also carried out a longitudinal study over a ten year period to look at the interaction between pulmonary health, family coping and adherence to treatment. They found that pulmonary health was better in families in which the parents emphasised integration, support for self and medical consultation. They found adherence with daily chest physiotherapy was linked to improved pulmonary health. Adherence was lower in those patients who were older and in those whose parents spent more hours working away from home.

Poorer pulmonary health was associated with a higher level of social contacts amongst family members. This finding is consistent with that of Geiss et al (1992) who found adherence was perceived by physicians to be higher when a patient's mother had less frequent social contacts. Interestingly, they also found higher perceived adherence to be associated with less satisfactory marital relationships between the parents of CF patients. They interpret the less satisfactory marriage as a symptom of the strain of trying to manage the child's disease. The authors therefore suggest this may highlight a need for a balance to be found between level of adherence and psychological adjustment to the illness within the family. However, as Drotar (1995) points out, the processes by which some families manage to achieve and maintain this delicate balance is far from clear.

## **Coping**

Kessler et al (1985 pp.550) define coping as "cognitive and behavioural efforts made to master, tolerate or reduce demands that tax or exceed a person's resources." Research on coping and chronic illness has considered both individual and family responses to the illness. Mullins et al (1991) in a study of mothers of

children who have CF suggested there are high levels of avoidance and denial in chronic illness populations. Increased levels of distress in the mother were associated with a greater number of stressful life events for the family and an escape avoidance style of coping. However the direction of causality was not established.

Kornas-Biela (1990), in a study of the parents of children who had CF, found that the most positive indicators in terms of parental coping and adjustment of the child were optimistic outlook, confidence in mutual assistance, and active encouragement of the development of the child's independence.

Bombardier, D'Amico & Jordan (1990), in a study of adults with a range of chronic illnesses, found a link between problem focused coping and better adherence to treatment, though the direction of causality was not conclusively established. Adherence was also positively related to the sufferer's belief that he/she has some control over the illness and that treatment would positively impact upon the disease course.

Hauser et al (1993), in a study of families and adolescent coping with diabetes, suggested that families which were cohesive and interconnected were better able to deal with the diabetes and the adolescents were more adherent to treatment. The emphasis was placed upon coping, mastery, optimism, flexibility in thinking, self reliance, co-ordination and active concern for feelings with some minimisation. In contrast, adolescents who were less adherent had families who were more disjointed. They showed higher levels of helplessness, lower levels of mastery, lots of minimisation of feelings, higher levels of anxiety and avoidance, a predominance of negative family legacies, poor co-ordination and little sense of the influence of past experiences upon their present efforts to cope.

In a study of adolescents with CF, Czajowski & Koocher (1987) identified six main themes to be related to coping. These were understanding the severity of the illness, responsibility for medications at home, information seeking regarding the illness, future goal orientation, involvement in school work and openness with peers regarding the illness. Principally they suggested adolescents who adhered to treatment tended to have adaptive beliefs and attitudes, in that they were more interested in and optimistic about their health. Those who were non-adherent tended to minimise their illness and use avoidant coping strategies.

They also found early adolescents relied most heavily upon coping strategies which encouraged interaction with peers. Middle adolescents still relied on peers, but forward planning for the future became more of a concern. The older adolescents were primarily concerned with establishing independence and autonomy. A realistic acceptance of the severity of the illness for this age group enhanced adaptive functioning. Poor adherence was expressed through passive resistance to treatment regimes rather than in an overt, openly rebellious manner.

### **Impact of CF upon adolescent development**

In most Western cultures adolescence is considered to extend from age ten or eleven through to the late teens and sometimes into the early twenties. Adolescence is considered to be a complex phase of development even for the physically 'normal' individual. During this phase of the life cycle the young person is thought to be required to become adept at forming relationships with members of the opposite sex, to adapt to his/her emerging sexuality and to become autonomous in both the behavioural and emotional sense. Adolescents are expected to prepare through education, and later work experience, to take on adult roles in society and to resolve the identity and value issues these new roles raise (Elliott & Feldman 1990).

These tasks present added challenges for the CF sufferer. Anxiety about sexual relationships may be complicated by concerns about the genetics of the disorder and outwardly noticeable signs of disease. These may include a persistent cough, a portocatheter for the delivery of intravenous antibiotics and physical immaturity due to malnutrition. Sawyer et al (1995) found that adolescent CF sufferers, particularly females, had significantly poor self esteem in relation to body image as compared to healthy controls. They discuss this in terms of delayed pubertal development which is more marked in female CF sufferers. Treatment issues, such as the requirement to be heavier than one might like in order to avoid malnutrition, may add further complexity to this issue, perhaps particularly for females.

The pursuit of careers or further education may be persistently interrupted by bouts of illness and increasing frequency of hospital admission, so adding complication to the taking on of adult roles. The development of autonomy, likewise, brings its own struggles, as the young CF sufferer has to take over the responsibility for a rigorous treatment regime in addition to those responsibilities adopted by non-afflicted teenagers.

Cognitive developmental theory of adolescence sees the young person as acquiring the capacity to reason in the abstract, enabling him/her to deal with hypothetical situations rather than just actual events. This stage of development is labelled formal operational thought by Piaget (Piaget & Inhelder 1958). This allows the adolescent to grapple with existential issues such as the meaning of life and the finality of death. These issues, as Pumariega (1982) points out, present particular challenges for the CF sufferer in view of his/her expected foreshortened lifespan.

The adolescent's relationship with his/her parents inevitably undergoes significant change as the young person struggles to develop into an independent, autonomous adult. Hall (1984) proposed that adolescent conflict revolved around communication difficulties, poor negotiation skills and poor problem solving abilities.

Eiser (1993) suggests that gaining autonomy and maintaining an inter-dependent relationship with ones parents need not be mutually exclusive. She suggests that the balancing of these two needs forms a normal part of growth and development for the adolescent.

Heaven (1994) suggests that one of the core tasks faced by the adolescent is to find ways to balance the demands he/she places upon him/herself with those of his/her parents and peers. All of these things may be contradictory to varying degrees. A high degree of flexibility in thinking is consequently required if the dilemmas faced are to be negotiated to the best advantage of the adolescent. The precariousness of this kind of negotiation is exacerbated for the adolescent living with a chronic illness, perhaps particularly a life threatening one such as CF.

Erikson (1959) regarded identity formation as the singularly most important task in adolescence. Achievement of a stable identity, according to Erickson (1959), involves the integration of those characteristics of significant others in the adolescent's life and those characteristics which are unique to the person. This process can, as pointed out by Pumariega (1982), be particularly complicated for the adolescent CF sufferer. Identity formation is unavoidably influenced by a greater degree of uncertainty regarding the future, outwardly noticeable symptoms and effects on physical growth, awareness of the genetic aspects of the disease and a greater degree of necessity for dependency upon parents and clinicians.

## **Adherence in adolescence**

Adolescence has become a topic of particular interest when considering adherence to treatment in chronic disease generally, as several studies have shown that adherence tends to decrease around this time (Gudas, Koocher & Wypij 1991; Patterson et al 1993). The reasons for this dip in adherence are likely to be multifactorial and complex, as indicated by the issues raised in the previous section.

Gudas, Koocher & Wypij (1991) studied perceptions of adherence according to reports of the patient, his/her parents and physicians amongst CF sufferers aged five to twenty. It was shown that perceived adherence was higher amongst younger children and was also related to increased levels of optimism and child knowledge of the illness. The relationship between these variables differed with age, suggesting the need to take a developmental perspective and to tailor interventions to the needs of the age groups at which they are targeted. This need is supported by Perrin et al (1991) who demonstrated clear systematic differences in children's reasoning about, and understanding of their illness, according to their age and illness experience.

Patterson et al (1993) suggest part of the explanation for decreasing adherence may lie in the idea that as children become older and more reluctant to adhere to treatment regimes, their parents try harder to enforce these. The parents having devoted many years to their child's illness are understandably reluctant to relinquish control for fear of negative consequences. Accordingly, the rebellious adolescent resists further so that parents become weary of the fight and leave the adolescent to his/her own devices. CF may therefore tend to become a battle ground for the adolescent struggle to obtain independent identity and autonomy.

McCracken (1995) suggests these difficulties may be avoided if the parents are able gradually to hand over responsibility for self care and treatment across the whole period of childhood, rather than waiting until the child reaches adolescence. This then enables the child to develop a sense of mastery and control over his/her disease. This reduces the likelihood of power struggles and facilitates an interdependent and collaborative relationship between the CF sufferer and his/her parents. This perspective is supported by other researchers who have focused specifically upon CF sufferers and advocate this as the preferred goal, as opposed to the traditional dependent/independent split (Gudas et al 1991; Pownceby 1996).

Other suggested factors of importance are peer influences, and the dilemma of feeling the need to blend in with ones peers whilst CF makes one seem intrinsically different. For example Koocher, McGrath & Gudas (1990) describe the case of a teenager having to go straight home from school for physiotherapy rather than to the coffee shop with the rest of the group.

McCracken (1984) proposed that flexibility is required within treatment regimes in order to facilitate the young CF sufferer engaging in an enjoyable recreational and social life. The availability of social support has been shown to be related to higher levels of adherence in adults, with clinical observations suggesting this may also be true for adolescent sufferers (Friedman & Litt 1986).

### **A case for further study of adherence in adolescent CF sufferers**

Although issues around adherence to CF treatment during adolescence have been touched upon in various ways in the literature, there have been few in depth studies specifically looking at teenagers' perspectives on having CF, and the effect of the disease and its treatment upon their lives. As the literature clearly points to there being a complex web of psychological and psychosocial factors influencing adherence at any stage in the life cycle, this area is obviously ripe for further study.

Relatively detailed, though still quite patchy, research has been carried out in respect to other chronic illnesses, particularly diabetes and asthma. However it could be argued that although some factors from such research may be generalised to CF, the emphases and crucial issues may well be different for CF sufferers. This could be a function of disease features which are specific to CF. These include a more complex and rigorous treatment regime, the knowledge that CF is ultimately terminal and marked genetic implications.

### **Critique of methodology used to study adherence to date**

Studies to date have tended to use a variety of questionnaires or self report diary type methods to investigate issues around adherence. In their study from the adolescent and child's perspective, Gudas et al (1991) used the Medical Compliance Incomplete Stories Test (M.C.I.S.T.) to assess general attitudes towards adherence. This test involves the child completing five stories about children going for various types of medical intervention. The stories are then transcribed and analysed along three dimensions of compliance/coping, optimism

and self efficacy. This instrument is advantageous in that excellent inter-rater reliability is easy to achieve, and it has been shown to discriminate between adherent and non-adherent adolescents with CF (Czajowsky & Koocher cited in Gudas & Koocher 1991). However, as only five scenarios are given the instrument has limitations as to eliciting rich information regarding the factors, beliefs and issues associated with adherence. Perhaps more fundamentally, the measurement of adherence is fraught with difficulty due to discrepancy between patient reports, actual behaviour and doctors' perceptions of adherence, therefore further limiting the value of such an instrument.

In the absence of published questionnaires on adherence, other studies, such as that by Abbott et al (1994), have used specially devised questionnaires or health diaries (Brown, Rowley & Helms 1994). Although these explore issues of degree of adherence to treatment, patients' perceptions of efficacy of treatment and reasons for non-adherence (Abbott et al 1994), the response sets seem to be quite limited and may force oversimplification of interaction between perceptions, beliefs and actions. Questionnaires, however, do have the advantage of allowing clear statistical comparisons and may have more scope from which to make generalisations. Nevertheless, as Banister et al (1994) suggest, even this type of quantitative study, involving statistics and rigorously controlled methodology, is not free from the experimenter bias and reflexivity that is often levelled as a criticism of qualitative methods.

Similarly, Brown, Rowley & Helms (1994) used health diaries with children and adolescents to explore the links between the symptoms, health status and illness behaviour. Although these diaries assessed psychological issues, to some extent these were very superficial, taking account of aspects such as behaviours of missing school or exercise, and requesting a simple rating of 'how bothered' the individual was by the symptoms. Such a rating does not enable exploration of contradictory view points and ambivalent responses. These may form the crux of the matter in terms of understanding adherence to treatment for chronic illness in adolescence.

Koocher, McGrath & Gudas (1990), in their qualitative study of critical incidents in CF patients and their families, produced some very rich data regarding nonadherence which added to the research base for interventions. However, as the study combined the perceptions of families and CF sufferers of all ages, their findings were not specific to adolescents. In addition by concentrating on critical

incidents it seems likely that their study would not have facilitated much insight into the factors associated with good adherence and positive treatment motivation.

The most comprehensive and informative project to date to consider issues of adherence to treatment in adolescent and young adult CF sufferers was the Coming of Age Project (Pownceby 1996). This study took the perspective of 104 adolescent and young adult CF sufferers attending 10 different CF centres and focused upon the transition to adult health care services and the impact of this upon adherence. Data collection included a comprehensive mixture of qualitative and quantitative methods.

The study once again concluded that the issue of influences upon adherence is elusive and highly complex. It was suggested that the seed for adherence behaviour, which is interrelated with family factors, health beliefs and coping, are sown early in life and become expressed when children reach adolescence and develop their own autonomy. The adherence levels established in adolescence then appear to remain relatively static throughout adulthood.

Although, on the face of it, this study addressed much of the gap in research relating to living with CF and its treatment from the teenage and young adult perspective, the level of analysis and grounding in psychological knowledge is in reality quite superficial. As a consequence, the finished project draws together and verifies most of the issues raised in current literature, but does not develop the theories any further. This leaves many questions unanswered and clearly highlights the need for more in depth qualitative research in this area, particularly that focusing upon the transition to adolescence.

### **A Case For Qualitative Research**

When considering the literature relating to adherence it could be argued that if the factors affecting adherence are so complex, the most important aspects will not be tapped by a simplistic enquiry. Stevenson & Cooper (1997) point out that traditionally psychology has its roots in a positivist position and has advocated the use of quantitative research methods in the pursuit of establishing the discipline as a pure science. The emphasis, therefore, has been placed upon objectivity, as opposed to subjectivity which is embraced within the qualitative paradigm.

The essence of qualitative research comes from the very different position of constructivism. Within the constructivist school of thought several different approaches to qualitative research can be taken. The core assumption, however, is that meanings do not reflect one reality as it exists in the world. Instead, constructivists advocate that meanings are constructed by individuals within social, cultural, political and historical contexts, so allowing for many versions of reality (Henwood & Nicholson 1995). This process is almost by definition highly subjective.

Qualitative research, by its very nature, incorporates the idea that people's experiences, attitudes and beliefs are often extremely complex and contradictory (Griffen 1986). As Blackwell (1992) suggests, the truly significant issues, in terms of adherence, relate to a person's beliefs, attitudes and value judgements about his/her illness and its care.

It seems that the conflicts arising in adolescence are likely further to complicate this. Detailed study of the adolescent's perspective, which tries to tap into his/her experiences and ways of making sense of his/her condition, could provide valuable insight which may be missed by questionnaire data. Gower (1985) suggests that qualitative interviewing enables the understanding of other people's constructions of reality, by enquiring about their attitudes and beliefs in such a way that the rich context that is the substance of their meanings is addressed.

If, as Charmaz (1990) intimates, health professionals can, through qualitative study, be offered alternative understandings of patients' beliefs and behaviour, they are then in position to act upon these insights as defined by the patient.

Eiser (1990) points out that increases in understanding the psychological aspects of chronic disease have been determined by theories and interventions developed in adult health psychology. She suggests that this may be restrictive as it does not always address the context of the child's development. A qualitative project, which embraces the richness of the adolescent CF sufferers experience, may liberate the psychological understanding of adherence from this restriction to some extent, by consideration of the adolescent developmental context. This may be particularly true when account is taken of the processual nature of chronic illness per se, a feature which makes chronic illness particularly suited to qualitative study (Conrad 1990).

## **Initial research questions**

In the light of the literature discussed so far, and perhaps particularly the findings of the Coming of Age project, the following research questions formed the basis of enquiry for this study:-

- What is the impact of reaching adolescence upon the meaning of CF for the sufferer and his/her ability to cope with the disease and its treatment?
- What kinds of illness and life experiences significantly influence the adolescents view of CF and its treatment? In what ways do these experiences impact upon adherence to treatment?
- What kinds of experiences enable or disable the adolescent CF sufferer from coping with the illness and its treatment in particular ways in particular sets of circumstances?

The ultimate aim, in exploring the issues posed by these questions, was to raise possibilities to inform further the interventions made with regard to adherence to treatment in adolescent CF sufferers, from a psychological perspective.

## **METHOD**

### **Grounded theory**

Grounded theory was first proposed by two sociologists, Glaser and Strauss (1967). They described a general qualitative methodology aimed at developing theory which has its basis, is grounded, in data which is systematically collected and analysed. The theory is developed in parallel with data collection. Grounded theory analysis principally begins with the data and remains close to the data. The researcher begins the study with some general research questions as opposed to clearly defined hypotheses associated with traditional empiricist research.

Throughout the process of analysis and theory building abstractions on different levels are made from the data and are then checked and refined by systematically returning to the data or by collecting further data. This process is referred to as theoretical sampling. This overall process of developing concepts and categories and testing against the data is described as "(constant) comparative analysis" Glaser & Strauss 1967, p.vii).

It is this feature of grounded theory which gives the method scientific rigour and allows it to "meet the criteria for doing 'good' science" (Strauss & Corbin 1990 pp 27). This, according to Strauss & Corbin (1990), is achieved by a mixture of creativity, which allows for the generation of new trains of thought or angles on the phenomena under study, and validation through constant and systematic comparison. They argue that if the procedures are carried out well the theory generated will be conceptually dense and free from biases and assumptions. Such biases may be present prior to, and may develop during, the analytic process. The resultant theory will be sufficiently abstract to make it applicable in a variety of contexts related to the phenomenon under study.

The use of grounded theory, since it was first explicated, has varied in actual practice from that methodology and theoretical perspective first described by Glaser & Strauss (1967). Researchers, who until recently have largely been sociologists, have taken different emphases depending upon the subject to be studied and their own theoretical roots.

Charmaz (1995) argues that grounded theory is compatible with both positivist and constructivist viewpoints and so can span the divide between empiricist and

interpretative methods within psychology. Charmaz (1990) purports to take a more interpretative position believing theory to be created through the interaction between the researcher's theoretical position, his/her relationship with respondents and the influence of these interactions upon the reading of the data. She advocates that her approach has its roots in symbolic interactionism and phenomenology. Symbolic interactionism assumes that a person will respond to the actions of others on the basis of the interpretations he/she makes regarding the intent of these actions.

Smith, Harre & Van Langenhove (1995) argue from a phenomenological perspective that true advances in psychological knowledge can only be made through the scrutiny of psychological meanings. They advocate that this requires an approach different from that of pure objectivity, as psychological meanings are phenomena which are reflections of the way in which individual people create meaning in relation to certain aspects of their world. This stance is then broadened out further by Charmaz (1990) by drawing upon Marxist theory which takes account of the ways in which social structures impact upon the psychological reality for the individual.

Glaser & Strauss (1967) and Strauss & Corbin (1990) take a more traditional positivistic stance, believing the methods they propose sit comfortably with the assumptions of an external reality which can be discovered and accurately recorded. They suggest that if the researcher is unbiased the reality of the researched phenomenon will be readily apparent.

The analytic procedures used in this project were guided by the principles laid out in Strauss & Corbin (1990) but drew upon the theoretical perspective taken by Charmaz (1990). Essentially, it was considered from the outset that the perspective of the researcher, and the interaction of this with the thoughts, feelings and descriptions of the respondents, create the meanings placed upon the data.

## **Procedure**

### ***Access/local context***

The emphasis of the study was driven by the wish of the team working in the Cystic Fibrosis clinic, particularly the consultant, to understand issues relating to adherence to treatment in adolescence in order to find ways to intervene more effectively. The team was about to be joined by a clinical psychologist on a sessional basis. It was hoped that the findings of this study would be instrumental in shaping the nature of the service provided by the psychologist to the team.

### ***Ethical issues***

As the study necessarily required the use of a clinical population ethical approval had to be obtained from the North Sheffield Research Ethics Committee before the project could commence. In order to comply with ethical requirements all participants were provided with an approved information sheet regarding the study (see appendix i) and were asked to sign a consent form before taking part (see appendix ii). In the case of younger adolescents, usually those under fifteen years of age, parents were also asked to sign a separate consent form (see appendix iii).

The main ethical concern was that in view of the potential sensitivity of topics to be discussed the interview process may inadvertently identify children or adolescents who were in need of psychological help. It was therefore stressed that in this event the person concerned would be offered access to the clinical psychologists within Sheffield Children's Hospital, or where appropriate, he/she could be referred to local child and adolescent therapy services.

### ***Participants***

Children and adolescents who suffer from CF and were over the age of ten, but below the age of twenty and attended the CF clinic at Sheffield Children's Hospital, were invited to take part in the study. All participants were approached informally by the researcher, either whilst attending the hospital for routine clinic appointments or during a period of admission. This method of approach was suggested by the consultant responsible for the sufferer's medical care as it was felt to be the least intrusive and threatening option.

Potential participants, and where appropriate their parents, were approached, introduced briefly to the project, given the information sheet and the opportunity to ask any immediate questions. They were then either followed up a few days later, if admitted on the ward, or at their next clinic appointment, usually six weeks later. This delay in responding was intended to allow time for participants to consider their involvement carefully and to avoid pressurising anyone to take part.

### ***The sample***

A purposive sample of eleven sufferers, five females and six males, were interviewed. Participants ranged in age from 10 years 7 months to 18 years 6 months, the mean age being 14 years. The gender split is representative of the population under the care of the CF clinic at Sheffield Children's hospital and of the population of CF sufferers in Great Britain (CF Trust 1995).

All participants were in relatively good health at the time of interview. Participants were selected initially according to age and availability and a willingness to participate. Subsequently, further respondents were approached according to theoretical sampling procedures (Strauss and Corbin 1990). This basically involves sampling for further interviews as the study progresses and the analysis develops, so that issues apparent in the data but not yet fully conceptualised can be pursued. Over the course of this study this meant that individuals whose CF was at a variety of different stages were sought and also younger adolescents who were about to, or had recently undergone, the transition to secondary school were selected. This was in response to the emergence of these issues, as potentially important concepts, as data collection and analysis progressed.

### ***Data collection***

Data collection was problematic due to poor and haphazard attendance at clinic visits and a reluctance by people, particularly the middle teenagers, to devote still more time to their illness. Although the clinical team, including the consultant paediatricians, were very keen for the research to be carried out, physical health issues inevitably took precedence for both staff and patients. Interviews consequently were continually cancelled or rescheduled due to time waiting for medical procedures and consultation. Additionally, the actual pool of potential participants was in practice somewhat smaller than originally envisaged (30 as opposed to 46) for a variety of reasons.

Four of the twenty-six people approached stated a preference not to take part and several others were considered to have opted out indirectly as indicated by non-attendance or making themselves unavailable for interview. Nobody voluntarily telephoned to arrange to take part or to ask questions about the study.

### ***Interviews***

Five of the interviews took place whilst the participants were admitted in hospital for treatment of infections with intravenous antibiotics. Three took place at routine clinic visits and three were conducted in the sufferer's own home. All venues were dictated by the preference of the participants concerned. All interviews were conducted in privacy in the sole presence of the interviewer. It was made clear to each participant, and where appropriate his/her parents, that confidentiality would only be breached if the participant divulged information which suggested that he/she was at serious risk of harm or posed a risk to others.

The interviews lasted between fifty minutes and one and a half hours. Tape recordings of the interviews were made with the consent of the participants on the understanding that only the interviewer would listen to the tapes and that they would be erased once the project was completed. The tapes were transcribed verbatim to aid analysis of the data.

The content of the interviews and topics covered was guided by the use of a semi-structured interview schedule (see appendix iv). The topics covered included general feelings about living with CF and the meaning of the disease for the person; other people's perceptions of CF and their ways of relating to the sufferer; the treatment itself including motivation, intervention/support from significant others; perceived effects of treatment in the short and long term; specific coping with a difficult time related to the illness; clinic visits and the impact of outcomes upon treatment adherence; perceptions of the future as compared to life now and in the past.

As data collection progressed and the analytic process developed further questions were added. These focused upon the realisation that CF was specific to selected individuals, and the transition from primary to secondary school, as these emerged as potentially influential experiences in terms of treatment adherence.

Each interview began with the completion of the Family Sculpt Task (Gehring & Wyler (1986) followed by the drawing of The Bag of Feelings (Binney & Wright 1997). These are techniques designed specifically for use with children and teenagers to help elicit information regarding family relationships and inner feeling states respectively, in a relatively creative and non-threatening way.

The Family Sculpt Task involves presenting the child or adolescent with a wooden board divided into squares and a number of wooden figures, large ones to represent adults, small ones to represent children, and some wooden blocks of various heights. The participant is then asked to place the figures on the board so as 'to make a picture of his/her family'. He/she is told that the picture should show how close everyone feels to each other and that the blocks can be used to elevate the powerful people in the family.

The Bag of Feelings requires the participant, with the help of the researcher, to draw a picture of an imaginary bag which contains all his/her feelings. The participant is then asked to imagine that the bag contains all his/her feelings, to select a colour for each feeling, and fill in the proportion of the bag that is full of that feeling. Each feeling can then be discussed as the child draws, or on completion of the bag, depending upon the wishes of the participant. (see appendix v for an example of a completed bag).

It was hoped that the use of these techniques would engage the participants from the outset and encourage them to focus upon emotions and thoughts, as opposed to concrete behaviours, actions and facts.

These tasks were then followed with a series of open ended questions aimed at exploring the issues listed above. This kind of semi-structured format was used in order to allow flexibility for the researcher to follow lines of thought and conversation raised by the participants as relevant, whilst providing enough consistency to ensure that certain issues believed to be pertinent to the topic could be raised. This format facilitates analysis of the interviews which does justice both to the research questions and agenda of the researcher and to the preoccupations of the interviewees (Banister et al 1994).

## **Analytic procedure**

The transcripts were analysed using the Grounded Theory procedures described in Strauss & Corbin (1990). They describe three main types of coding known as open coding, axial coding and selective coding. Each of these procedures will be described separately. But, as Strauss and Corbin (1990) point out, in actually carrying out the procedures the researcher tends naturally to switch between one level of coding and another. This is particularly true of the processes of open and axial coding.

### ***Open coding***

Open coding is defined by Strauss & Corbin (1990) as "the process of breaking down, examining, conceptualising and categorising data" (pp. 61). This is initially done by the process of line by line coding. This involves labelling each line of data in terms of the phenomenon to which it refers (see appendix vi for section of coded transcript).

As this process is conducted, incidents within the data are continually compared so that those pertaining to similar phenomena can be given the same name or conceptual label. Once particular phenomena have been pulled out from the data the concepts which seem to be related to each particular phenomenon can be grouped around this to form a category. These categories can then be labelled in such a way as to remind the researcher as to which phenomena and concepts they pertain. The label should be on a higher level of abstraction than the concepts placed under it. These categories in themselves then become phenomena at a more abstract level. These are however considered to be provisional at this stage as they await verification through the process of axial coding.

The concepts can then be developed in terms of properties and dimensions. Properties being the characteristics or attributes pertaining to a category and dimensions referring to the location of a specific property along a dimension. For example the category normal versus different could have the following properties and dimensions:-

### General properties

### Dimensions

circumstances	allow total normality	maximise difference
realisation (for self, by others)	no awareness	awareness of difference
outcome	beneficial to be different	harmful
identity	CF integral to identity	pretend doesn't exist
wellbeing	very well greater normality	ill max. difference
prominence	stand out	blend in

Each time a category is present in the data it can then be located at the appropriate point along each dimensional continuum. Each occurrence of a general property will be located at a different point along the dimensional continuum. This leads to the building up of dimensional profiles for separate occurrences of a category which can be grouped to form patterns.

### ***Axial coding***

The process of axial coding involves restructuring the data following open coding, in a way so as to describe the relationships between categories and subcategories. This achieved by use of "the paradigm model" (Strauss & Corbin 1990, pp. 99-107). This involves denoting relationships of subcategories to categories in terms of causal conditions, phenomenon, context, intervening conditions, action/interactional strategies and consequences.

In its simplest form this essentially means the following:-

Phenomena are derived through open coding. The causal conditions, of which there is usually more than one, leads to the occurrence of the phenomenon. For example, deterioration and/or a severe infection leads to alarm in the sufferer. The properties of the causal condition then determine the dimensions of the phenomenon. For example, irreversible deterioration in lung function leads to intense alarm. Causal conditions and their properties are identified by concentrating upon a particular phenomenon and looking through the data for the things that precede it.

The context refers to the conditions under which certain properties and dimensions apply and certain actions will be taken in response to a phenomenon. For example,

under conditions where alarm is intense the sufferer may try to push this to the back of his/her mind, adhere more rigorously to treatment and attempt to regain control of the illness.

Intervening conditions are broader more general conditions which impact upon action interaction strategies such as culture, time, career, history, space, economic status individual characteristics and so on. For example, if a person with CF feels ill, but has a long way to travel to the hospital, he/she may decide to hedge his/her bets and wait to see if the condition improves. If however the individual lives close to the hospital he/she may decide to drop by, just to be on the safe side.

Action interaction strategies are the actions or interactions which occur, or are taken in response to, a phenomenon within particular contexts and under a specified set of conditions. These strategies are purported to be processual and to have a particular result or goal which may be intended or a result of reflection by the person or another.

Finally, consequences refer to the results of action / interaction or in some instances the failure of action / interaction in achieving a desirable outcome. This can be in terms of people, places or things, can be actual or potential and can occur in the present or the future.

For example, the CF sufferer who responds to the alarm experienced as a result of deterioration in lung function may, if he/she believes poor adherence to treatment to be a contributing factor, increase the level of adherence. If he/she once again acquires a serious infection, despite these actions, this would constitute the consequence of further deterioration, increased alarm, and despair at the benefits of treatment as a consequence of failed action interaction. The overriding consequence may then be resumed and continued poor adherence.

Throughout this process the researcher continues to look for further properties of categories and the dimensions of any newly identified properties.

### ***Selective coding***

This is the final stage of analysis and involves choosing a core category under which all the other categories can be related both to the core category and to each other. It is through this process that the final tight integration of the theory is

achieved. The relationships between categories are validated and any gaps in the categories and relationships are filled in through the process of writing and rewriting, as the researcher returns to the data to search for the answers to further questions as they arise.

### ***Enhancing theoretical sensitivity***

Theoretical sensitivity is, Strauss & Corbin (1990) emphasise, a crucial feature in building a good, conceptually dense grounded theory. They describe a range of techniques for enhancing theoretical sensitivity which can be used at all stages of the analysis. The techniques are aimed at broadening out the researcher's thinking in relation to the data and enabling an in depth analysis of what the data contains.

These techniques include the asking of questions, analysis of a word phrase or sentence, the flip-flop technique, systematic comparison of two or more phenomena, far out comparisons and waving the red flag.

The asking of questions is self explanatory and is geared towards the development of categories and their properties. The researcher is encouraged to ask questions such as who?, what? where? how much?, when?, why? and how?

Analysis of a word phrase or sentence simply involves brainstorming all the possible meanings or connotations of a particular word, phrase or sentence that the researcher feels may be of significance. For example the word 'normal' is important in the context of this project.

The flip flop technique basically involves turning a concept upside down and contemplating the opposite. This allows for comparisons to be made between the extremes of any one dimension. For example 'what it means to be normal' can be turned on its head to explore what it means to be 'different'.

Systematic comparison of two or more phenomena simply refers to choosing things and comparing them in lots of different ways. Strauss & Corbin (1990) give the example of comparing reactions to a slim woman with an overweight woman. Far out comparisons can also be made. These involve comparing two phenomena, which on the face of it may be totally unrelated, in order to generate further thought about the phenomena under study.

Finally, Strauss & Corbin describe 'waving the red flag'. This entails the researcher being extremely alert to assumptions in the data which are based on cultural perspectives. The researcher can find these in the data by keeping a close eye out for phrases such as 'never', 'its always been that way', 'everybody does the same thing'. Questions should then be asked as to why these things are apparently so definitive, enabling a fresh perspective to be taken in relation to the issue at stake.

### ***Memo writing***

The process of memo writing helps the researcher to keep track of all the products of coding. Memos provide a way of elaborating codes and of beginning to build relationships between categories. The process of writing memos helps the researcher to abstract from the data and go beyond the individual case to identify patterns, but at the same time to stay close to the data. Charmaz (1995) states that through the process of memo writing the researcher can achieve 'intimate familiarity' with the data. This, according to Loftland & Loftland (1994) cited in Charmaz (1995), is what constitutes the criteria for good qualitative research.

Memos can take the form of written notes to the researcher about leads to follow up on, questions or ideas about what the data analysed so far may mean and clear descriptions of categories and the associated properties and dimensions. They may be very concrete in the form of instructions to the researcher or highly theoretical. As the analytic process develops memos become more complex, clear and accurate. Memos are primarily a record for the researcher, though later memos may form part of the final write up of the research.

### ***Example memo***

#### ***Theoretical note re family responses/ expression of concern 16/3/97***

What is it that differentiates intrusive concern from a more containing kind of concern? Is it that intrusive concern does not allow for any positives so is more of a hopeless, tragic, overbearing kind of concern? How do people faced regularly with this kind of concern avoid or deal with it? Does this relate to hiding the fact they have CF from others more carefully? If they cannot escape this kind of concern does it lead to fear, sadness, resentment of illness, lifestyle? If so does this then have implications for treatment adherence, in what ways, under which circumstances?

### ***Interpretative validity***

Strauss & Corbin (1990) suggest the validity of a grounded theory lies in the rigor with which the procedures described are applied to the data. If these procedures are carried out with sufficient care they argue,

"the method meets the criteria for doing 'good science': significance, theory-observation compatibility, generalisability, reproducibility, precision, rigor and verification" (pp. 27).

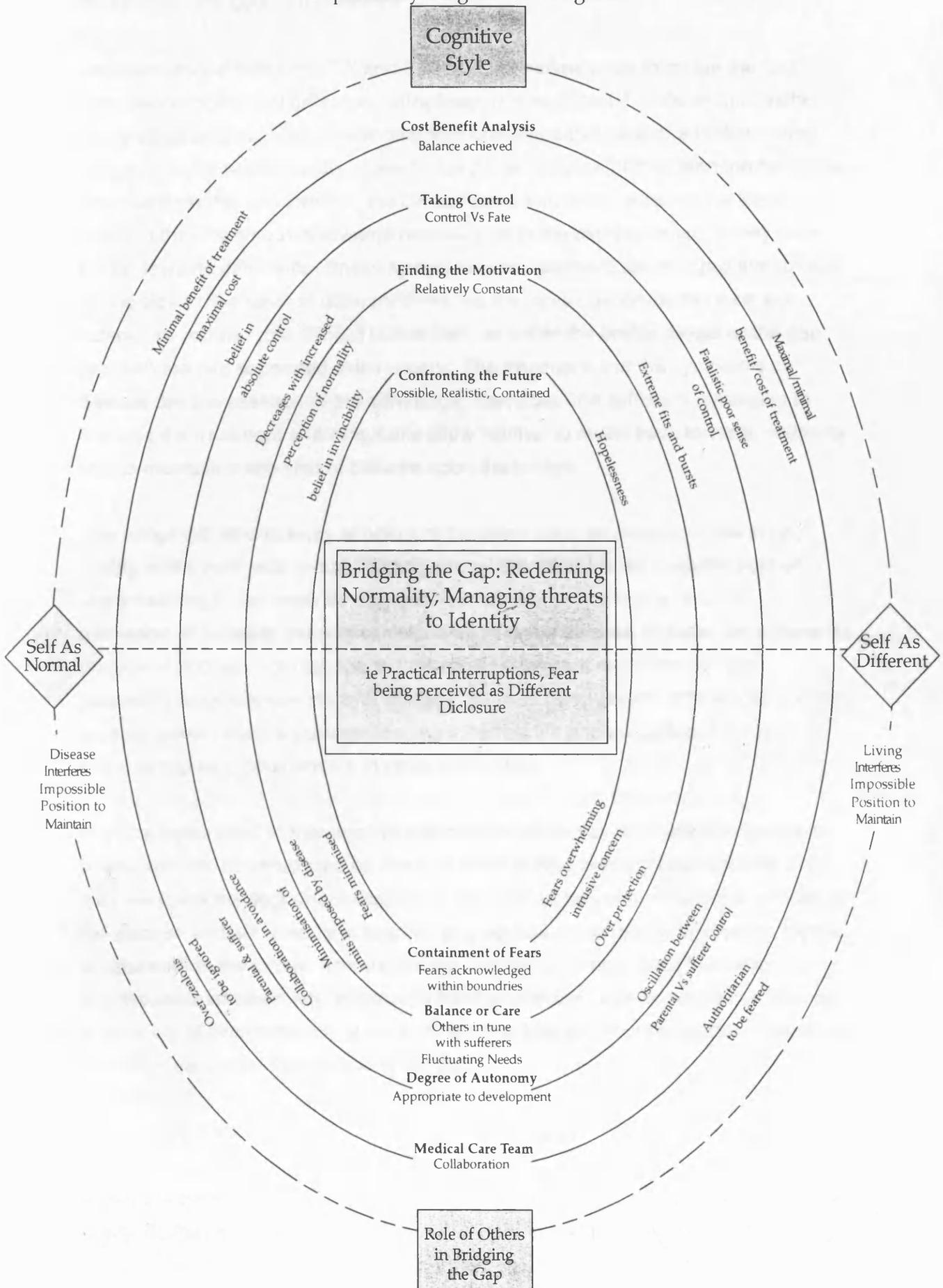
Throughout the process of analysis in this study, the researcher kept a check upon the extent to which the procedures laid out in Strauss & Corbin (1990) were being followed by the ongoing discussion of sections of coded data, at various stages of coding during a weekly grounded theory seminar.

## **ANALYSIS**

The outcome of the analysis will be described in terms of the particular story which seems to be being told by the sufferers who participated in the study. In accordance with the suggestion of Charmaz (1995) quotes will be used extensively to illustrate the different features of the story in order to emphasise the human perspective and make the conceptual analysis more accessible. To avoid confusion the term sufferer will be used throughout when referring to the person who has CF.

When quotes are used, the transcript from which they have been taken, labelled A-K, and the paragraph from which the particular quote has been extracted, will be indicated at the end of the quote using the appropriate letter and number. Where attention is being drawn to a particular developmental theme, and the age of the sufferer making the comments is felt to be important, this information will also be added, though the age will be denoted to the nearest year in order to preserve anonymity and confidentiality. Names have also been changed for this purpose.

# Explanatory Diagram of Categories



## **BRIDGING THE GAP: an overview**

The main task of living with CF and its treatment seems to be to bridge the gap between normality and difference effectively. It is as if the CF sufferer and his/her family could be visualised as standing with one foot either side of a broken swing bridge. In order to survive they have to keep their balance. If they lean too far in one direction then this can threaten the CF sufferer's existence, either in the literal sense, if they lean heavily towards normality, or in the psychic sense, if they lean too far towards difference. Illness symptoms and treatment serve to pull the sufferer to one side or the other at different times. As the illness becomes more severe balance becomes more difficult to maintain, as either the bridge sways or the gap between the two separated sides widens. The treatment and the symptoms of disease are the essence of the difference. Therefore, the sufferer's strategies to manage the treatment and symptoms allow him/her to revert back towards normality and to maintain some kind of balance upon the bridge.

The bridge will tend to sway at points of transition such as starting a new school, mixing with a new peer group, forming sexual relationships, or a severe bout of illness leading to the need for aggressive treatment or possibly a hospital admission. Increasing severity or deterioration of the disease requires the sufferer to adapt and find ways to manage the increased treatment requirements and potentially more obvious signs of disease, so as to minimise the differences and find an acceptable balance between leading a normal life and accepting the need for some recognition of difference in order to survive.

The strategies used to manage the gap can be learned by the familial response to illness and can be influenced by this and relationships with significant others. This may influence the degree of acceptance the sufferer has of him/herself in relation to the disease and will determine to some degree how precariously, or steadily, he/she is balanced on the bridge. The beliefs the sufferer develops about his/her illness and the value of treatment can provide him/her with the tools to maintain a balance in a variety of circumstances and can determine how far, how seriously or how often the individual places him/herself in danger.

The story of the child or teenage CF sufferer in terms of living with the disease and its treatment can therefore be told in terms of one core and two other main categories. The core category of 'bridging the gap between normality and difference' will be explained in terms of its subcategories, which pertain to the development and maintenance of the CF sufferers' identity in relation to the disease. The two main categories, namely 'the role of others in bridging the gap', and 'cognitive style' will then be explained in relation to this core theme.

## **BRIDGING THE GAP BETWEEN NORMALITY AND DIFFERENCE**

### **i) Redefining normality: identity in relation to CF**

- ii) Threats to identity:**
- a) *Practical interruptions to normality***
  - b) *Fear of being perceived/ treated as different***
  - c) *Hiding the signs: managing fear of discovery***
  - d) *Significance of the relationship in determining disclosure***

### **i) Redefining normality: identity in relation to CF**

In bridging the gap between normality and difference the CF sufferer is principally required to form and maintain an acceptable identity in relation to his/her CF, so as to keep his/her balance on the bridge and effectively span the divide. If the individual is to be successful and survive this necessitates maintaining a balance between normality as it suits him/her in view of CF, and normality as he/she believes it to be perceived by others who do not have CF.

When the CF sufferer accepts that CF is a part of him/herself and so is integral to the individual's identity, this leads to the likelihood of the treatment being accepted as part of day to day life. If a balance is achieved between CF being a part of identity, but not an overwhelming part, this seems to promote a routine attitude towards treatment. The sufferer seems to be able to acknowledge the negative side and be realistic about the limitations this may impose, but then take a proactive stance rather than dwelling upon what might have been. This then seems to enable the sufferer to let go, to a greater degree, of the things he/she cannot have control over, and take charge of the aspects of the disease over which some degree of control

can be taken. This seems to be a kind of normality within limits and could be conceptualised as constructive acceptance.

"Its a part of me .....it doesn't exactly tie me down and that so bad that ....because I think I live normally apart from having to do my physio .....yeah there's a bit of treatment involved but nothing that's going to hurt or is really bad so I'd say get on with it you're alright....." F16, 8

"Well its not nice.....but you've just got to try to get on with it ..... I do think what it would be like to be normal sometimes but there's not much.....there's no point because you're not going to be normal so you might as well get on with it" D9

The differences perceived in identity expression by the sufferer are minimal and not grossly different from the modes of identity expression he/she imagines using if he/she did not have CF. CF is not considered by the sufferer to impose extreme restrictions upon future aspirations, skills and achievements. Although some differences can be acknowledged these are rationalised, enabling the sufferer to form a balanced view in relation to aspects of his/her identity in terms of skills, hopes and dreams that are not necessarily related to CF.

"Its part of me.....I'm used to doing it so" F23

"I don't think it'd be that different .....apart from if you were the type who wanted to smoke and all this you'd probably do that but you don't when you've got this" F18

"I know CF limits some things like urm.....and I mean I don't dream about being a football player or anything because I'm not good at football.....and I don't .....some people want to be astronauts.....I don't dream about that.....because I've got .... I'm not so that's really limiting me ..... I don't like heights.....I'm interested in drama and I'm very interested in computers" I42

It seems that under these circumstances treatment can become so much a part of routine that the difference actually becomes the norm. The sufferer assumes his/her normality rather than a normality perceived to apply to everyone else. This then makes the missing of treatment seem odd, as it becomes so much part of routine, and so also serves to minimise the annoyance felt at the burden of treatment. The greater the perceived effort required for treatment and the greater the element of self reliance hinted at, the more likely this is to lead to a degree of non-adherence. However, a strong sense of normality in relation to this seems to allow the sufferer

to combat this and adhere relatively closely to the treatment requirements. This issue links to the degree of independence the sufferer has which will be discussed fully in *'the role of others in bridging the gap'* (subcategory; *degree of autonomy pp 58-60*).

"I've like almost trained myself to live with it ..... I think because it isn't anything that I've done ..... and you get half way through your life ..... Its been habit ever since I can remember" I30

"getting up at five thirty in the morning to do my D-nase that's a bit.....I don't feel like it ..... I don't mind my physio when I'm tired because that's relaxing ....but I don't like my PEP mask when I'm tired because I have to do that myself ..... there's not that much I mind about it because I've just got used to it as a way of life" D32

On the other hand two other options exist. When the sufferer's identity centres around CF and CF is perceived to impact upon every aspect of life, the perception is held that the person's identity expression would be drastically different if he/she did not have CF.

"(tongue tied) I'd probably be a mad raver ..... I'd probably be on drugs all the time or a womaniser ..... because I have got it I listen to a completely different type of music ..... don't approach women ..... don't take drugs ..... " E28-30

Under these conditions CF is perceived to have placed grave restrictions on a persons life, the treatment requirements being the core of these restrictions. This then serves to make the burden of treatment overwhelming and the fight to be 'normal' even stronger. The need to preserve a facade of normality is even greater so that treatment obstructs this quest for normality and is more likely to be missed, ignored, or forgotten about.

"I just don't take it, just forget about it and do what I want (laughs) that's why I end up in hospital" E59

The other extreme of the dimension is that the sufferer tries to pretend that CF is not part of his/her identity by avoiding the use of the label CF and not taking on board the full implications of the disease. The strategy of denial is used to maintain the desired identity of total normality and to try to avoid having to incorporate CF into the identity, except on a vague basis. This then has implications for the

sufferer's beliefs about the necessity and value of treatment which are discussed at length in the category '*cognitive style*' (pp 64-80).

"I just wanted to be like.....like other people as much as possible so....." G7

"I don't really think of myself as having CF ..... I don't think I do really.....I still don't think I do ..... its just I cough a lot and have physio.....not you know I'm not different.....urm.....I don't think of it as my lungs are diseased sort of thing I just think oh they're a bit crap" A13

If the sufferer is heavily relying on avoidance as a way of managing the disease and maintaining a normal identity this can have direct implications for treatment adherence. Awareness of the disease is so far removed that he/she may be more likely to forget aspects of treatment. It is as if the sufferer finds it extremely difficult to tolerate and manage the constant reminder of difference that the treatment provides, and the possible implications of this for the future.

"you just don't think about it really ..... I should do .... like tablets , I just go out and don't even think about it ..... maybe I should, maybe its just getting the little things ..... I don't think about it you only forget like if I had them in the car I'd still forget (nurse in clinic advised this prior to interview)" J7

This perception of total normality on the part of the sufferer can be helpful in terms of adhering to the treatment regime and for the sufferer's acceptance of the disease if he/she is unaware that treatment and CF is not part of everybody's life. However it is inevitable that there comes a time when the sufferer's cognitive development allows for the realisation that this is not a 'normal' part of everybody's life. This gives rise to the need for reappraisal of what the treatment means and questioning of behaviour that has until now been taken for granted. This could arise out of a reluctance by the parents to explain that they are different and take on board the reality of life with a child who has CF. (*see category 'role of others in bridging the gap', subcategory 'containment of fears' pp. 49-54*).

"I thought it was what everybody else did so it didn't bother me when I was younger..... not till I was about 7 or 8 actually, then I thought hey no-one else is doing this, what am I doing sat here taking these when nobody else does" B47

When this realisation of difference occurs it can lead to the response of missing treatment in order to maintain the facade of normality and minimise the perception

of difference. However, if the sufferer is satisfied with the difference and comfortable in the knowledge that the treatment is his/her normality, though it might not be anybody else's, then the treatment will be more likely to be continued as before.

"when I was in hospital ..... and my brothers came to visit me ..... but I never went to visit them ..... and they weren't doing stuff like I was" G44

"no ..... because I'd always done it so" G46

The sufferer's identity in terms of CF and the consequent established internal normality does not seem to be static. Adolescence introduces new dimensions, such as cognitive development and the capacity for abstract thought, to the formation of identity which pose particular challenges for the CF sufferer. The stability of his/her identity fluctuates to varying degrees and can be threatened by and influence a number of factors which will be described as further subcategories. The sufferer's ability to cope with these threats will impact upon treatment adherence in adolescence.

## **ii) Threats to identity**

### ***a) Practical interruptions to normality***

In the face of deterioration of the disease or a bout of severe illness the issue of CF as part of the identity may be forced, as the disease symptoms and the treatment required to manage the disease become more prominent and potentially more obvious to others. The limitations imposed by the disease on the sufferer's established normality in his/her day to day life are increased, so challenging the individual's beliefs about what he/she can and cannot do. Normality is more difficult to maintain as the number of instances where the disease imposes a difference increase. The sufferer, therefore, is constantly required to adapt to changing demands and find a balance between normality and difference which is acceptable to the individual. This may necessitate the sufferer reincorporating CF into his/her identity in some way.

"I suppose more like this year and last year I've thought about ..... I have got CF more than I used to ..... because its only just really started to affect me .....like say if I'm watching tele

and its an active holiday or something and I think that'd be cool but then I think oh but I can't do it.....I was quite mild and I was just normal really.....completely but now .....its just breathlessness really and that's just a complete arsehole because you can't do anything without thinking well would I get breathless if I did that" A13, 20

This can have a positive effect upon adherence. If the sufferer's physical state is so poor that it prevents him/her acting normally and without treatment the difference would be obvious to others, or he/she believes it would be obvious to others, this leads in an increase in motivation for treatment adherence in order to maintain an acceptable balance of normality. If this balance is disrupted it poses a threat to the sufferer's identity in the form of fear of being perceived or treated as different. The risks to normality imposed by missing treatment in these circumstances are perceived to be greater than those imposed by sticking to it. When the physical state is such that the sufferer can appear normal without adhering to the treatment and treatment would actually be considered to interfere more with normality than the odd display of a symptom, this makes adherence to an aspect or more of the treatment regime less likely.

"sometimes I'd go a day and just have one physio session because then I was clear all the time, practically anyway ..... and you know it didn't make any difference at all but now I couldn't miss a physio and go out and just act normally because I'd be really breathless and just like coughing all the time" A35

If the sufferer tries to maintain semblance of complete normality and the disease is dismissed and treatment forgotten or negated, this can be shattered by the imposition of the deteriorating physical state and in the extreme case, hospital admission. The sufferer carries on regardless, succumbs to the allure of a pretence of normality and can, as a consequence become seriously ill. In the absence of a balance between acting normally, but recognising the need for acknowledgement of some degree of difference there can be grave consequences for both physical and mental health.

"I went on holiday and didn't take any medication the whole week because I was with friends and we had like a flat of our own and we came back and I was like urgh and I stayed in hospital for two weeks ..... I was on the brink of packing it in" E76

The physical state imposes a necessary change in behaviour in terms of treatment once again to redress the balance of normality. However, once the physical state

improves, behaviour returns to normal for the sufferer, unless he/she is able to find ways to contain the anxiety and anger provoked by the enforced realisation of difference, which in many ways can be described under such circumstances as a rude awakening.

"I used to complain about it ..... go aahhaah but most of the time you just forget about it actually ..... forget about it most of the time but then there comes a point where you can't ignore it like when you're in hospital and you just start aahaaah and get really annoyed because there's nothing you can do about it ..... you just have to wait for it to go away again ..... and once its all gone you just go back to normal" E13

If the sufferer focuses heavily upon difference the quality of his/her life may be impaired by treatment demands, although they may have the result of a longer life. If however, at the other extreme, the focus is placed entirely upon assuming normality, quality may be enhanced in the short term, but the ultimate impact is detrimental to length and quality of life.

Fears of difference and the implications of the disease can be contained by drawing comparisons with others who do not have CF and are perceived to provide markers of normality.

"if its windy that'll take my breath away, and I think well, I don't know but I think normal people find that as well" A41

Worries can be rationalised by this process in a way which acknowledges normality as opposed to difference as far as is possible. Comparisons can be made with people who have had similar treatments, such as drips, though for different reasons, required physiotherapy, have been admitted to hospital at some point, or whose lives have been affected by illness in such a way that some similarities can be drawn.

"Even if I have this week off (school) I've only missed as much as the rest of my friends ..... because like Mark he had two weeks off because he had a swollen gland or something so I've only had as many days off as him anyway so I'm right pleased about that" K38

If the person is well and the disease is perceived to be relatively mild then normality is easier to assume.

"I think some people who are worse than me might be able to say the more care and things, I probably have a more normal life for someone with CF" I17

The treatment itself plays into this as it gets in the way of everyday life and can be perceived to be time consuming, inconvenient and a chore. Clearly as the disease worsens, and the level of treatment is by necessity more aggressive, this upsets the balance of normality to a greater degree, so increasing the effort required to adhere to the treatment regime. Similarly as the sufferer grows older, even if this does not happen as a function of deteriorating health, this can occur simply as a function of lifestyle changes as the young adolescent develops a social life and interests outside of the family. Clearly it is not unreasonable to suggest that when, as may well be the case, these two factors collide the strain upon maintaining a balance may become even greater.

"When I was younger I suppose it was easier but ..... easier to do ..... wasn't doing as much ..... wouldn't be doing as much in the day like going out ..... " C53

The issue seems to be about finding an acceptable degree of interruption to desired normality for the sufferer, which may make it difficult to incorporate a religious and meticulous following of the treatment regime. The process of how these decisions are made are discussed in '*cognitive style*', (subcategory '*cost/benefit analysis: active decision making*' pp.77-80).

"I don't think it'd be any different.....I don't think so ..... basically I do what I want anyway really ..... I still have physio obviously, I don't always take my creons but ..... " J16

Organisational issues can be considered further to interrupt normality when lack of resources are perceived to add to the burden of effort and treatment and increase the need for hospital admissions.

"being able to take pumps home and stuff..... because I wouldn't ..... because I could be at home now but because of pumps I've got to be in here ....." E106

There are occasions when it is beneficial to maximise the difference between self and other, or at least the practical restrictions imposed by the disease, in order to avoid taking part in an unwanted activity, to receive desired attention and pampering or to excuse impertinent behaviour.

"I suppose sometimes if I don't want to go to college I probably ..... I'll pretend not to feel very well so mum doesn't hassle me" G31

"I couldn't do outdoor PE if it is snowing and foggy (laughs) ..... I don't really like doing it then anyway so ..... it doesn't really make that much change because my mum and dad always try to make sure that I do everything that a normal person does" D23

" They'd probably punch me in" E43  
(referring to family if he didn't have CF)

At other times the difference is a source of frustration and regret when it dictates that the sufferer cannot carry out a desired activity.

"The doctors don't like you doing horse riding but I absolutely love animals .... I am allowed to go but not in the stable ..... and silly things you might like to do like working with animals..... I don't think I'll ever be able to do that but its something I'd love to do like having pets....." B20

The task once again, in terms of practical restrictions, is to find the balance between leading as normal a life as possible, so minimising the restrictions imposed by the disease, but at the same time not dismissing the importance of special consideration in certain circumstances. For example, it may be acceptable to go swimming when well, but the implications of going when currently symptomatic may be too severe to ignore. Problem solving can be an effective strategy to manage this balance.

"before we got the dog we had to borrow somebody else's dog, just for a couple of weeks to see if my chest was alright". D25

#### ***b) Fear of being perceived or treated as different***

The fear of being considered as different comes to the forefront at many points in the CF sufferer's life. These times principally relate to when issues of disclosure arise or when comments by others, unwittingly or otherwise, denote the perception of a difference. Disclosure could be construed as posing a threat to the stability of identity and the balance of normality achieved.

The most fundamental purpose of assuming as much normality as possible, in order to ensure the optimum quality of life, appears to be to avoid the perception of difference by others and so avoid the risk of rejection, stigmatisation, over protection or preferential treatment. This management of the fear of being seen as different seems to have many and varied implications for adherence to all aspects of the treatment regime but particularly those which are potentially more visible to others such as creons, oral and, to some extent, intravenous antibiotics.

When the CF sufferer is faced with what are considered to be stigmatising or judgmental comments by others who do not know the cause of the apparent difference, the sufferer may wish that other people knew the truth, or the real difference, in order to avoid confrontation with these kinds of hurtful assumptions. These kinds of comments generally arouse anger in the person concerned which can, to a degree, be tempered by taking the other's perspective or empathising with the lack of knowledge. These are nevertheless, unpleasant experiences for the sufferer, as the perception of difference is thrust unavoidably in their face.

"People who ..... you know think its really funny to say "oh that's a bit of a smokers cough".....and you think oh yeah that's funny ..... its just ignorance, they don't know that's not it but you just think piss off" A23

By maintaining an appearance of normality incidents such as these can be avoided as they relate, in these circumstances, to strangers or to those who do not know about the disease.

"so its just the breathlessness really ..... it wouldn't be a problem if I wasn't so concerned with that ..... the way I look and stuff like that.....you know my image" A23

If the appearance of normality is challenged by someone with knowledge of the disease, this arouses stronger feelings of anger. The comment or action poses a more serious threat to the sufferer's self identity as normal and basically like anyone else.

"its the doctors more like you know..... once this doctor came into me with all these students and they were looking at my hands and a student said 'oh I think her fingers are a little bit clubbed' ..... and he said a little bit ..... I'd say they were very clubbed actually and its just like you know insensitive like that ..... and emphasising the differences sort of thing.....that really pissed me off actually" A15

This kind of event may have ramifications for adherence as the sufferer may then feel compelled to deny the existence of a difference more strongly, so potentially leading to a negation of the need for rigorous treatment. Clearly this does not foster good doctor/ patient relationships either, the implications of which for adherence or self care will be discussed in terms of the role of others in bridging the gap.

***c) Hiding the signs: managing fear of discovery***

The desire to present a normal image can have a positive effect upon adherence if the treatment is considered to prevent the obvious signs of stigma or aids the presentation of the image the sufferer wishes to present to society. For example if closer adherence to treatment enabled a male CF sufferer to attain a desired more muscular physique.

"I've always wanted to weight train but you can't really weight train unless you've got the fat first and to get the fat you've got to ..... you need your lungs to be well so I'm going to get my lungs well first ..... that's me jogging and creons to get fatter ..... I'm tall I should be wider, that's about it really" J33

In contrast, the female desire to be thin can have negative and positive effects on adherence in concurrence, depending upon which aspects of the treatment regime allow the female sufferer to attain or maintain the physical image she wishes to present. For example a young woman who wishes to be slim may not adhere closely to the dietary recommendations but may take plenty of exercise.

"they might go oh you've lost weight and I'll say I'm going away so I'm going to get right toned and fit ..... I'll put on weight after that .....I try and get my weights done every night ..... to tone my body and I've got a bike trainer so I try to do that as often as I can ..... then I just have a low fat diet so I keep healthy...." F29-30

The person's image can be more obviously threatened by aspects of the treatment which may become visible. This may lead to skipping one or more aspects of treatment or the delaying of treatment until a later point in time. The immediate benefit of avoiding the possibility of standing out as different in some way and running the risk of rejection outweighs the longer term benefits of adhering to the treatment regime.

In these circumstances the sufferer may still do the treatment but will go to great lengths to hide it. This requires a considerable amount of effort and the more sure or fearful the sufferer is of a negative response the greater the lengths he/she will go to, to hide the treatment.

"taking the tablets at school wasn't a problem but it would have been a lot easier if I didn't have to ..... trying to do the magic trick and stick them in your mouth without anyone seeing" J25

The strategies the sufferer adopts to hide the treatment, such as concealing tablet taking by sneaking behind bike sheds, going to the toilet, or hiding them under serviettes, serve the purpose of bridging the gap between normality and difference. The sufferer maintains the appearance of normality but at the same time acknowledges the need for different behaviour.

If the sufferer cannot find an appropriate strategy to conceal treatment, or the perceived effort in doing this is too great, then the treatment is skipped. This can happen when the sufferer's credibility is likely to be threatened and if an attempt to hide the treatment is more likely to fail.

"There's no way I'm taking tablets in a pub ..... probably drop them on the floor anyway"  
J20

If the sufferer is able to take the view that others hold an attitude of indifference then this, in turn, allows him/her to feel less in need of hiding the treatment

"its only when I'm eating my dinner and they ask what are those and stuff so its people like that, they just basically don't know so they don't care really" I24

The fear of being recognised and treated as different and possibly rejected, along with the issue of who, when and how to tell, may also be particularly acute at points of transition, such as the transition to secondary school, or for older sufferers the transition to college. This may mean that new strategies to counteract fears of rejection, by avoiding others having knowledge, may have to be found, as the sufferer is likely to be forming new relationships. This situation can be assuaged to some degree if the sufferer is undergoing the transition amongst some close and well established friends who know about and accept the disease.

"There was Ben and Stephen and everyone from my class and my friends class ..... so they knew anyway" I23

Otherwise fears of rejection can become paramount and so the bridging of the gap between normality and difference requires more skill and assumes a higher profile within the sufferer's daily life. New rules may prevent the efficacy of established strategies to protect a normal identity, requiring the development of new ways of managing the fear and potential threats.

"Going up to Holmesfield because like its ..... you're trying to keep it secret all the time ..... say I'm on antibiotics and its dinner and we're not allowed to take drinks outside, you've got to have them in the dining hall ..... so I have them in front of people and my friend, well not a friend.....says what are they..... I'll say antibiotics and they say what do you need those for as if they're not and I'll think oh God ..... I don't want people thinking I'm on drugs or anything like that" B53

The dangers of concealing the disease can, under these conditions, become apparent by the misinformed comments of others. This can cause alarm in the sufferer as it challenges his or her position of avoidance, disrupting his/her balance upon the bridge.

"My Maths teacher the other day said she had a second cousin with CF and you don't usually live past 17 and things like that ..... then a friend goes up to another friend and says oh I feel right sorry for Katy she's going to die when she's 17..... which made me a bit upset really ..... I don't think she knew anybody had it in school so....." B4-5

If the sufferer lacks the strategies to manage fear of the potential consequences of the disease or rejection, should it occur, or fears rejection to the degree where they cannot tolerate any outward sign of difference whatsoever, he/she may feel almost as if the existence of even a minute degree of difference has to be denied in order for the individual's sense of self to be maintained. This may mean that openly carrying out aspects of the treatment regime is virtually impossible. Paradoxically, in the extreme, the will to preserve the self could lead to unnecessarily premature death.

"It didn't start until I got to secondary school, in the infants it didn't matter .....it was like and everyone knew and it was like OK.....I didn't mind sort of ..... I did mind taking my tablets

at lunch though ..... but apart from that it didn't matter until I got to secondary school .....  
when I met new people and I was like Noooope" E92

If the person cannot envisage worthwhile advantages and/or expects a negative response attempts to avoid disclosure and so possibly non-adherence to treatment or poor self care are made more likely. Physical care is compromised in order to preserve the desired self image and identity.

"I don't know I don't want people to start feeling sorry for me.....then again I don't want them  
to pick on me either" B9

However if it is perceived to be advantageous for someone to know, this can make disclosure more likely.

"I don't mind if they know but I won't just go up to someone and say I've got CF.....in some  
cases I don't think they have to know" D28

"I went on this Whitehall trip and ..hum..and everyone had 3 lives and you were allowed to  
step off a piece of apparatus, they were balancing ones and you were allowed to step off  
them 3 times and I couldn't do this one so my leader picked me up and carried me to the end  
of it (laughs) because he knew I had CF" D28

There are times when disclosure is perceived by the sufferer to be necessary and, in a sense, the potential disadvantages of not telling outweigh those of telling, or in order to achieve a certain outcome a disclosure has to be made. When the sufferer wishes to be able to adhere to treatment without judgements or wrong assumptions being made by peers, adherence is promoted and effort reduced as the necessity to hide treatment is negated. In order to be able to take this stance the sufferer needs to have the confidence that others will not reject, or that he/she can handle rejection should it occur, along with a strong conviction in the value of sticking rigidly to the treatment regime. The conditions which may give rise to the development and maintenance of such a strong conviction will be discussed in '*cognitive style*' (pp. 64-80).

"I tell people who have to know because I spend a lot of time with them, they're going to think  
she's taking a lot of drugs" F21

Further disclosure can be instigated by a forthcoming admission to hospital, or following a lengthy hospital admission, when the sufferer anticipates that others are going to be asking questions and there is a need to explain absence, for example to teachers. Under these circumstances the sufferer takes the view that he/she might as well come clean rather than having to lie or leave everyone guessing. The extremity of circumstance and the degree of perceived necessity to tell will vary from sufferer to sufferer and across circumstances. One dimension along which this may vary is the significance of the relationship to those to whom a disclosure is to be made.

#### ***d) Significance of the relationship in determining disclosure***

The intimacy and value of relationships can also influence the degree of fear of rejection, the likelihood and the perceived necessity of disclosure. In the younger adolescent or child the fears of rejection centre around more distant relationships with the fear of those on the outside of the sufferer's peer group finding out and using it as a weapon.

"well if I know them quite well ..... I'll let them know but if say like people in other classes, like higher classes ..... I wouldn't let them know because they might ..... because they are bigger they might start calling me names and that". H25 (age 11)

Close peers are regarded first and foremost as a potential source of support rather than a threat, making disclosure within the immediate peer group easier and more likely. This, in turn, will reduce the effort required to follow the treatment and probably aid adherence.

"because they'll help me a bit more" H26 (age 11)

If the younger adolescent sufferer does not feel able to use his/her peer group as a source of support, and is keener to avoid the issue of CF and its treatment, this can lead to extra suffering and anguish.

"it gets you down, like if you're sat in Burger King and having to take your tablets, with your friends and they're all sat like that (pulls confused looking face).....they don't know what to do with themselves sometimes, I think they feel right awkward.....I don't want people to feel awkward around me as if they shouldn't be there....." B15 (age 13)

Issues of disclosure, within the sufferer's immediate social circle, can be handled by the parents who usually know the sufferer's friends and their parents, so providing some degree of safety and protecting the sufferer to some degree from misunderstandings. As the sufferer grows up and develops more of a separate identity from that of his/her family issues of disclosure, and by definition the fear of rejection, must be handled more often by, and at the discretion of, the sufferer.

"my mum knows them anyway ..... their parents she knows so she's probably told them anyway, there's not usually a lot of times when I tell them" K27 (age 12)

If disclosure is made at a distance as is usually the case, and the issue is handled by parents, this avoids the sufferer having to explain and potentially feeling rejected. The perception of rejection can result either from a misunderstanding in the process of the explanation or by the reaction of the person to whom a disclosure has been made anyway. By disclosing at a distance any embarrassment revealed by the person can be avoided. In fact, the whole issue can be avoided, if those concerned so desire, thereby minimising the perception of difference, as any differences are not brought into the forefront of the situation.

"I'd prefer if it was written in a letter or something because I don't like talking to someone"  
D28 (age 14)

"once somebody's said, it you know the friend doesn't really know a lot about it so you end up having to explain to about six people at a time about it all (pulls a face) ..... I don't mind really it just gets a bit embarrassing and....." K27 (age 12)

In contrast, the older adolescent perceives potential battles to lie within the peer group or amongst friends. In particular the issue of intimate and sexual relationships seems to increase the stakes, with the formation of new relationships particularly arousing anxiety.

"Like if I've got a new boyfriend and he's not the type you can tell ..... its just embarrassing"  
F14 (age 18)

Increased anxiety seems to arise out of the fact that within an intimate relationship visible evidence of difference, or markers of the disease and its treatment, are much more difficult, if not impossible to hide. Aspects of physical evidence, such as a porto-catheter, within the context of a sexual relationship, may be anxiety provoking,

as they provide irremovable and inconcealable evidence of difference and so have more stigma attached to them. By contrast, for the younger sufferer a porto-catheter may be preferable to more visible needles or long lines, as they can be concealed under clothing. For the older sufferer the stigma may be perceived to be such that, even in the face of acceptance by the significant other, the individual can still feel repulsion at him/herself. The porto-catheter, in this context, being an outward reminder of difference, an 'in your face' reminder, that cannot be ignored.

"it's easier if you have it in your chest because you'll still be able to write and go to school and things and no-one can see it there and nobody asks you about it" K43 (age 12)

"I just told her I had a portocath as well and she said oh she's seen worse.....I thought like what!" E16 (age 17)

In the face of revealing the disease and in the context of an important and intimate relationship, when the CF sufferer is not sure of the other person, the sufferer may handle the fear of rejection by anticipating it and trying to preempt the other's response making it easy for the other person to reject him/her. The consequence of the sufferer being accepted, is one of relief, as the significant other has not rejected, despite being given the easy option to do so.

"I said I don't mind if you don't want to see me again because I'll understand why and she said don't be so stupid ..... and I was like ooh" E18

Similarly, in the anticipation of unwanted preferential treatment, protection or sympathy, the person may include warnings against this in the way in which they tell other people about their illness.

"I don't want sympathy, I don't want anyone to even ..... I just tell them and that's it, I don't want anyone .....are you alright, I'll buy you a drink..... I don't want anything ..... I'm not bothered about sympathy, I'm not a charity case ..... there's nothing wrong with me ..... so I just tell the people who are important in my life" F21

If the reaction to disclosure within a similarly valued and intimate relationship is negative this serves to heighten fears of rejection and the desire to conceal the disease. However, if such a reaction is then resolved, within the context of a caring and significant relationship, the sufferer comes to expect that disclosure will ultimately be beneficial, in that it will eventually lead to a caring attitude and a

greater degree of openness. Again the greater the degree of openness, the more likely it is that the sufferer will be able to find the motivation to look after him/herself well and adhere more closely to the treatment regime.

Under conditions where the disclosure occurs without the initial knowledge or consent of the sufferer, fears of rejection, before the full extent of the other's reaction is known, can only be managed reactively, rather than proactively. The degree of control over disclosure can vary. The sufferer may choose to tell as discussed above. At other times the sufferer may find that another person has already been told by someone else, or that he/she just knew, but it is not clear to the sufferer how he/she came to know. This can be very disconcerting and can sometimes lead to further disclosure to avoid upset by a significant other hearing 'on the grapevine', as opposed to directly from the sufferer.

The sufferer may feel that this is unavoidable and become annoyed at the repeated interruption to the pretence of normality and threat to his/her identity. The individual may expect rejection and so preempt it, but if the relationship is important he/she may try to discuss the issue. This allows the sufferer to regain control by making sure he/she tells the story and conveys the issues as he/she sees them. This serves to avoid, as far as possible, the influence of stigmatising comments. If the sufferer has experienced disclosure positively in the past he/she is more likely to embrace this possibility as opposed to running away. The explanation in the sufferer's terms allows the individual to normalise, as far as possible, the meaning of the disease.

If the sufferer, however, still harbours fears of possible comeback, or wishes to try to preserve normality as it was before the other person knew, he/she may avoid discussing the issues. This may avoid the possibility of a negative reaction, or maybe even actual rejection, and avoid the need to confront negative feelings, allowing things to continue as normal on the surface. A positive reaction is therefore assumed.

"I just started going out with her and she knew ..... we haven't sat down and talked about it or anything but I think its o.k..... I think she's fine she never brings it up or anything" J22

Alternatively, or as a consequence of this, difference can be enforced in the sufferer's social life by others finding out about the CF without this necessarily being the sufferer's intention. This prevents the same facade of normality from being maintained.

"so its like I can't have a normal relationship because that always comes up.....everything I do that always comes into the situation" E16

In the face of actual rejection, or if a disclosure is about to be made and rejection is anticipated, this fear can be managed by the belief that negative responses are the fault of the person to whom the disclosure has been made, rather than any failing or lack of value in the sufferer.

"Generally I think they take it alright if they are the right people" F21

Such a belief can, in part, be arrived at by the repeated experience of positive consequences to disclosure.

"I don't usually try to hide it unless ..... its unless I know they are going to cause me trouble over it which I don't think I've ever had to deal with" I20

If the sufferer discloses and is treated normally, indicating the difference is accepted, this paradoxically allows the person to feel normal. The sufferer may actively influence this by acting normally, in the way he/she did before the disclosure, thereby making it more difficult for others to treat the sufferer differently and allowing others to habituate to the knowledge, as the differences are not constantly being played out.

"When a few more people found out they were like oh I'll buy you dinner .....are you alright.....and I thought you're just doing it because you've found out, if I didn't have this you wouldn't be offering me this or that but then they get used to it. They forget about it and so do I ..... its not like I'm a charity case and I'm poorly they just think arrgh she can cope, I act like I'm not bothered so they don't normally go out of their way" F22

When the sufferer has been on the receiving end of prejudice or rejection, or is for whatever reason preoccupied with this, the sufferer seems to hold the belief that living with CF will become easier as he/she grows older, thus anticipating that others will understand the predicament faced more empathetically. The focus seems to be upon being accepted and considered as normal and of equal value to the next person, rather than upon physical deterioration. The concept of being a human being first and foremost takes over from continued health, or at least assumes equal status. This is something that becomes even more valued in the face of inevitable

deterioration in health. This is also something which has potential for change, whereas ever declining health for more effort may be perceived to be less within the confines of control of the sufferer and is clearly ultimately beyond the confines of the control of humanity.

"I think a lot more people understand as you get older ..... they always say children are cruel aren't they" B50

"It might be easier .....I'll be able to talk more easily and people will be more mature ..... so if I tell them they won't be like oorh .....well hopefully not and if they are well I won't think they're are very old or whatever..... they'll be a bit stupid" E93

## **THE ROLE OF OTHERS IN BRIDGING THE GAP**

The strategies the CF sufferer uses to manage the gap are inevitably, to a large extent, learned from the familial response to his/her illness. Relationships with family may influence significantly the view the child or teenager has of him/herself in relation to the CF. The quality and nature of the friendships developed, will therefore determine, to some degree, how precariously or steadily the sufferer is balanced on the bridge. The responses of the medical care team and the interaction of these responses with family dynamics will also influence this position and potentially adherence.

The role of others in bridging the gap will be described in terms of the following subcategories:-

- i) Containment of fears**
- ii) Balance of care**
- iii) Degree of autonomy**
- iv) Medical care team**

### **i) Containment of fears**

One of the most important factors in helping the sufferer to manage his/her illness and its treatment and so maintain the balance between normality and difference seems to be the way in which the sufferer's fears are responded to and concern is expressed by the family. This may be a function of the family's, perhaps particularly the parents, containment of their own fears and their communication of that sense of security to the sufferer. When the family is actively involved in the sufferer's illness and its treatment this seems to allow the sufferer to derive comfort and a sense of well being from the family.

"well ..... when I see my sister and things like that ..... when I see my family" K7

(In response to question about what things make him feel happy)

The family can demonstrate this care and a proactive response to the illness in the practical sense by supportively helping the sufferer with the treatment regime, in some cases by actually becoming involved with charity events and fund-raising for the CF Society.

"My grandparents on both sides they always help me with things because they do things for CF and that ..... they'll help with jumble sales, on my mum's side and they go to dances to raise money for CF and things like that" H1

" Like well they always help me do my physio and encourage me and they look after me" H2

When the family demonstrates this kind of collaboration in the treatment or recognition of the disease, the sufferer seems to learn to value him/herself and feel that the family has invested a lot in him/her. The individual is then able to take on the role of looking after him/herself. The family accepts the need to adapt to the difference, embrace it and turn it into something constructive, rather than try to continue as normal or respond to it as a disaster. The sufferer's treatment and disease assumes a high, but contained, priority and the parent is seen as encouraging the sufferer to carry on even though he/she may feel like skipping some aspect of the treatment regime.

"you might not do it as much but my mum is always get it done, get it done ..... so we'll say yeah we can't be bothered so ..... like to look after us is her main priority, to feed us, cloth to look after us but sometimes I can't be bothered ..... but generally I'm good I don't miss a lot it's just like that ..... I think oh just get it done" F23

Under these conditions the sufferer seems secure in the knowledge of the family's support in times of need and comfortable in allowing the parents to carry some of the burden of the disease.

"I'll put her a few squares away (sister) because she's not really close but you know if there's an emergency and things" I1

"family yeah, my mum wouldn't have to bother going to meetings, she does a lot of fund-raising, jumble sales stuff like that, they'd probably be lost like 'ooh we've got nothing to do with the hospital ..... they'd probably think its a lot more off their minds than it is ours" F19

When fear or discontent is expressed by the sufferer, either through open communication or by acting out and refusing treatment, the parent is able to hold the fears for the sufferer by offering realistic reassurance and accepting the limits imposed by the circumstances. This conveys the message that there is no cause for alarm and that the situation is not going to escalate out of control.

"Well my mum is quite good because she says stuff like 'well next time you come in you won't have much to worry about will you because you'll probably be able to get better.....and if that's the best you can do that's the best you can do'....." D57

The sufferer is then able to appreciate the parents' concern and feel secure in the knowledge of their support without feeling overwhelmed by it. The parents put a boundary around the sufferer's fears in a proactive way, encouraging him/her to take control over the situation within realistic and honest limits, as opposed to wallowing in self pity.

"obviously I know that my mum and dad really do worry about me and stuff and they say that they are because I can tell when they are worried or not" D26

Within this context the peer group of the sufferer is still important as an alternative source of support, but the reliance for comfort and reassurance is still actively provided by the family, particularly at times of distress.

"I'm a bit low at the moment because my chest is bad ..... I've not got to get higher blows its just that I'm in for about a week ..... I want to put down that I love my mum dad and sister" D5 (whilst completing bag of feelings, interview conducted during hospital admission)

"my mum, dad and sister they try and understand but some of my friends like Susie who I talked about before, ..... she doesn't seem to care because she's not phoned up at all while I've been poorly ..... and my other friend Jessica she knows that I collect cacti and she's phoned me up every day and told me that she loves me and stuff so...." D16

When concern is expressed in a much more sympathetic and intrusive, as opposed to empathic manner by the parents or any significant other, this seems to convey the message that there is something really terrible to worry about. This emphasises the need to think and behave differently. The extra burden upon the sufferer and/or family leads to fears being exacerbated rather than contained, although the intention is obviously an expression of care.

"Like my uncle he always says he feels right sorry for me ..... some of my aunts they look at me when I'm taking my tablets and think ooh poor thing ..... but I don't want people to think like that" B12

Concern can also be expressed intrusively through favouritism. This again serves the purpose of the sufferer being singled out as different in a way which is not actually necessary for the individual's continued health and survival, again thrusting the issue of difference and the significance of the disease in the sufferer's face.

"one aunty in particular she treats me like I'm the favourite sort of thing ..... and sometimes I find that a bit annoying because its like she sort of ..... well it seems to me like she's making up for time she might lose if I die early you see" A29

Pressure to adhere to treatment can also be construed in this way and actually results in the sufferer feeling angry; a strategy aimed at pushing the intrusive person away so as to maintain the sufferer's feeling of and belief in his/her normality.

"I mean I know my sister doesn't mean it but she's on at me all the time and it gets on my nerves, sometimes I feel like telling her to shut up because it gets me mad, bossing me around" B18

Concern can be experienced as particularly intrusive when it occurs in the face of deterioration in the disease, and is expressed by a significant other who has usually avoided the issue of the disease and has not been overly involved in the treatment. The concern intrudes upon the sufferer's ability to deny the seriousness of the condition, so forcing realisation of the implications of the progression of the disease and challenging his/her perception of normality. This arouses alarm in the sufferer and reassurance given is recognised as being false. The fear aroused can be defended against by the expression of anger at the intrusive person. This may have detrimental effects on treatment adherence. It may lead the sufferer to avoid treatment as part of the avoidance of the seriousness of their condition, in the ways explained in '*bridging the gap between normality and difference*' (pp. 28-48). Alternatively, it may lead to increased adherence in order to avoid the feared consequence. Further implications of avoidance are also discussed in terms of the sufferer's cognitive style in relation to CF (pp. 64-80) These are inevitably influenced by the cognitive style of the sufferer's parents and other significant caregivers.

"Dad he's being a bit of a git at the minute ..... I think he tries to forget that I've got it anyway ..... I don't know, I think he finds it pretty hard to deal with anyway ..... urm I think really its just dawned on him that I'm as bad as I am ..... I don't think he thought ..... because he wasn't really very involved when I was little because he was always quite busy so I think its shocked him that I've got this bad ..... so that's why he sort of ..... it seems very

false to me that he's like he's really concerned all the time but I don't think its false its just because I'm not used to it because he's always put on this pretence that I'm o.k..... I think anyway I don't know....." A6 & 28

In the extreme, where fears are very difficult to contain, the sufferer feels that he/she is the reason for the family remaining as one unit and the whole structure is organised around CF. This could be conceptualised as being so intrusive as to be overwhelming for the sufferer, making the drive for normality much stronger and the will to adhere to treatment weaker. This relates to the formation of an identity for the person which is defined largely in terms of CF (*see 'redefining normality: indentivity in relation to CF' pp. 29-33*). If the sufferer does not adhere to treatment then this may make him/her more ill, so keeping the family closer together as there is more need to worry.

"me sort of thing ..... if I didn't have CF they probably wouldn't have a reason to stay together ..... looking after me ..... worrying" E35

When fears are not well contained the sufferer may also feel a burden to his/her parents. He/she may focus on the things that the parent has missed out on in life in a way which seems more filled with regret than would be the case if fears have been well contained and a proactive stance has been taken to CF and its treatment.

" urm.....I think my mum would've got a job a long time ago as well.....well for the last couple of years she has done but before that she didn't because of looking after me and all the treatment" G12-13

" I feel sorry for mum and dad because they don't get normal life" B2

Under conditions where fears are not contained adequately at home the sufferer may seek support and containment from peers, as opposed to the family, as he/she feels that an unbearable burden is being placed upon his/her parents. This can be particularly true in times of crisis when, rather than seeing the family as a support, the strain may dictate that fears are less able to be contained, tension is high and parents are less available, promoting an existential crisis in the sufferer. Accordingly the situation is managed through self pity and a heavier reliance on peers for support.

"last year when my grandma died I think God I've got that on top as well and CF to cope with as well ..... I sit and feel right mad because I think that's happened and I've got CF on top of it or I fall out with my mum and things like that ..... it makes me feel a hundred times worse.....laid on my bed and cried and all sorts was going through my mind like why did I have to get it, it sounds silly because I wouldn't wish it on anybody else but why me"

B35-37

"I just go and phone some of my friends and see if they want to go out or come round or something because if I talk to my friends I seem to come round a lot quicker because they tell me their problems as well and it takes my mind off what I've got..... " B38

In contrast, containing encouragement promotes an attitude of taking control over the illness and enables the sufferer to accept, within limits, CF as a part of his/her identity and to form a realistic attitude towards which aspects he/she can take control over. This enables the individual to be positive whilst acknowledging the negatives, as he/she has a secure base to fall back on at times of dread.

## **ii) Balance of Care**

The balance of care really pertains to the role of others in enabling the CF sufferer to bridge the gap between normality and difference through their actions. For the CF sufferer relationships with significant others contain a tension. The sufferer wishes people to demonstrate that they care and realise some degree of difference or specialness, but at the same time wishes to be treated as normal and not be overprotected. There is a desire for others to be in tune with needs for empathy and achieve a balance in demonstrating care.

"people worry but you know they just see me as a normal girl so ..... I suppose its a bit better like that because I wouldn't want them to think that I need a wheelchair and stuff ..... wrapped up in cotton wool and stuff " D26

When the disease imposes a restriction upon the activity and behaviour of the sufferer, and if others are too aware and try to look after him/her, this is perceived as being overprotective as it draws attention to the difference between the sufferer and the other. If, however, the other forgets about the CF and treats the sufferer as completely normal, but the sufferer cannot act completely normally due to physical limitations, this emphasises the difference to the sufferer and leads to annoyance at the perceived lack of empathy on the part of the other. The sufferer in these

circumstances desires the other to find the correct balance of care and to corroborate in treating him/her as normal, minimising the difference. In a sense, when the disease mitigates against normal behaviour, the gap can be bridged and the sufferer's perception of normality maintained by the careful consideration and subtle allowances of others who do not behave in ways which emphasise the differences.

"My sister I expect her to think about it and think oh yeah Becky can't do this and this but then if we're walking up the hill quite fast and I'd be like oh come on Kerry slow down and she'd be like come on and I'd be like oh God I can't breathe ..... and then you think oh I wish she understood a bit but....." A14

Empathy is particularly desired in terms of the extra demands of treatment for the sufferer. If the gravity of this is denied the sufferer may feel judged and misunderstood, and the treatment becomes even more of a bind and an unbearable difference. This may be particularly true if fears are not that well contained. If, on the other hand, the sufferer feels that others empathise with him/her this seems to enable the individual to take a balanced perspective regarding the opinions of others.

"they think that its something that crops up now and again like asthma ..... but I don't think they know enough about it to understand all the treatment and everything" D30 (referring to teachers and peers)

"some people don't understand so you just think well ..... they treat you as if you should just do it like eat more to put on weight ..... but its harder than just saying it ..... " G62

"no like my sister, sometimes it upsets me because she's always telling me and I think if you knew what it was like having to take tablets all the time, every time you eat..... she says she wouldn't mind if she had to, to look after herself but she would if it was all the time..." B15

If the difference is acknowledged within limits and accepted, again the sufferer paradoxically feels normal. When the difference is ignored this can allow a pretence of normality, but sooner or later the disease is going to intrude upon this, causing a threat to the sufferer's balance of normality and his/her identity. This affects treatment as it epitomises the difference. Any aspects of it which are not going to redress the balance of normality are missed at least occasionally. This potentially

places the sufferer in danger of deterioration and so of losing his/her balance on the bridge.

When the sufferer has a balance in terms of normality and difference he/she is able to accept that others have limits to their understanding and perceives that they try to understand as much as possible. This does not appear to be a function of chronological age, but rather relates to the sufferer's style of interrelating with the family and the disease. Younger sufferers simply express these issues in more concrete ways.

When fears are contained, this seems to enable the sufferer to feel that others empathise, or at least enables the sufferer to appreciate more strongly the difficulty others may have in achieving the desired degree of empathy. This may be because the people around him/her are more empathic and less intrusive, enabling the sufferer to contain the CF in some way. He/she feels secure in the knowledge that others care and is able to accept an inevitable degree of different treatment from others, without feeling threatened. The responses of others are therefore less likely to provoke anger and rebellion which may be detrimental for treatment adherence and self care.

" They do understand up to a certain extent but they don't understand and know what its like ..... but they try their hardest so I can't complain really can I " D16 (age 14)

The sufferer seems to obtain a sense of security from his/her friendships and perceives a positive balance between careful consideration and disregard for their CF.

"Yeah I think Matthew is understanding because he ..... I've got something in common with each of my friends I think ..... because like that's like Matthew and me we're very alike if you see what I mean and urm ..... he's urm .....he's not always into ..... he likes having a laugh with people but when someone's not very well or something or something's wrong with them he's understanding about that so he ..... he doesn't just go straight for picking on them or anything he actually thinks about what he says" I12-13 (age

12)

Other sufferers seem less committed to the idea of other people attempting to understand and are apparently unable to give concrete examples of others

attempting to understand their predicament, or the examples given are perceived to convey a serious lack of understanding.

" ..... urm .....apart from other people with CF ..... I suppose people try and understand or they understand ..... some people probably do ..... " G4 (age 17)

"urm ..... not sure really .....not really sure ..... I think some people might do but....." C21 (age 16)

"No..... like my sister, sometimes it upsets me because she's always telling me and I think if you knew what it was like, having to take tablets all the time, every time you eat .... she says she wouldn't mind if she had to, to look after herself but she would if it was all the time" B15 (age 13)

In contrast, some sufferers believe that people don't know but are accepting of this and are able to give a coherent explanation of why they don't know. This takes on board the reality that they are not completely normal but neither are they 'freaks', so conveying a balanced opinion.

"sometimes when my lungs are clogged up I just think well I don't know if they can imagine ..... but I don't know if they can imagine themselves because I don't know if we get any feeling ..... you know if I cough they might think oh yeah like asthma but I don't think they really do ..... you feel alright unless your lungs aren't that good ..... they probably don't know" F15 (age 18)

When the sufferer's life is consumed by CF he/she takes the view that others cannot possibly understand, because of the gravity of difference, and convey feelings of huge difference, isolation and insecurity.

" I don't think so ..... unless you ..... they have a day in the life of ..... well unless someone came and took away one of their lungs or something ..... then they'd suddenly realise and go oh dear" E18 (age 17)

" When I get tired and ..... stuff on my mind all the time and why I can't just talk to people like everyone else does ..... and communicate with people like everyone else does..... just go and approach people ..... " E19 (age 17)

The perception is one of difference, the focus is placed on the things that stand out as being away from the norm rather than similar to everyone else, provoking anxiety and a greater desire to be normal.

" They don't know what to do with themselves sometimes I think they feel right awkward ..... I don't want people to feel awkward around me as if they shouldn't be there" B15  
(age 13)

### iii) Degree of autonomy

This category relates to how the degree of autonomy that the child or teenage sufferer is permitted to have has implications for self care and adherence to treatment. Parents are seen as the main influence in developing patterns of adherence. Responsibility is then transferred to the sufferer as he/she becomes older and begins to understand more about the disease. If the sufferer believes in his/her ability to care for him/herself this is seen as a positive shift and promotes continued adherence.

"it was a pain when you were younger because you're doing this and this but you don't really know, even though I understood it from being quite young its probably easier now because you can do more yourself, look after yourself better ..... when you're younger its up to your mum and if she doesn't look after you, you've had it haven't you because you're too young to really know what's going on" F32

In circumstances where the sufferer has a greater degree of autonomy and the parents fail to take charge in ensuring that the treatment regime is adhered to at an early age, this leads to reduced adherence, despite the fact that the parents may be very concerned and protective towards the sufferer. The parent and sufferer effectively collude with denying the necessity of carrying out the treatment rigorously, avoiding taking on board the full potential implications of the CF.

"I suppose it'd be between about 7 and 10, something like that ..... and I didn't like physio and I didn't think I needed it and mum'd be like come on lets do your physio and I'd be like no ..... mum always had a battle on her hands ..... and she just got fed up of it and would say right fine do what you want ..... and she ...well she was worried and everything but it didn't make a lot of difference" A43

"they leave it up to me" C20

If parents are seen as being in charge and they collaborate in ensuring adherence this leads to increased likelihood of the importance and necessity of treatment being internalised by the sufferer. The parents taking charge, but allowing for some degree of negotiation, makes the sufferer feel he/she would still carry out the treatment without encouragement or a reminder from the parents. An organised but flexible routine facilitates this.

"My parents aren't pushy but they make me do it and I think that's right" K33

"well my dad usually says about ..... he'll come up about eight or something like that and he'll come up at quarter to or something..... and I get mad with him (laughs), he's usually alright with the time but the last couple of nights he's come up quarter of an hour early so I've told him to go back downstairs ..... and I just can't be bothered to start ..... so I suppose it is my dad more than anybody but I mean ..... I don't really need anyone to tell me I mean I know I've got to start at a certain time" K21

A heavier handed, authoritarian approach can also have a positive effect upon adherence and instil the importance of the regime but runs the risk of rebellion when the sufferer starts to assert his/her autonomy, so backfiring into reduced adherence. In order for the sufferer to attain independence the parent has to relinquish control at some point. The greater the degree of control exerted the more difficult this is to relinquish. The sufferer therefore has to make more effort to escape the control. Faced with reduced treatment adherence parents work more intensely to maintain their control. Importantly, if treatment is adhered to under these conditions, it is carried out as a means of 'getting them off the child's back', rather than out of a sense of need or an internalised belief in its value and importance.

"If I do miss it it's not usually missed altogether its just delayed .....I usually do it so she'll stop hassling me....dad he doesn't say a lot but when he does I tend to do what he says"

G25-27

"I know my parents always used to ..... I can't say they shouted at me it sounds like they were abusing me (laughs) but they did ..... it does though...I don't know they used to but now I shout back ..... I used to hate it...but now...I can understand why ....its only basically my mum she insisted on it so....." J12-13

" basically I do what I want anyway really ..... I still have physio obviously....I don't always take my creons but urm ..... I don't know mum she'll see I've had my dinner and she'll say have you taken your tablets and it gets right on my nerves" J16

The value of treatment is instilled in terms of the negative consequences of missing it, rather than more deeply in terms of value and care for the self and the positive aspects of carrying it out. It is internalised on the basis of fear. The sufferer, therefore, is more likely to have to deny and avoid the feared consequences of the disease to assume the normality that he/she desires, and so misses the treatment. This process is circular in that the sufferer cannot be independent until he/she is allowed to be, yet the parents cannot allow the individual to be independent until they feel safe enough to trust that the sufferer will care adequately for him/herself.

In order to promote good patterns of adherence the sufferer and parents need to find, maintain and adapt to a balance of control over the treatment responsibility. This is particularly demanding in adolescence as the parameters for the sufferer to take on responsibility and the parents to relinquish control are fluctuating. However, this is more likely to be achieved when the family are able to take on board the implications of the sufferer's illness but contain their own fears to the level where they can be in calm control when necessary, and not be petrified of relinquishing control or be so terrified of the disease that they try to assume normality by denying, to a considerable degree, the necessity of treatment.

#### **iv) Medical care team**

The interactions with the medical care team and perhaps particularly the doctors may reflect the CF sufferer's relationship with his/her family. The individual's responses to medical intervention and suggestions will be influenced by the family's and his/her individual style of relating to CF and to authority figures. The degree of similarity in these relationships may have implications for the doctor-patient relationship and consequently for treatment adherence.

When the sufferer is secure in the knowledge of support from family and accepts some degree of difference such that he/she can only do his/her best to manage the disease, the attitude towards the relationship with the medical care team reflects a spirit of some collaboration. The sufferer describes decisions about his/her treatment in such a way as to indicate that he/she played a part in making a choice.

"I'm starting a three monthly (pattern of intravenous antibiotics) where you have one every three months, I might not need it in three months but it'll be better for me in the long run ..... its better to keep a pattern I think because my lung function is down from what it was when I had a three monthly pattern of drips so we're going to try that again and see if that improves my breathing." D38

Under conditions where the sufferer experiences the family as being intrusive and tends towards avoidance as a strategy to maintain normality and effectively maintain his/her position on the bridge, a clear pointing out of a deterioration by the doctor may be experienced as intrusive. This enforces a harsh confrontation with reality, and runs the risk of the sufferer retreating further into avoidance in the absence of other effective strategies to manage his/her fears.

"you think oh my blowing is going to be down and you think well hopefully it won't be ..... then you go in and its down and he goes ooh your blowing is down and you're like well I can see that actually and then you just feel a bit pissed off ..... because you feel he's having a go at you because you're not well and he's not obviously, not intentionally but it feels like that sometimes" A52

The desire again is for empathy; for the doctor to be in tune with the sufferer's view point and not thrust threatening information in his/her face. Without this empathy the sufferer may feel unjustly criticised and that his/her efforts are not being rewarded.

" I know well I think I know a certain doctor doesn't think I do my physio which I know I can understand him thinking that because I've been a bit slack over the past year but I think I've done really well with my physio" A54

Adherence is a fluctuating rather than a fixed phenomenon. Responses which give the sufferer a sense of being labelled as poorly adherent foster an environment in which the sufferer feels misunderstood, guilty and blamed. This may make the sufferer more likely to be 'careful' with the truth when discussing aspects of treatment adherence

The desire for empathy, from hospital staff, becomes stronger as the adolescent sufferer tries to establish some kind of independence and autonomy, feeling that he/she has a greater awareness of his/her needs and treatment requirements and therefore wishing for a collaborative rather than a dictatorial approach. This requires a perceived recognition on the part of the sufferer that treatment requirements are

understood within the broader context of his/her life and the need to maintain as much normality as possible. This is, as opposed to a perceived view of the sufferer in the narrow context of his/her disease, the essence of difference.

If the sufferer feels he/she is being dictated to, resentment and an attitude of rebellion, may be instilled. Potentially this places the sufferer in a precarious position on the bridge by forcing him/her to compensate for the emphasis on difference with a swing back towards normality.

"maybe with the doctors treating you a bit more older. They get me mad ..... they think they can tell me what to do when I'm an adult nearly ..... it does my head in ..... like Dr Smith ..... if you lose anymore weight I'm..... I'm like what are you going to do tie me down and tube feed me ..... I don't think so..... he'd like to try it ..... they need to understand the weight business ..... I'll do the opposite of what they say .....they annoy me" F37-

38

This is an aspect of care that is perceived by the sufferer to be relatively easy to change, but would make his/her position much easier to tolerate. This serves to exacerbate the resentment felt if the sufferer considers that he/she is being treated without due respect for his/her autonomy.

"it doesn't take much does it" A53

If the style of interaction with the doctor is reminiscent of the authoritarian style of the sufferer's parents this may serve to exacerbate rebellion rather than offering the desired alternative model of negotiation and collaboration.

"I suppose its like the doctors not to have a go at you but try and help you more ..... to talk to you and explain ..... its not that effective if they start shouting at you..... if they have a go its a bit like being at home " G57-60

In contrast, in the context of the sufferer having relatively unquestioning trust in the doctors and less of a sense of autonomy, but the treatment adherence is largely motivated by fear, when the sufferer is threatened this can lead to a determination to prove the doctors prognosis wrong, and so increase adherence. The danger, however, may arise when, despite efforts, the very fear the sufferer was trying to avoid becomes a reality and the gap between normality and difference widens. This places the sufferer in a precarious position, as treatment motivation is derived from

fear, yet the sufferer's fears are poorly contained. Consequently, an increase in fear may lead to a response of avoidance and a swing in the direction of normality. In this event only serious deterioration in health can enforce recognition of difference and begin to redress the balance. When the parents find it difficult to contain the sufferer's fears this may exacerbate the swing.

"I'm going to do it this time ..... I'm going to put more weight on, it went wrong last time but I'm going to show him this time .....show him I can do it ..... but like if things did go wrong and I had to have IVs I'd sit there and just cry and think oh god ..... its only usually like my bad is losing weight, usually there's not much more than that with me but if there were then I'd worry....." B45

## COGNITIVE STYLE

The balance of normality and difference which the CF sufferer has assumed, or assumes at different points in his/her life, will influence the cognitive style the individual uses in order to maintain his/her position upon the bridge. This is inevitably influenced by the responses of others, particularly those who have significant roles in the sufferer's life in the ways discussed in '*the role of others in bridging the gap*'(pp. 49-63). Up to a point, distinctions between the role of others and individual style of managing the gap are unavoidably false, but in order to explain the possible interactions, distinctions have been made upon this basis.

The individual cognitive style in managing the gap is thus divided into the following subcategories:-

- i) Confronting the future**
- ii) Finding the motivation**
- iii) Taking control**
- iv) Cost benefit analysis: Active decision making**

### **i) Confronting the future**

In order to sustain or even develop a conviction in the value of treatment in terms of an investment for the future, the sufferer somehow has to confront the possibilities for the future and find a way of managing the inevitable fear this arouses. The degree of efficacy with which the adolescent sufferer can manage these issues will influence his/her motivation to adhere to treatment and care for him/herself, the decisions he/she makes in relation to this and the degree of control he/she takes over the illness. The style of dealing with fear is influenced by the response of family, friends and the medical care team and inevitably, to some extent, the sufferer's developmental stage and the stage of the disease.

When the sufferer is able to confront his/her fears of the future in a realistic and optimistic way, but can also contain these fears, this seems to allow the individual to keep the focus upon the pragmatics of treatment as a way of managing his/her fears. The sufferer is able to talk about and confront the issue of death and deterioration, but is not disablingly preoccupied with this. He/she copes by keeping a here and now focus. The sufferer appears able to balance a proactive and

philosophical attitude to the possibility of premature death and ultimately take the stance that there are no certainties in life. He/she might as well make the best of it. This again normalises the predicament faced by making a comparison of the degree of certainty that everybody has in life, the ultimate aim being to lead as normal a life as possible.

"I might have to do more ..... like a physio in the afternoon ..... and like my D-nase, now I'm just having it once but I don't know if I'll have to stop it now if I'm having a three monthly drip....." D49

"I can't really say I think I'll have good patches and bad patches. It might get easier for the fact that I've got more urm ..... they'll have more knowledge and stuff and they'll do more research and stuff but it might get worse as in the ..... they find something out or something, or something happens or something I don't know" I40

"urm..... not ... well ..... a bit ..... I don't worry ..... I don't worry about .... about my health because I just think well ..... I'll have more of a ..... well everyone dies whatever happens ..... but I don't really think about that I just think about getting on with things and having fun so I don't really think about that" I41

If the sufferer has tried to avoid the potential implications of CF then a bout of severe illness, which forces a confrontation with the future, may alter the his/her conviction of belief in the value of treatment . This is particularly so if the deterioration is attributed by the sufferer to poor adherence to treatment, or if treatment adherence is seen as a way to better health in the future. Increased treatment adherence due to shock, however, does not appear to last, as the associated level of fear is too high to sustain and ultimately would be mentally and physically exhausting for the individual concerned. Therefore, the sufferer has to find ways of managing this fear in order to maintain his/her sanity. This means that the level of fear ultimately has to subside and consequently so does the renewed motivation for treatment.

"I just thought oh my God I'm dying sort of thing, and then I started to do it more ..... but now..... even now (still seriously ill) I'm getting slack again because ..... then it was like oh god I've got to do something quick otherwise I'm going to die but now its like oh well I've been here a year and I'm fine now you know so they can't tell me I'm going to die because I'm fine sort of thing" A45

The treatment is adhered to, to a point where the sufferer is able to resume some sort of normality, and then the adherence drops off to prevent interference with his/her established normality. This has, until now, questioned the value of treatment and denied any significant degree of difference and consequently the need for different behaviour from the sufferer. As the disease becomes more severe and the avenues left open from which the sufferer can derive hope become fewer and narrower, one way of sustaining some level of hope is to disbelieve the doctor's prognosis in the way described by respondent A above. If the doctor's prognosis involves a condition of virtually total and unwavering adherence, then by wavering but still surviving with worthwhile quality of life and so beating the prognosis, the sufferer can sustain enough hope to continue. Without hope the treatment is pointless. Hope is necessary to enable the sufferer to rebalance him/herself between normality and recognise the increasing difference only to a tolerable degree. Avoidance of future issues can, in essence, place the sufferer in a very stable position for a time well away from the difference. Ultimately this becomes maladaptive and the sufferer becomes precariously balanced, as the nature of his/her illness swings the individual towards enforced difference.

Shocks of varying degrees can have an impact upon treatment adherence. However, it seems that if the worst possible shock, that is a serious threat to life, is confronted, this shock cannot be repeated and the same level of fear and consequent impact upon adherence can be neither repeated nor sustained. If the sufferer has faced the ultimate scare and really believes that he/she is going to die, the impact of any future shocks of a similar nature are tempered by the experience of having survived a similar crisis, despite the warnings of others. In this event it is unlikely that any further shock tactics will result in an increased and sustainable level of adherence.

" It was the ultimate shock and that didn't last so probably not, no" A56

"because it was so intense last year that feeling ..... I thought I was really going to peg it ..... urm..... and I thought am I going to be here next Christmas sort of thing ..... and now sometimes I think that but I don't think am I going to be here next year I think am I going to be here in 2 years ..... its sort of ..... I don't know ..... its just getting used to thinking about it all the time ..... you can't let it be just oh god all the time can you ..... I got to thinking well I could die in a car crash tomorrow so what is the point in worrying about it " A50

The sufferer has to reconcile the fear with a reference to normality and the possibility of death for anybody. The reality of the difference he/she is facing under these circumstances is too great to confront in its totality on a continuous basis. The passivity and disbelief as regards the severity of the disease is re-established and though the sufferer is aware of the possible consequences of taking this stance, this is not enough to promote the level of adherence desired by others.

"its really tempting fate ..... I know it is but ..... I'm just lazy I think" A46

Under circumstances where the sufferer feels that CF makes him/her very different from the norm and the individual is thus unable to manage his/her fears by normative comparisons with others, this makes the prospect of a foreshortened future extremely difficult to face. The feelings of difference serve to make the sufferer feel isolated, as he/she believes that others have an incomparable degree of certainty and so a very different outlook for the future and very different hopes. The realism becomes so overwhelming that it cannot be balanced by any comparable degree of optimism.

" I mean like at the moment my future plans are like ..... you know the next couple of years ..... if I didn't have CF I'd probably, because my friends always go ..... I don't know what its like for them but they talk about when they're 90 and stuff ..... and what job to go and get and for me its like what job am I going to get its well ..... I don't really care because there's not much point ..... as for when I'm 90 I mean I don't think so....." E37

The degree to which fears can be contained depends inevitably upon the severity of the disease. When the disease is less advanced the possibility of death is a considered, though much more distant, reality. This makes it easier for the sufferer to confront the harsh realities but quickly regain a proactive perspective rather than being disabled by fear or despondency. The sufferer is able to be humorous about the prognosis when others are contemplating a future which is not real for him/her, and is able to place boundaries around his/her fears so as not to get engulfed by them. This can be achieved by a variety of strategies such as distraction, normative comparisons with other CF sufferers and those who do not have CF, pulling out the more positive aspects of life and a focusing upon future hopes and aspirations. The ability to do this seems to have been developed first and foremost from the containing reassurances by family, but also by friends. This task is inevitably more easily achieved by the younger adolescent CF sufferer as the future is literally more distant and his/her capacity for abstract thought may be less well developed. This

may therefore, make it easier to maintain a concrete focus and avoid becoming overwhelmed by existential issues.

"Yeah when I think I'm going to die, that's the only time say when you're lungs are that clogged up you can't breathe and you die of that or you have to have a heart and lung transplant and you don't survive or liver damage and go yellow, I think oh my god why don't I just go and stab myself (laughs) that's the only thing ..... I hope I don't die painful, then I start crying so I try and make it, it sounds nasty but make it as a joke so i don't get that bothered .....my friends .... they're all going on about wrinkles and I'm like ohh I'll be dead before then, I won't get any wrinkles I'll be dead before 60 ..... and they're like oh that isn't fair so I'm like shut up I'm only joking" F26 (age 18)

"If I'm sat thinking ..... I'll probably go put some music on right and forget about it, stop worrying God I'm only 17 ..... I don't think what if I've only got 20 years I think have a good time, get married, have some children, marry someone great and take it from there"  
F27 (age 18)

"Only like when I get a bit older and if I got right poorly I might start worrying but if I work hard I could stay quite well so..... not really no other than that" H54 (age 11)

If the fears are well contained and the sufferer feels that he/she has some degree of control, then the individual is able to contemplate some desires for the kind of future he/she would like.

"I'd like to be a hotel receptionist ..... I think I'd like to have babies" D49 (age 14)

For some the future, in terms of the implications of their disease, is too frightening to contemplate, but the sufferer is still able to hang on to his/her future hopes by avoiding thinking about the prognosis. Plans for the future, in and of themselves, can help the sufferer to place a boundary around his/her fears.

" ..... I don't really think about it ..... "G30

"there's a lot of things I've thought I'd like to do but the main thing is like to get somewhere in music ..... like in a band or open a shop or something ..... " G52

The absence of any plans, however, can be a sign of avoiding issues related to the future.

"No..... I just take each day as it comes" C75

The potential danger in this again lies in whether or not the sufferer takes this to the extreme and tries to live normally, as if he/she does not have CF and denies the necessity and importance of treatment until this is thrust in his/her face by a crisis.

A commonly occurring strategy for managing fears of the future is, not surprisingly, to harbour hopes of a cure, at best, or, at least more effective, less time consuming and less noxious treatments, and draw hope from the best possible case scenarios.

" I'd like to see a cure obviously, but if not a cure then I'd like to see more advanced treatment that works a lot better and stuff and get closer to a cure and like quicker more comfortable ways of doing physio, just a better way of living really ..... but things are pretty good at the moment anyway so.." D53

"There's all these documentaries and stuff about it and I think, well one says they're coming up with a cure and one thing which boosted me was, I don't know when it was there was something in a magazine about someone who lived to 50 with it which is really good" I32

These are for sufferers realistic possibilities when they are in relatively good health. However when the disease is more advanced it is much more difficult to derive hope and alleviate anxiety in this way. This could be terrifying when, at the same time, due to deteriorating health, this is rapidly becoming one of the few potential positives and a sole motivation for treatment adherence as the other possibilities are depleting. Strategies open to the sufferer with less advanced disease, such as focusing upon the positives and normative comparisons with others, begin to lose their meaning just when containment of fear is needed most.

"like sometimes I think what's the point in having a girlfriend, its not going to go anywhere because I'll just die so ..... what's the point in going to college I'm not going to work anywhere ..... I'll just bum around ..... then I think well if suddenly a cure comes along I'll be a bit buggered ..... I'll think oh dear what do I do now (laughs)..." E102

"They haven't come up with a cure for anything like that ..... no major disease so why should they ..... I can't think they're going to come up with a cure for CF ..... in the time like ..... " E103

## ii) Finding the motivation

The belief that treatment has both immediate benefits and is an investment for the future, seems to promote adherence. This, of course, depends upon the sufferer having confronted and made a reasonably realistic appraisal of the likely future implications of having CF.

"urm ..... I think its keeping me well ..... it doesn't really disability anything I do because I'm not very good at running or anything anyway but I'm ..... It doesn't like when I go and play badminton it doesn't bother me ..... when I'm older it'll help for then because it stops it all piling up and stuff" I28 (age 12)

"the doctor said you get to a stage where you're not going to get any better but obviously you can get a lot worse if you don't stick with your treatment when you're younger" D37 (age 14)

It seems that it is easy to recognise the benefits of treatment at an intellectual level but there is variation in the degree of conviction. Some, like the respondent above, indicate a clear belief in the value of treatment whereas others hint at a far greater degree of scepticism. This may, in part, be mediated by the age of the sufferer and stage of the disease. The younger sufferer is perhaps less likely to have had his/her beliefs challenged by severe illness, possibly in spite of perceived good treatment intentions.

"urm .... I suppose if I stick to it I might not be back in here so soon ..... and I might feel more well in myself ....." G29 (age 17)

" If I do it it'll probably make me feel better and stop me from getting ill" C38 (age 15)

This is not enough to consistently influence increased adherence.

"sometimes ..... not always" C39

Conviction in the belief that treatment is worthwhile in the short and long term can be derived, at least in part, through trust in the doctors. If a sufferer has been seriously ill in the past and is then consistently better under the care of a different physician, this can be considered to provide evidence of the efficacy of treatment and provide motivation to adhere to avoid a similar adverse experience in the future.

This can also provide justification for an authoritarian attitude on the part of the doctor.

"well I've got to keep my weight up haven't I ..... ever since I've been under Dr Smith I've been alright ..... he shouted at me once because I'd not put on weight but I've never been really poorly under him" B32

"I'm sure it'll make me feel better, I mean obviously it is because I'm like I am now and I want to stay like this and I don't want to be poorly like I was because I mean I wasn't very well at all when I was little ..... I mean I had a tube in for about three years and I was like this (sucks cheeks in )" K 34

Under these circumstances however, the motivation seems to arise out of fear of having to do worse treatment, or more treatment, rather than out of a sense of making the best of a bad situation. This appears to relate to the response of others, since when fears are contained, treatment is less threatening and beliefs about benefits are realistic. When the sufferer's fears are not contained, fear can motivate adherence, but if the sufferer does everything as they should and then falls prey to worse illness and more noxious treatment, this can lead to a whole questioning of the value in the first place.

"Well you get better a lot quicker don't you and it saves having to have a lot more if you don't ..... say if you're on antibiotics and you don't take them it could be IVs next time and they're twice as bad" B32

When the benefits of treatment are deeply instilled in the sufferer's belief system and treatment is taken as a part of life that only minimally interrupts normality, this seems to allow the sufferer to derive motivation much more easily. He/she is able to respond actively to illness by increasing treatment in a sensible and uncomplaining way, without the perception of a huge effort.

"when ..... I'm unwell I think I naturally just switch on to doing more physio and things" I34

Similarly, if there is a conviction in the benefits of treatment, anxiety in relation to a clinic visit can increase adherence out of the will to receive good feedback. If the results are good this can lead to increased motivation, fuelled by enthusiasm and relief that things are going well. Conversely, if the results are bad, this can lead to the determination to take control and put things right for next time. These properties

of motivation seem to apply in the context of the self directed sufferer with supportive family and peer relationships, who believes he/she has a degree of control over the illness, but at the same time has a realistic awareness of the limits of this control. Regardless, the increase in adherence level does not seem to be sustained.

"I usually get verbal diarrhoea afterwards and I don't stop talking ..... and I usually start repeating what they've said is good about it and what I need to work on ..... and then I'm determined to work on it ..... and I know it sounds like I'm not trying hard but after a while the novelty wears off of doing it all" D15

The level of self awareness is such that the sufferer is able to anticipate reasonably accurately the outcome of clinic visits and prepare him/herself accordingly.

" we went to see 101 Dalmatians ..... that was because we were expecting a drip but we usually go to the pictures anyway whether I know I'm going to get a good report or not" D14

If the sufferer does not have this level of awareness then the individual lays him/herself open to more shocks, as results at the clinic strip away the defence of denial. If the sufferer has no other strategies to deal with his/her fears then he/she is left with no option but to retreat back into avoidance at the earliest opportunity, in order to redress the balance towards normality.

"usually when I come to have an appointment at the clinic ..... usually I think I'm weller than I am I suppose ..... like they do a test to see what my blowing is like and its usually lower than I think ..... usually I've felt alright at home ....." G32

Increased self awareness can be enforced by a shock which requires increased recognition of vulnerability in order to reduce the chances of a similar frightening experience in the future. Avoidance is no longer an advantageous, practical or possible strategy.

"I know how bad I feel so I think oh God better go in quick ..... last year I just thought oh I'll be alright" A49

The CF sufferer is caught up in a cyclical process of increased adherence in response to adverse circumstances or renewed enthusiasm when effort invested is believed to be paying off. This then subsides until renewed motivation arises out of

circumstances such as a forthcoming clinic visit, illness or well-being. Regardless, there are limits, as the task can never be completed but has to be continually integrated and balanced in the sufferer's everyday life. Normality, of a kind to suit the sufferer, has to be sustained. The demands of a continuous, high level of adherence do not often match these wishes, as they demand too much digression from normality and an intolerable degree of difference and effort.

"I suppose its the only way you can live with really, I don't think I could do anything 100% for the rest of my life ..... Its like anything ..... I can't do anything forever" G36

"I suppose its like you've done and you've done it forever and you're going to be doing it forever so ..... if its just an assignment you can just do it so you're not going to just keep putting it off ..... even though I do but..... you do it eventually and you know that's it its ended with so ..... you're glad to have finished it..... with treatment you've finished it for now but you know its always going to be there so ..... it never ends....." G63

### **iii) Taking control**

The degree to which the sufferer feels in control of the illness will be influenced by his/her ability to confront and manage future fears, beliefs about the benefit of treatment and the individual's illness experience. The more in control the sufferer feels, the more likely he/she is to believe in the value of the treatment and so sustain the motivation required to carry it out rigorously.

If the sufferer has limited conviction in the value of treatment he/she seems to perceive that fate plays a larger hand in determining his/her well being than does the individual him/herself. The sufferer has some sense of the benefit of treatment but his/her attempts to stay well can be scuppered at any time by events beyond his/her control. This leads to a lapse into passivity, waiting for luck to change before responding with increased adherence. When the sufferer perceives him/herself to have only limited control, motivation for treatment is only really derived when the sufferer is feeling well.

The lack of active coping strategies makes it very difficult to fight back in the face of adversity. The sufferer will feel depressed in the face of poor clinic results and deteriorating health and feel good in response to improving health. However, these results are not attributed strongly to the sufferer's own actions, therefore he/she

does not see him/herself as being in a strong position to make any changes. Under these circumstances, no amount of pointing out the dangers in poor adherence to treatment is likely to have more than minimal impact. The sufferer ultimately has little sense of control over his/her illness, and being dictated to is unlikely to promote this as it, again, takes away the individual's control over his/her life.

"Whether I could be bothered to continue.....too much hassle" E77

"Yeah I can't remember what happened.....I just got depressed and then..... I started college, I still wasn't well, wasn't completely better, but I was well enough to go to college.....then I had some IV's, I think they were the best IV's I've ever had.....and after I finished those I was like yehey.....I was like really happy.....in party mode" E79

"Well I haven't been in hospital since before Christmas and I've been quite well ..... but I'm only in hospital now because I had flu ..... which has like stopped me oooohh..... and I didn't take my medication because I was feeling crap ..... so that was what brought me in here, if I didn't have the flu I wouldn't be in here ..... so I'm just annoyed" E67

"I wasn't taking everything ..... but I was taking more than I usually do ..... just because I was feeling quite good" E73-74

When the disease is severe in this way the sufferer has to undergo a process of adjusting to new limits and progress towards a greater degree of certainty, but in an unwanted direction as the disease advances. The degree of control that the sufferer can realistically take diminishes and normality, as it is perceived to be for others, shifts further out of reach. This requires the sufferer to span a much wider gap and to assume a much more precarious and more difficult to manage position upon the bridge.

When the disease is less severe the sufferer can manage the fear of the limits of his/her control over the disease, such as the fear of sudden illness, by minimising it with the philosophy that if he/she does all he/she can to keep as healthy as possible, then the fear is less likely to become a reality. This belief can also be instilled by the containing encouragement and reassurances of family and friends. If fears are not well contained this may lead to increased adherence to treatment until fate plays a hand, and its value is questioned due to observation of the fate of

others. This leads to an exacerbation of fears and further questioning of the degree of control the sufferer can take.

"I mean all of a sudden catching something like pneumonia, a thing like that you can't always recover from ..... Andrew died last year and Mark the year before but ..... I mean knowing them and Mark was always poorly but you'd never think that, I mean he did come home a couple of times but then all of a sudden he got poorly then never came home really"

B57

The sufferer may try to combat his/her fears relating to the disease by the belief that he/she will be able to find ways to avoid the disease getting the better of him/her, no matter what. This may contain the sufferer's fears in so far as he/she feels invincible, but does not necessarily facilitate optimal adherence. This may then place the sufferer in a precarious position if, and when, this invincibility is brought into question by, for example, the progression of the disease.

"When I wake up.....which I think I have done, I think I'll be fine ..... I think I should be fine ..... when I think about the future I just think ten cars and the cash ..... that's all I think about really ..... that's the only thing I think about..... if I need some expensive drugs then I suppose I'll just buy them....." J43

If the sufferer has a realistic but optimistic outlook regarding CF and its prognosis, when challenged by fate in this way he/she seems able to put this down to bad luck, but not question the value of the treatment in improving the prognosis ultimately. The sufferer has a much more stable sense of control because he/she has been able to look reality in the face, but then contain the fears, accept the limits, pull out the positives and continue to strive for as much control as possible.

"well if I go home then mum and dad say right you'll have to have another session of therapy or we'll have to start really getting down to it ..... urm but... because I know that my mum, dad and I are doing our best, the doctors have told us we're doing our best" D58 (explaining response to poor outcome at clinic visit)

"No not really, the last time was last year..... that was because I had a virus" D59 (in response to question how often she is admitted to hospital)

The sufferer is able to maintain the belief that what he/she does makes a difference and so react to adverse circumstances by reasserting their control and by increased

adherence. This feeling of being in control in itself lessens anxiety about the illness and keeps the sufferer in a kind of proactive spiral.

"I don't know why but I'm generally not bothered ..... if my blowing goes down I think oh I can improve it its not as if I'm going to die or anything ..... you could've caused it by not having your nebuliser or whatever so I don't sit and worry ..... I just think I can sort it out" F29

Sometimes, in the face of severe illness the sufferer can switch from a defeatist and so anxiety provoking, out of control stance to regain a positive, more proactive stance. This change can come about by a variety of mechanisms. If the sufferer is able to regain a sense of control over his/her illness this can lead to a lifting of mood enabling an increased motivation to get well to develop.

"before Christmas I just felt ..... oh why am I bothering sort of thing ..... and that was when I was quite worried about it ..... I suppose it's still there but I'm trying to be more positive about it" A8

"now I've been away and I've had a chance to rest I'm quite determined to get better and I think sort of positive as well" A4

This change can be brought about by an opportunity to go back to a more normal lifestyle, a break from the imposed difference, for example hospital, and a chance to reflect in a safer environment where the difference is there, but is manageable, because it is not continually being 'thrust in your face'.

"there's nothing in particular, I'm just feeling positive probably because I've been shopping "

A12

Circumstances which are not within the sufferer's control can also play a part in the ability to take control and foster a proactive attitude. The sufferer's limited sense of control is boosted by fate working in their favour. Even a change in the season can mean that proactive attempts to take charge are less likely to be interfered with by the chance catching of an infection.

"during the winter I feel quite down because I deteriorate quite a lot ..... but in the summer I'm normally quite positive" A9

This is however a relatively precarious position to be in as the sufferer is waiting to see in which direction the wind is going to sway the bridge, as opposed to trying to anticipate and counteract this.

#### **v) Cost/benefit analysis: active decision making**

The beliefs that the sufferer holds about the value and importance of treatment will influence the more immediate decisions he/she takes with regards to carrying out the treatment, the feelings he/she has following missed treatment, and the rationalisations he/she make about digressing from the treatment regime in the short term. Decisions are based on the sufferer's illness experiences, recommendations of the doctors and others responsible for his/her care, understanding of, and positive experiences of, the treatment.

Decisions about treatment are often dependent upon health status. This is not however a straightforward linear relationship. When the sufferer is feeling ill or tired he/she is less likely to adhere strictly to the treatment, particularly those aspects of it requiring the most effort. Equally, when the sufferer is well he/she may take the decision to skip treatment, and rationalise this with the belief that he/she can afford to do this because he/she is in relatively good health. The sufferer is feeling normal and does not want, therefore, to disrupt this with a reminder that he/she is by necessity required to behave differently from others. This may be particularly true when the CF sufferer is out of his/her usual environment, such as on holiday or staying over with friends. The costs of interruption to the enjoyment of the different routine may be considered to outweigh the benefits of carrying out the treatment. The sufferer is living closely to how he/she perceives others' normality to be. Also the pressure of others is easier to ignore, or may be absent altogether, if away from the family and the sufferer feels well and relaxed and so finds it easier to justify missing the treatment.

"like when we go abroad and you haven't got much time then its a healthy atmosphere ..... sometimes I can't be bothered and like half do it ..... I'll catch up tomorrow but then I don't, I know myself I won't but at the time you're getting ready to go somewhere and you think 15 minutes of that ..... I'll smudge my make up" F23

Adherence can be promoted under circumstances where the negative consequences of poor adherence are immediate, so the decision is made that, in this instance, the costs of missed treatment outweigh the benefits of adhering. If, on

the other hand, the benefits are perceived to outweigh the costs, the sufferer may decide that he/she can reasonably suffer minimal, if any, adverse effects from missing the treatment. The decision is taken to miss treatment rather than risk dissent from normality as perceived by others.

"tablets I miss sometimes but not very often and only with things like where you only need one or two ..... you know like if I had a Mars Bar I wouldn't miss them with that...." A35

If an aspect of the treatment is not considered to be vital by the sufferer when no immediate adverse effects are likely, this makes it easier to forget treatment and actively decide on a course of partial adherence, as opposed to religiously following the regime as prescribed. If there is no immediate effect then it is obviously much harder to believe in the possibility of long term damage. This therefore makes it easier for the sufferer to assume normality by avoiding the real, serious potential implications of poor management of the disease. This may be equally true for parents as it is for the sufferer. A parent who tends towards denial will find it very difficult to instil a true sense of the long term benefits of treatment and so reduce the likelihood of cost benefit decisions being made with a long term, as opposed to short term, perspective. The option is taken for normality in the short term, as opposed to an investment for greater normality in the longer term.

"the antibiotics sometimes I miss one day purely through forgetting but I don't think that matters" A35

Alternatively, treatment is missed as the sufferer is able to decide, and convince him/herself, that this has been compensated for in some way and therefore there will be no harm in missing it. The sufferer takes this decision about his/her care, regardless of the advice given by the doctors. It is based on what he/she believes is required and his/her perceptions of the purpose, and mode of effect of the aspect of treatment concerned. It is as if the sufferer perceives he/she can span a wider gap on the bridge temporarily, or manage more sway towards normality, as he/she can pull back to a more balanced position to rest and recuperate at a later stage.

"it wasn't like every night ..... there were days when I'd have my regular two physios but sometimes I'd just leave it to one but because I'd had P.E inbetween it was just like having another physio session anyway" A43

The efficacy of these supposed substitute treatments is, however, an important factor which is taken into consideration. There is an active trade off between effort required and perceived efficacy.

"not as hard treatment, I mean I kind of have my PEP mask but it doesn't do as good as physio ..... if they could find something that'd be as small as a PEP mask you know, be just as compact kind of treatment but be just as effective., more effective than physio then I'd go along with it ..... I wouldn't want to do PEP masks everyday because I know it wouldn't be doing me as much good as having a proper good physio" K25

If the sufferer believes in treatment as an investment for the future he/she may then use a previous track record of good adherence as a way of rationalising a reduction in adherence to some degree at a later stage. The sufferer may compare his/her track record with what he/she believes the track record of fellow sufferers to be like and use this as a means of justifying a slackening in adherence. This can be conceptualised as a way of managing the guilt the sufferer feels at missing some aspect of the treatment and possibly relates to the individual having been previously managed in a very authoritarian way by his/her parents.

"when I was young I never used to miss it ..... now I probably miss it once a week, one session which is nothing compared to everyone else in here but to me, I mean before I never used to miss it ..... I remember I could sit down and I'd be lucky if I could remember one time in a whole year ..... like that I missed it and that was it ..... I mean now its nothing"

J28

The sufferer again takes decisions which give the optimum level of benefit in terms of leading a quality, worthwhile, and as normal as possible a life. The immediate consequences of misjudging this balance can be discomfort from over doing treatment as well as under doing it.

"If you can't get anything off your chest then don't keep trying because sometimes you try and try and you can't get anything off and you can give yourself a sore chest and throat" H12

The observed and experienced perception of benefit is more likely to lead the sufferer to make a similar decision, or to take a similar course of action when faced with a similar situation in the future, so promoting adherence.

**"my lung function is down from when I had a three monthly pattern of drips so we're going to try that again and see if that improves my breathing" D38**

**If the sufferer is able to pull out and keep the focus upon the positives rather than the negatives, he/she is more likely to make decisions based on belief in the potential benefits of treatment. The sufferer is able to be more realistic as he/she can confront his/her fears, contain them and act upon them. The decisions he/she takes are based on a way of coping which actually becomes the individual's way of containing his/her fears.**

**"so just pick out all the good, well not the good bits but the better points and weigh down the bad points ..... don't get it out of proportion" D43**

## **DISCUSSION**

The main findings of this study will be summarised and discussed in terms of their relationship to the literature cited in the introduction. As the study progressed, however, it became apparent that other aspects in the literature were relevant, and these will be discussed as appropriate. Further, the initial research questions raised (p. 14) have also been developed throughout the analytic process but, as will be seen, remain implicit within the discussion of the theory proposed.

It will be suggested that many of the findings could be unified and made coherent via the explanatory framework offered by attachment theory. This will therefore be introduced and discussed in terms of possible implications for the study and understanding of adherence to treatment in adolescent CF sufferers. The limitations of the grounded theory approach, and the particular stance taken within this study will be considered, along with some more general limitations. Finally suggestions will be made as regards the implications for clinical intervention.

### **Relationship of the analysis to the literature**

The analysis of the data collected for this study reflects several aspects of the current literature relating to adherence to treatment for CF in adolescence. However, it seems that the study expands upon many of the issues raised, and provides possible new ways of conceptualising links between the different aspects of the relevant literature which, in turn, generate new ways of understanding the factors influencing adherence during this life stage.

The core theme which runs through the analysis is, perhaps not surprisingly given this stage of the life cycle, related to forming and maintaining a stable identity. As Erickson (1959) suggests this is the most fundamental and important task in adolescence. For the adolescent CF sufferer it seems that this does, as Pumariega (1982) suggests, pose particular challenges. This study has shed some light upon how these challenges may threaten treatment adherence, and upon some particular life experiences, and family styles which may exacerbate the challenges faced in adolescence to the point where treatment adherence is threatened.

According to this study, the task of the CF sufferer is the formation of an identity which encompasses CF to a tolerable degree, but at the same time does not dismiss CF to the point where treatment is avoided, in an attempt to avoid the

implications of the disease. To a varying extent, the adolescent CF sufferer has to find ways, as do most adolescents, of being accepted. The treatment and signs of disease pose an extra potential threat to this and so impact upon the CF sufferer's identity formation.

Threats to identity are ever present, but can be exacerbated at points of transition such as starting a new school or college, or by a deterioration in health status which is either relatively temporary or marks a stage in the progression of the disease. Since most of the literature on adherence relates to adulthood, or borrows models from adult health research, the impact of developmental factors upon adherence is not well conceptualised, as yet. The literature to date has ambiguous findings as regards the impact of increasing severity of illness upon treatment adherence (Abbott et al 1995). The main factors suggested to impact upon this are those of increased treatment demands (Czajowski & Koocher 1987) and immediate benefits of treatment (Abbott et al 1994).

This study suggests that the reason for continued ambiguity in the findings of quantitative studies of this issue may lie in the complexity of the relationship of severity of illness to adherence. The theory proposed implies that increasing severity of illness may increase adherence to a degree, if and when missing treatment may make the outward signs of the disease obvious to others, so posing a threat to the identity of the sufferer as normal in the eyes of others. In contrast, as the disease becomes more severe, the chances of a cure become more remote and make it more difficult for the sufferer to sustain hope, leading to a questioning of the value of treatment. Similarly, a dramatic increase in the severity of the illness may lead to a peak in treatment adherence in response to the fear this evokes. However, this feeling may be too intense to sustain, leading again to a reduction in adherence, though adherence may remain higher than it was prior to this crisis. According to the theory proposed in this study, adherence fluctuates considerably and a measurable, consistent effect of disease severity upon adherence levels will necessarily be difficult to attain consistently.

Points of transition pose a threat to continued adherence as the sufferer is either required to hide both the treatment and signs of disease, or disclose that he/she has the disease to new people. This is an area which has not been specifically raised in the literature relating to adherence. However, Admi (1995), in a grounded theory analysis of older adolescent and young adult CF sufferers, suggested that individuals with strong self identities felt free to disclose their CF in certain social

situations, but equally felt comfortable hiding the signs in other situations, where this was felt to be appropriate.

Strategies to manage disclosure were found retrospectively to change through adolescence, with younger children giving concrete explanations, but the issue becoming more complex with the acquiring of the capacity for abstract thought. This then requires a change in approach to disclosure and clearly provokes anxiety for the individual as he/she is able to hypothesise about potential events and outcomes related to disclosure. Information management was then shaped by the actual experiences of disclosure and 'turning point events', meaning events when the issue of whether or not to disclose is brought to the forefront.

Clearly a point of transition for the individual can provide a whole catalogue of 'turning point events'. Consideration of the findings of Admi (1995) verifies the proposal made in this study that positive experiences of disclosure are more likely to lead to further disclosure. It also seems to add weight to the suggestion that adherence, particularly for the younger adolescent who is still unsure of disclosing, and whose identity is in a greater state of flux, is likely to be related to the felt need to conceal the disease from others and strategies available to the sufferer to manage these potentially threatening situations.

Admi (1995) suggests that total concealment of the disease was unusual. However the disclosure stories tended to minimise the implications of CF by not mentioning the prognosis or the deteriorating nature of the disease, and by comparisons with other, less stigmatising and often less serious, conditions such as asthma. Importantly, Admi (1995) purports that audiences for disclosure were selected by the sufferers on the basis of how able they perceived the audience to be to manage the information given. This may reflect the issues raised in this study which indicate reticence to explain to others and a reluctance to tell, perhaps particularly, in those individuals whose families are apparently less able to contain their fears. If the family have not been a good audience for information about the disease this may set up the expectation that others will not be. This may lead to greater efforts to conceal the CF resulting in reduced treatment adherence.

The accounts given in this study reflect the apparent importance given in the literature to the role of the family and coping in relation to adherence. Many positive features relating to adjustment to CF and adherence seem to co-exist within the same scripts suggesting the influence of an overall family perspective. The

subcategory of '*containment of fears*' within the category '*the role of others in bridging the gap*' (pp. 49-54), in its simplest form, could be analogous to the suggestion in the literature that adherence is highest in families in which there is an atmosphere of co-operation, integration, expressiveness and optimism (Patterson 1985; Patterson et al 1993). This study could be argued to expand the conceptualisation of this issue. It suggests the reason for these factors aiding adherence is that an approach of this kind places boundaries around the sufferer's fears, and allows the individual to gain a sense of comfort and well being from the family and an increased sense of personal value.

This study could be seen to add to the conceptualisation of these issues, as it offers ideas as to how families achieve a balance between level of adherence and psychological adjustment to the illness. This goes some of the way to addressing Drotar's (1995) comment regarding the lack of understanding of these processes. To pretend however that all the gaps have potentially been filled would be a gross overstatement. The main benefit of this study is that the findings do not suggest that the families which fare less well are simply the opposite of those that do, i.e. pessimistic, disintegrated, poorly co-ordinated and poor in expressive skills.

The links in the literature between family style, coping with chronic disease and the impact of these upon adherence were also reflected in the main findings of the study, particularly within the categories '*the role of others in bridging the gap*' (pp.49-63) and '*cognitive style*' (pp. 64-80). The idea of the cohesive, interconnected family leading to a sense of mastery, optimism, self reliance and flexibility in thinking, proposed by Hauser et al (1993), could be related to the ideas laid out in the analysis that those sufferers whose fears are contained, and who are allowed a degree of independence and autonomy that is steadily negotiated, may be more likely to hold strong beliefs in the value of treatment and it's long term benefits. Also, this may enable sufferers to take control of their illness in a positive way and to be actively making decisions about treatment in terms of costs and benefits.

Essentially, the concept of interdependence (Eiser 1993) as being the preferred goal which is most likely to promote adherence, as opposed to the traditional dependence/ independence split (Gudas et al 1991; Pownceby 1996), is also upheld within the theory proposed here (see subcategory '*degree of autonomy*' pp.58-60). The accounts given in this study clearly reflect the suggestion of McCracken (1995) that if families are able gradually to hand over responsibility for

treatment and self care across the course of childhood, this may reduce difficulties arising with treatment adherence in adolescence and enable the child to develop a sense of mastery and control over his/her disease. However, the depth with which this is conceptualised as a process impacting upon adherence is added to by this study. The links between the different issues of family, identity and individual cognitive style are more clearly illustrated.

The analysis of this study also addressed the issues of doctor patient relationship. As Korsch et al (1971) suggested, a perceived empathy on the part of the doctor may facilitate adherence. This issue seemed to be apparent in the accounts of the adolescents who took part in the study, but the dimension of the interrelationship between the sufferer's cognitive style, his/her relationship with the family and those with the medical care team was added as an important factor. It seemed that when the CF sufferer experienced his/her family as supportive and collaborative he/she may be more likely to experience the physicians in this way. If, on the other hand, the sufferer was used to a more authoritarian attitude at home, he/she seemed to describe a similar relationship with the doctors and to resent this. Similarly, if the sufferer was dealing with the disease by leaning heavily towards normality, the pointing out of the differences by a doctor could be experienced negatively. This again is clearly a complex issue and challenges the assumption, which seems implicit when reading existing literature, that the doctors style of interacting with different groups of patients will be relatively consistent.

It was also clear that Donovan's (1995) assertion that patients make their own decisions about treatment based upon their beliefs, circumstances and available information was a feature in the accounts of the respondents, though a much stronger feature in the stories of older adolescents. This presumably is another facet of increasing autonomy, independence and awareness of the disease. Again, this taps into the developmental perspective taken of adolescence and highlights the difficulty faced by physicians and adolescent sufferers, of staying on the same wavelength over time. This could be due to the ever changing parameters of what constitutes the open communication and flexibility purported in the literature to facilitate good adherence. These factors, according to the analysis of accounts given in this study, may fluctuate with varying degrees of regularity as the experiences and identity formation of the adolescent, and consequently his/her relationships with others, alter as he/she attempts to maintain the balance between normality and difference in the context of rapidly changing demands, and developmental stage.

Though it is clear that many aspects of the literature to date are reflected within the analysis of the accounts given in this study, the nature of this literature is fragmented. However, the relevant literature and the theory proposed here could be explained, relatively cohesively, by relating the findings to attachment theory. The consideration of attachment theory in this context opens avenues for speculation as regards outcome in CF in terms of treatment adherence, further development of the theory proposed and possible strategies for intervention. Rutter (1995) states that the major features of attachment theory have been consistently supported empirically, and have been expanded to theorise about aspects of relationships and social development beyond infancy and into adulthood. He emphasises the potential importance of the implications of attachment concepts for clinical practice.

### **Attachment theory**

Attachment theory was first proposed by John Bowlby in the 1950's in response to research showing the damaging effects of hospital and institutional care on young children (e.g. Goldfarb 1943; Skodak & Skeels 1949 cited in Bretherton 1985). Attachment theory argues that the quality of early parent-child relationships and interactions leads to predictions of parental regularity and sensitivity on the part of the child, which guide the behaviour of that child towards the parent.

The development of the strange situation test (Ainsworth et al 1978) provided a classification system for the security of infant's attachment to his/her caregiver, usually the mother, and led to the identification of three main patterns of attachment; secure, avoidant and ambivalent. The strange situation test involves settling the child with the parent in a playroom, a stranger is then introduced and the caregiver then leaves the child alone with the stranger for three minutes before returning. Later, the parent leaves the child alone for three minutes. The infant's attachment security is classified according to the child's behaviour upon reunion with the parent.

Secure children protest at separation, greet their parent on return, seek comfort and then resume playing and exploring their environment. Avoidant children typically show no or very little distress on separation, ignore their parent upon return and carry on as if he/she were not there. Ambivalent children tend to show extreme distress on separation but refuse comfort from their parent, then oscillate between comfort seeking and angry resistance of his/her reassurances and do not resume

exploration (Ainsworth 1982). A further group of children were originally seen as being unclassifiable. Main & Solomon (1986) observed that the behaviours of these children upon reunion were very disorganised and disoriented and proposed a further category for these children. However, as it remains unclear whether these are not simply an extreme version of ambivalent and avoidant patterns (Crittenden 1989) this category will not be applied within the context of this study. This pattern is, anyway, considered to be relatively rare and has little relevance in relation to the sample of respondents used.

Bowlby (1969); (1982); (1988) cited in Bretherton (1985), suggested that the children will, through their attachment experiences, build up models of the world of people and objects and the self in relation to these. These models governing expectations of regularity in care giving become internalised as internal working models which guide the child's, and later the adult's, behaviour when encountering new situations. The nature of the model the child constructs depends upon the responsiveness and sensitivity of the caregiver(s) and the extent to which the attachment figure(s)' behaviour facilitates the formation of a secure base from which the child can actively explore the environment. The model is constantly updated in the light of experience and growing cognitive capacities. However painful, insecure attachments can lead to defensive exclusion of painful material from consciousness.

Essentially, children who know that their caregivers will respond sensitively and according to their need (secure pattern) will learn to communicate openly and see themselves as worthy of help and comfort. Secure children tend to demonstrate more positive affect, be persistent in problem solving situations and be rated as being more socially competent and popular with peers. They are apparently more able to tolerate negative emotions whilst maintaining constructive interaction with others (Sroufe 1983; Sroufe et al 1984).

Children who know that, by and large, their parents or caregivers will respond punitively or adversely to expression of need, will learn not to communicate their needs and will demonstrate an avoidant attachment pattern. They will tend to cut off or hold back emotive displays as they have learnt that this reduces conflict with caregivers whom they experience as rejecting or insensitive ( Cassidy & Kobak 1989). Their problem solving abilities will tend to be restricted ( Main et al 1985).

The child who cannot predict how their parents or caregivers will respond to need, whose parents are inconsistent in terms of responsivity and sensitivity will develop

melodramatic ways of signalling need which involve heightened expression of negative emotions, such as fear, anger or distress. These children may appear, either to be clingy, as this behaviour maintains contact with the caregiver, or to be angry and rejecting. Both these patterns of behaviour interfere with the child's confidence in exploring their environment and coping with novel situations (Ainsworth et al 1978). This behaviour is characteristic of the ambivalent pattern of attachment.

Bretherton et al (1985) suggested that the concept of internal working models of attachment takes the scope of attachment theory beyond childhood and provides a way of understanding adult relationships and parents relationships with their children. The internal working model the child develops and carries through to adulthood is predicted to shape future relationships.

The Adult Attachment Interview (George, Kaplan & Main 1985) provides a way of assessing attachment experiences retrospectively. The use of this instrument led to the development of classifications of adult attachments which have been found consistently to predict the attachment security of their children (Main, Kaplan & Cassidy 1985; Fonagy, Steele & Steele 1991). These studies showed that parents of secure children valued the importance of attachment relationships, and gave coherent and balanced accounts of their own childhood experiences and relationships with attachment figures. This was true also of some parents who had very unhappy upbringings but had managed to make sense of this and revise their internal working models accordingly. Under the rating system for the Adult Attachment Interview (Main & Goldwyn 1985) these parents are classed as autonomous and free to evaluate or secure, producing secure children.

Parents of avoidant children were unable to recall much of their own experiences as a child, denied the relevance of these experiences, gave bland, idealised accounts with few clarifying examples, or discussed their relationships with parents in a dismissing manner. These parents are classed as detached or dismissing in attachments. Finally, parents of ambivalent children gave confused, lengthy accounts which were often confused and contradictory. They gave the impression of still being entangled or enmeshed in their past and were able to relate many associated thoughts and feelings but were unable to draw any conclusions. They conveyed a tendency to discuss their parents with derogating anger. These are characteristics of the preoccupied classification.

## **Attachment theory and Cystic Fibrosis**

Attachment theory has scarcely been applied in the context of chronic illness and does not feature more than peripherally in the literature relating specifically to adherence to treatment or chronic illness in adolescence. It could be argued that this concept of the influence of parents' attachment upon the attachment security of their children, via the internalisation of their attachment figures in the form of internal working models, intersects with the analogy of '*bridging the gap*' used in this study. Adult and childhood patterns of attachment and internal working models could be the most economical concept to explain the range of strategies used by the adolescents in this study in bridging the gap between normality and difference.

However, it should be noted that the connections made are speculative. As Rutter (1995) points out, though attachment theory has allowed for huge advances in the understanding of the development of relationships and of the associated difficulties which may arise, it must nevertheless be emphasised that attachments are clearly not the whole of relationships.

The proposed relationship of the three different attachment classifications will be explained in relation to the theoretical findings proposed in this study. The literature relating to attachment in adolescence specifically, though still in very much a formative state, will be incorporated to illustrate the links made as appropriate. The clear distinctions between classifications of attachment relationships and the speculated implications for CF and adherence to treatment are inevitably false to a point. Clearly, as with any relationship there are many shades of grey and perhaps more importantly, the patterns of attachment between parents and their children within any family may differ and may vary over time (Rutter 1995). However, in order to explain the speculative links coherently, within the scope of this project, the three basic distinctions, which are a common feature of the majority of the literature concerned with attachment theory, will be used.

### **The position of the securely attached CF sufferer**

#### ***Familial response***

If the perspective of attachment theory is taken it could be argued that the family of a securely attached child, which will be determined by the internal working models of the parents, will also be the family who are able to face the diagnosis of CF. They

will be able to steer between the two extremes of an avoidant response characterised by denial and minimisation, and an ambivalent, enmeshed response characterised by melodrama, hiding poor containment of fears and a sense of being overwhelmed by disaster.

It is proposed that the secure family will be able to recognise their own and the sufferer's fears and provide containment of these fears, through reassurance for themselves and the sufferer. Wright et al (1995), in a study of attachment security, found that although a clinical and normal control group of eight to twelve year-old children all expressed fears in relation to separation from their parents, the fears of the controls were readily "contained" (Winnicott 1971; Casement 1988 cited in Wright et al 1995) and rationalised by the children in their stories. In contrast, the fears of the clinical group, though similar in content, tended to escalate beyond the control of the children or adults, typically their parents.

It could be argued that if fears are contained, the secure family with a CF sufferer will be able to face facts and take on board the necessity for treatment in producing a good outcome, so making the worst feared scenarios less likely in the long run. The parents will be able to present a positive, self-directed approach to the future and will be experienced by the secure child as supportive, responsive and empathic (Wright et al 1995). This empathy and responsiveness is important as it enables the child to learn that his/her emotions are tolerable, shareable and will not overwhelm the parents (Hopkins 1991).

The secure family may be more likely to be proactively and collaboratively involved in the sufferer's treatment and to be perceived to be available to the sufferer at times of crisis. The sufferer will experience the family and significant others as being in tune with his/her needs. The individual will feel that the degree of difference is accepted, so allowing paradoxically for him/her to feel normal. When a mismatch occurs between the sufferer's needs and the responses of others he/she will be less likely to feel his/her balance is overly threatened by this, will be able to reflect upon the situation and empathise with the other's position. On the whole the secure sufferer will be the one who has a sense of security in his/her friendships and perceives a positive balance on the part of others between careful consideration and disregard for the CF. (see quote I12-13 p 56).

This model is then the one which becomes internalised by the sufferer. This has particular implications for adolescence as this is the period when the internalised

parents take over from the actual parents, to a greater degree, as the sufferer develops into adulthood. (see quote K21 p.59)

### ***Identity formation***

Ryan & Lynch (1989) in a study of the attachment relationships of adolescents and young adults suggested that a secure attachment facilitates internalisation and identity consolidation and that the parents acceptance and support, which characterises this style of interrelating, enables the adolescent to feel loved, have higher self esteem and to utilise parental support when required. The relationship between successful identity integration and security of attachment is supported by several studies of late adolescents (Kroger 1985; Lapsley et al 1990 cited in Rice 1990) though the literature to date has not considered the processes in achieving this through early and mid adolescence. However, Rice (1990) points out that the relationship of attachment status to identity formation is still unclear and speculates that the finding of an association may depend upon the measure of identity used. Rice (1990) also proposes that the relationship between attachment and identity development may become more pertinent at points of transition. In the context of this study it does seem reasonable to speculate about the impact of family style and relationships upon identity formation in terms of CF. Descriptions given in terms of family responsiveness and containment of fears seem to be related qualitatively to the stance taken in relation to CF in terms of identity and the management of transition by the sufferer.

The secure sufferer will, in the sense of this study, be the one who is most steadily balanced upon the bridge and will be able to bridge the gap between normality and difference most effectively. The sufferer will be able to accept CF as integral to his/her identity, accept that it places some restrictions upon his/her life, but not allow this to become overwhelming (see quote F16, 8 & D9 p.30). He/she will have the most optimistic outlook for the future. The treatment then becomes a normal part of daily life as the sufferer makes the best of a bad situation and takes control over those aspects of the disease which he/she can realistically influence. This places the sufferer in a better position to negotiate the demands of treatment with social and peer influences as they move through adolescence. Kobak & Sceery (1988) demonstrated greater problem solving abilities in adolescents assessed as secure using the Adult Attachment Interview (George, Kaplan & Main 1985).

Secure sufferers may also, perhaps as a result of a stronger self identity, be less likely to encounter threats to identity as harshly as their less secure fellow sufferers. The secure sufferer may be more likely to be the one who is able to disclose CF to the degree where the necessity to hide treatment is negated, as he/she will be able to take the view that others, apart from close peers, are not likely to be that interested anyway and that anyone who uses it as a weapon for torment is not worthy of the sufferer's positive regard. (see quote F18 p. 44).

The secure sufferer may, in accordance with this attitude, manage points of transition more smoothly as he/she is more likely to anticipate the support rather than rejection of others, particularly those who are considered to be close. This is not to say that the secure sufferer may not find disclosure threatening at times, perhaps especially in late adolescence, as more intimate relationships come to the forefront. These relationships are after all considered to be most intense, as from an attachment perspective they serve both basic human needs of protection and reproduction (Crittenden 1993). However, secure sufferers may be more likely to be able to handle their fears effectively and so be less likely to find themselves in danger of losing their balance upon the bridge. They could be more likely to be able to handle negative reactions well, problem solve around these and so in the end create a positive outcome. (see quote F22 p.47).

### ***Autonomy/independence***

Attachment is seen as a dynamic relationship which changes in accordance with the developmental tasks faced. Individuation happens with, rather than from, the parents. This is analogous to the concept of interdependence (Eiser 1993). Ryan & Lynch (1989) suggest that experiencing the parents as supportive and accepting as opposed to detached, is more conducive to the accomplishment of the tasks faced by the adolescent and young adult. This concept could be applied to the taking on of self care and treatment responsibility for the adolescent CF sufferer, as this is one of the key tasks of adolescence that must be mastered to ensure survival into adulthood.

The secure family may be more likely to take charge of the treatment in the earlier years of the sufferer's life, as they are able to take on the board the necessity of good adherence and maintain a balanced, though bearable, awareness of the implications of poor adherence. The secure family may also, accordingly, find it easier to relinquish control gradually to the sufferer as they develop. The sufferer is

more likely to have internalised the importance and value of treatment, and so is less likely to deviate from the acceptable norm established by the parents. This, in turn, may allow the parents to relax and feel they still have some influence whilst simultaneously allowing the sufferer to have some autonomy.

This is synonymous with the concept proposed by Bretherton (1987) that a successful attachment is one that permits, from an early stage, optimal autonomy within the context of optimal support. This is set in opposition to the notion that attachment is a symbiotic relationship from which the child must eventually emerge as a separate and differentiated individual. It could be argued that this distinction is considerably more important in the context of a serious chronic illness such as CF. Attachment theory proposes that although attachment behaviour becomes less obvious as children develop cognitive skills which enable them to cope with stressful situations, the natural response, even in adults at times of distress, is to seek proximity with attachment figures or to refer to internal representations of attachment figures (Bowlby 1988).

### ***Relationship with the medical care team***

The secure sufferer is more likely to apply this internal working model to the medical team, thus is more likely to describe a collaborative and supportive relationship with the physicians and the rest of the medical care team as he/she is able to take on board information about the disease and potential implications to his/her best advantage. Though the secure CF sufferer in adolescence may respond rebelliously to a lack of regard for his/her autonomy this may be more likely to be moderated by a balanced relationship of interdependence with the parents. The secure sufferer may be more likely to be able to negotiate his/her position successfully so that he/she feels at least some degree of participation in decisions regarding treatment and self care.

### ***Cognitive style***

In terms of cognitive style the secure CF sufferer may be more likely to confront issues relating to the future, manage the fear aroused by this and maintain a sense of control over his/her illness and treatment. He/she may be more likely to have internalised the capacity to contain fears through normative comparisons, humour, distraction, focus upon the positive aspects and hopes for the future and drawing upon available social support. (see quotes F26 & 27 p. 68) This assertion is

supported to some extent by Wright et al (1995) who observed that children in the non-clinical control group, when faced with fears related to separation from their parents, were able to generate a variety of coping strategies such as positive self talk, social support, reassurance and distraction. This was not true of those children in the clinical group considered to demonstrate avoidant or ambivalent attachment patterns. Principally the children in the control group demonstrated a sense of mastery over their experiences.

The CF sufferer who is able to gain a sense of mastery over his/her illness may be more likely to take control and have an internalised conviction of belief in the necessity and value of treatment. This may in effect become the sufferer's normality, so placing him/her in a position of optimal balance upon the bridge.

It could be speculated that the secure sufferer will be in such a strong and balanced position that even in the face of adversity, such as considerable deterioration, or occurrence of a distressing life event, though the position may become temporarily precarious the sufferer is, perhaps, less likely to place him/herself in jeopardy for any significant amount of time. The secure sufferer's sense of mastery may allow him/her to quickly regain his/her balance through new approaches to coping and problem solving or through drawing upon past experiences. (see quote D58 & F29 pp. 75, 76) Decisions regarding treatment therefore are more likely to be taken to the best advantage of the sufferer with a balanced view of the immediate and longer term consequences. The secure child, according to Crittenden (1993), is able to assimilate cognitive and affective information and revise his/her internal working models relatively rapidly. It could, as Crittenden (1993) suggests, be argued that this capacity is to some extent a precondition for flexibility in thinking and effective coping in the face of adversity.

Additionally, the secure sufferer may demonstrate a greater level of self awareness so enabling him/her to anticipate difficulties and so prepare for these to some extent. Main & Goldwyn (1985) state that the descriptions of early relationships given by the parents of securely attached children will be characterised by the capacity for self reflection. Shulman (1993) suggests that the adolescent whose parents are warm, supporting and encouraging of competencies tend to plan the course of action they wish to take to deal with stressful events.

Clearly, the cognitive style and coping strategies used will depend to some extent upon the developmental stage of the sufferer (Shulman 1993). Younger sufferers

may be more likely to rely upon a joint family approach to coping but as they approach adulthood these strategies may become more internalised, leading to a more autonomous cognitive style of dealing with CF, its treatment and associated fears. However, it is argued that at each stage of development security of attachment will be an organising variable.

### **The position of the avoidant sufferer**

#### ***Familial response***

Within the context of attachment theory it could be speculated that the family of the avoidant CF sufferer will lean far more heavily than the secure family towards denial of the seriousness of the condition and minimisation of the potential implications of the diagnosis. The sufferer and his/her family will be in a more precarious position upon the bridge, as this approach sways the balance too far in favour of normality, avoiding the necessary treatment.

The avoidant family may find the fears associated with the CF very difficult to face and so may deal with the situation almost as if it does not exist. Accordingly, the parents will be less effective in containing the sufferer's fears than the parents of the secure sufferer. The avoidant sufferer will therefore tend to view him/herself as all powerful and hide behind bravado and the pretence that he/she can cope and does not need the support of anyone else to find solutions to his/her predicament. (See quote J 43 p. 75)

The sufferer whose family show an avoidant response to the CF may, in the early stages, minimise the significance of the disease but then in the face of a crisis may be more likely to experience expressions of concern as intrusive, so exacerbating rather than placing a boundary around his/her fears. This may be particularly true when adverse circumstances, such as serious deterioration, enforce a confrontation with the avoided fears by the sufferer's family. (see quote A6 & 28 pp. 53). This may be too frightening to face so reinforcing the efficacy of an avoidant stance. The denial of any degree of difference may become stronger, reducing the likelihood of adherence to treatment. The sufferer and family lean further towards normality and so potentially take up a more precarious position upon the bridge.

The family may find it difficult to take on board the necessity for treatment and the longer term implications of poor adherence, potentially making it more likely that the

very scenarios they are trying to avoid become a reality. The necessity to maintain an air of normality will be stronger than for the sufferer in a secure family. This may mean that the sufferer will be more likely to experience others as being out of tune with his/her needs and to feel misjudged and misunderstood by the responses of significant others. This poor attunement may then serve to exacerbate feelings of difference which are difficult to tolerate, and so threaten the balance of the sufferer upon the bridge. The avoidant sufferer may feel greater degrees of anger towards unresponsive others, though this may not be outwardly expressed, and is less likely to empathise with their attempts to understand or to be able to take the perspective of others (see quote A14 p. 55).

Attachment theory would suggest that it is this model of avoidance and the expectation that the expression of need or emotions such as fear, anger or distress will be ignored or greeted with rejection which becomes internalised as the sufferer approaches adulthood. This may then, due to the avoidant stance taken in relation to the CF, lead to a greater likelihood of serious deterioration earlier than necessary.

### ***Identity formation***

Ryan & Lynch (1989) suggest that adolescents and young adults with poorer attachments experience their parents as less loving and accepting than those with secure attachments. They tend to be less willing to rely upon parental support at times of stress, despite denying this and idealising their parents. It could be hypothesised that the CF sufferer in these circumstances will try to pretend that CF is not a part of his/her identity and the implications of the condition will be integrated into the identity in a very vague way (see quote A13 p. 32). The sufferer may then literally be more likely to forget aspects of the treatment regime. Treatment serves as a constant reminder of the very things he/she finds it difficult to face, thus the individual is less able to effectively take control over the disease.

Insecurely attached adolescents have poorer problem solving skills (Kobak & Sceery 1988), suggesting that perhaps the avoidant CF sufferer will have more difficulty in balancing the conflicting demands of peers, family and treatment requirements in adolescence. The sufferer's relationships may be more characterised by fears of rejection (Ryan & Lynch 1989; Kobak & Sceery 1988), making attempts to hide the difference more likely so as to maintain the pretence of normality and reduce the likelihood of rejection, or misjudgements regarding his/her character. The avoidant sufferer is perhaps in accordance with these fears more

likely to find points of transition difficult and to react to threats to his/her identity with feelings of anxiety, distress or anger. (see quotes A23 & A15 p. 38). Kobak & Sceery (1988) found higher ratings of anger, anxiety and distress in difficult social situations in those adolescents classified as dismissing, the equivalent of avoidant, using the Adult Attachment Interview (George, Kaplan & Main 1985).

The avoidant sufferer may therefore adhere less closely to the treatment regime and may find it more difficult to take a proactive stance in the face of deterioration or anxiety provoking social situations related to the disease. In the face of threat he/she may be more likely to try to handle fears by further avoidance, in effect working harder to deny the difference to any degree, so leaning even further towards normality.

### ***Autonomy/independence***

If, as Ryan & Lynch (1989) suggest, experiencing the parents as detached in adolescence is less conducive to mastery of the tasks of this life stage it could be hypothesised that the avoidant CF sufferer will find it more difficult to take on responsibility for his/her treatment and self care effectively. There may be more likely to be a swing from dependence to independence, the traditional split which has been demonstrated to have negative effects upon adherence (Pownceby 1996; Gudas et al 1991).

The avoidant family may be less likely to take charge of the treatment in the early stages of the sufferer's life as to do this requires the facing and dealing with the potential implications of the disease for the future. This may mean that either the parents or the sufferer are in charge, rather than there being a collaborative approach to treatment. ( see quote A43 p. 58). If the parents take charge of the treatment in an authoritarian manner this may mean the value of treatment is instilled in terms of the negative consequences of missing it, rather than in terms of its intrinsic value. (see quotes G25-27, J12-13, J16 pp. 59,60).

Ryan & Lynch (1989) suggest that insecurely attached adolescents are less likely to rely on their parents for support. Kobak & Sceery ( 1988) found that avoidant adolescents who were dismissing of the value of attachments manifest higher levels of hostility in close relationships, including those with peers. These adolescents are more likely to seek support outside of the family, within the peer group, but are less likely to be successful in utilising the available support to their best advantage due

to fears of rejection (Ryan & Lynch 1989). This type of situation could therefore lead to the scenario of optimal autonomy within the context of minimal support, as opposed to within the context of optimal support, the criteria for a successful attachment proposed by Bretherton (1987).

It could, therefore, be hypothesised that avoidant adolescent CF sufferers will be the those who use the CF treatment as the battle ground to assert their independence from their parents. They may be the ones who are more heavily influenced by the peer group, but the strong drive for acceptance, driven by underlying anticipation of rejection as opposed to support, will make disclosure of the disease more threatening, attempts to hide the treatment more likely and accordingly reduce the likelihood of adherence. The sufferer is again more likely to be placed in a more precarious position on the bridge and may lack some of the essential support required to redress the balance.

### ***Relationship with the medical care team***

The avoidant sufferer and his/her family may find it more difficult to take on board information regarding the disease. The blatant pointing out of the potential dangers of poor adherence and the differences for the sufferer may be experienced as highly threatening. The distress related to this threat may not be immediately obvious as the avoidant sufferer, according to the predictions of attachment theory, will tend not to show his/her fears until he/she is on the brink of disaster. The sufferer and family may also be more likely to counteract the threat with a withdrawal further into avoidance, rather than being spurred on to increase efforts in relation to treatment. The sufferer may be less able to problem solve and may find it more difficult to negotiate his/her position so that he/she can feel actively involved in decisions relating to medical care. He/she may instead assume a position more characterised by denial of medical opinion and take complete control. Ironically, this type of drive for complete self reliance and independence may be more likely to result in hospital admission so enforcing dependency.

### ***Cognitive style***

The avoidant sufferer may tend to minimise the potential implications of the disease for the future and may, as a result, only be able to give an impoverished picture of future plans and aspirations (see quote C75 p. 69). The sufferer may have internalised a model which means he/she finds fears and emotions associated with

the future difficult to contain and will have few strategies to deal with these emotions apart from passivity and denial. Wright et al (1995) observed this phenomenon in children with insecure attachments who demonstrated a lack of, or inappropriate, coping strategies when relating fears of being separated from their parents. The avoidant CF sufferer will be less likely to gain a sense of mastery over the CF and may internalise the belief that treatment has only limited value, almost to the extent that the individual believes he/she can survive without it, so leaving the rest of the prognosis up to fate.

In the face of distress the sufferer may end up in a particularly precarious position as he/she may have a reduced self awareness and will be less able to anticipate difficulties (see quote G32 p. 72). Also, poor recall (Kobak & Sceery 1988) and a past stance of avoidance may mean he/she has less sense of being able to draw on strategies which led to a successful outcome in similar circumstances, so perpetuating poor coping resources. Serious deterioration may lead to a temporary increase in the sufferer's sense of control. Since this increase is motivated out of fear which is likely to be poorly contained, motivation to take control may be more likely to subside as the sufferer, who may find it more difficult to alter their internal working model accordingly (Crittenden 1993), has to fall back on the old avoidant coping strategies.

### **The position of the ambivalent sufferer**

The implications for treatment adherence for the ambivalent CF sufferer may be similar to those of the avoidant sufferer in that the final outcome may be poor adherence. However the path taken to arrive at this outcome is qualitatively different.

### ***Familial response***

It could be hypothesised from an attachment perspective that the ambivalent family will experience the CF in relatively catastrophic terms. The family may find it difficult to contain their own and the sufferer's fears as regards CF, but rather than simply avoid the issues they will also at times be overwhelmed by their emotions.

Attachment theory would predict that the family will find it difficult to form a coherent story about the illness and its implications and will oscillate between minimisation and the belief that everything is fine to the expectation of disaster.

Concern may be more likely within this context to be expressed in terms of sympathy which, rather than containing the sufferer's fears, conveys the impression that something awful is about to happen, so promoting heightened anxiety (see quote B12 p. 51). Wright et al (1995) observed that children in the clinical group described fears associated with separation from their parents which became out of control and ended badly.

Kobak & Sceery (1989) found that those adolescents and young adults who were classed as having a preoccupied or enmeshed working model of attachment (ambivalent pattern), using the Adult Attachment Interview (George, Kaplan & Main 1985), experienced their families as more supportive than those who were dismissing of attachments (avoidant pattern). They were rated by peers as being more anxious and reported higher levels of distress.

These heightened levels of emotional expression may lead to the sufferer, in an ambivalent family, feeling that he/she is the reason for the family remaining in existence as a unit and may be more likely to perceive him/herself as a burden to his/her parents, despite also being acutely aware that his/her parents care. This sense of burden may have a negative impact upon adherence as the will to be normal, so relieving everyone of the burden, may be stronger, reducing the likelihood of optimal levels of adherence. The sufferer may be more prone to blame others and feel it is unfair that he/she is different. Accordingly he/she may never accept the condition and make the best of it (see quote B35-37 p. 54 & B15 p. 55). In the context of the analogy used in this study the response of the family often leans heavily towards difference but this is a very difficult position to sustain and the sufferer has to swing back in the direction of normality in order to compensate and maintain a sense of sanity.

The gravity of the perception of difference may make it less likely that the sufferer can even conceive of others understanding his/her predicament so in the extreme enhancing the sense of isolation and insecurity. The sufferer may however have a sense of his/her parents as a potential source of support and so crave more attunement with others. Kobak & Sceery (1988) suggest that the preoccupation of ambivalent adolescents with attachment relationships may lead them to cope with high levels of anxiety by dependent or clinging relationships.

### ***Identity formation***

It could be hypothesised that the ambivalent CF sufferer will form an identity which centres around, and is overwhelmed by CF. The model of catastrophe will be internalised and CF will accordingly be considered to impact upon every aspect of the sufferer's life. The sufferer leans heavily towards difference and may feel that his/her identity expression would be dramatically different if he/she did not have CF (see quote E28-30 p. 31).

Kobak & Sceery (1988) suggest adolescents with enmeshed/preoccupied models of attachment tend to demonstrate less ego resilience than their secure counterparts and are less self assured in relationships. In the context of this study the ambivalent sufferer could be said to have a relatively fragile sense of identity which can easily be threatened. Accordingly the ambivalent sufferer, like the avoidant sufferer, may then try particularly hard to conceal the disease so making adherence more of an effort in order to fight against the sense of difference and bolster his/her identity (see quote B53 p. 41). The slightest perception of difference may be overwhelming to the sufferer and so pose a threat to his/her existence in the psychic sense. The assumption is made that as the sufferer finds the CF so overwhelming this will be the reaction of others to whom a disclosure is made. The sufferer may therefore crave the empathy of others, yet at the same time fear that entering into relationships, from which he/she may gain empathy, will result in rejection when the expectation that the other person will be overwhelmed is realised.

### ***Autonomy/independence***

In accordance with the suggestion of Ryan & Lynch (1989) that insecure attachments are less likely to effectively facilitate accomplishment of the tasks of adolescence, it could be hypothesised that, like the avoidant CF sufferer, the ambivalent sufferer will also find it difficult to take on board responsibility for the treatment. The burden of taking on the responsibility of treatment may be perceived as overwhelming for the sufferer leading to a swing back in the direction of a pretence of almost total normality, so reducing adherence. This may then enforce a reminder of the difference as the sufferer may become seriously ill. This recognition of the reality of the disease may then throw the sufferer straight back towards difference and the overwhelming nature of the associated fears. The sufferer is in effect oscillating between normality and difference and finds it very difficult to assume a position of balance (see quotes E76 & E13 pp. 34, 35).

Rather than rapidly becoming independent however, the sufferer may fluctuate to a greater extent than secure adolescents between dependency and independence in an attempt to gain the support he/she craves. The sufferer craves relationships but at the same time feels very threatened by them, so never really gets what he/she wants (see quote E16 & E93 pp. 45, 48). The sufferer may tend to become angry with others and accordingly, be rejected by or reject others. This could lead to a situation of minimal autonomy in the context of optimal support, interchanged with optimal autonomy and minimal support, as opposed to the desired situation of optimal autonomy in the context of optimal support (Bretherton 1987). The sufferer's internal working model is derived from patterns of intermittent and unpredictable support, perpetuating the swaying of the bridge as the sufferer oscillates from one extreme to the other.

### ***Relationship with the medical care team***

Like the avoidant sufferer, the ambivalent sufferer may also find it more difficult to problem solve and so negotiate a desirable degree of autonomy as regards decisions relating to his/her care. The sufferer will find negative information overwhelming. Therefore he/she may be predisposed to remember this in a confused and contradictory manner making it difficult for him/her to form a general perspective as regards the CF and the treatment. The sufferer may oscillate markedly between wanting to be dependent upon the medical care team to wanting almost total independence. When the team are not able to keep up with this flip between these two extremes the sufferer will be likely to become angry, and to make the team angry, by his/her oscillations.

### ***Cognitive style***

The ambivalent CF sufferer may find it very difficult to form an overall perspective as regards CF and the value of treatment. Like the avoidant sufferer, he/she may have very few internalised strategies for dealing with difficult situations and emotions related to the disease. Accordingly, it could be speculated that he/she too will experience difficulty in gaining a sense of mastery and control over the disease. The sufferer may oscillate between the passive responses of hope and despair but have little sense of what flips him/her between the two states. The sufferer will be unable to draw conclusions and may drown in feeling states to which he/she can assign little cognitive coherence. Motivation for treatment may then vary accordingly.

Crittenden (1993) suggests that ambivalent children will take more risks in an attempt to gain the support of their attachment figures and will increase the stakes until they attain the desired response. It could, therefore, be hypothesised that the adolescent CF sufferer may reach more risky decisions regarding treatment than his/her secure or avoidant counterparts in the quest for normality and for support. This could be counterproductive in the long run as the sufferer may decide to risk behaving as if he/she were totally normal and miss treatment, in the extreme case, altogether. This may then have the effect of the sufferer becoming ill and so again overwhelm him/her with the difference. Essentially, the cognitive style of the sufferer may serve to maintain the oscillating position from normality to difference upon the bridge. In the face of difficulties, if the bridge then too sways, the sufferer is in danger of being flung into the abyss below (see quote E76 p. 34).

Crittenden (1993) suggests that ambivalent children have learned that the expression of affect has the desired outcome, whereas cognition is misleading. The ambivalent child tends in mental processing to emphasise affect and disregard or defend against cognition. It could, therefore, be speculated that if the CF sufferer who is ambivalent survives a crisis he/she will remember acutely the associated emotions, but will have little sense of the associated thought processes and of how he/she came to survive (see quote E77-79 p. 74). The sufferer may have a stronger sense of the link between health status and inner feeling states, feeling anxious and depressed when unwell and on top of the world when in good health. He/she may have very little sense of control over the illness and so little sense of how he/she maintains any kind of balance upon the bridge (see quotes E67 & 73-74 p. 74).

## LIMITATIONS

Although this study has added new perspectives to the understanding of adherence to treatment for CF in adolescence, and possibly raised issues which could be applied to adherence within the context of other chronic illnesses during this life stage, there are a number of limitations to which attention must be drawn before applying the theoretical perspective generated. Each of these limitations will be discussed in terms of the implications for the completeness of the theory generated, and the potential for the applicability of the theory within particular contexts. It remains a theory building study generating hypotheses rather than conclusions.

### The sample

The sample of CF sufferers from whom data was gathered in this study has one main limitation in that the majority of those who took part were either in early or late adolescence. The sample included only two female and one male in middle adolescence, aged thirteen to fifteen, at the time of interview. This means therefore that the perspective of middle adolescents is incomplete. Attempts were made to sample from this age range more thoroughly but practicalities dictated that this was not possible within the time constraints of this study. This age group were, in any case, fewer in number within the population available and seemed more reluctant to devote more time to their disease, making them a difficult population to access.

This phase of adolescence is thought to be a time dominated particularly by increasing independence (Elliott & Feldman 1990). If more data had been obtained from middle adolescents this may have generated a greater wealth of phenomena relating particularly to the subcategory '*degree of autonomy*' (pp. 58-60) and possibly in terms of variation in cognitive style. Czajowski and Koocher (1987) found that coping styles in this age range were more influenced by planning for the future than at other stages of adolescence. However, the analysis of the data collected does have enough depth and density to add to understanding of the impact of increasing autonomy and independence and cognitive style upon adherence to treatment. As Strauss & Corbin (1990) argue, sampling within the grounded theory method is carried out on the basis of incidents rather than persons, therefore the theory generated here is by no means negated by the content of the sample either in terms of numbers or distribution across the age range.

The sample did include, through the use of theoretical sampling, sufferers with a wide range of severity of disease and who were perceived by members of the clinical team to be adhering to varying degrees to the treatment regime. Accordingly a range of family styles of dealing with CF and treatment were also revealed. However a greater saturation of categories may have been possible if it had been possible to include more sufferers in mid adolescence.

### **Saturation of categories**

Strauss & Corbin (1990) propose that in a grounded theory study sampling should be continued until theoretical saturation is reached. Essentially this means that all the categories are conceptually complete and that further sampling would not be likely to add to the explanatory power of the proposed theory.

Within this study it was not possible to achieve this aim due to practicalities and time constraints. The category of *'the role of others in bridging the gap'* (pp. 49-63) could conceivably be expanded by further sampling, focusing on family interaction, particularly in the light of the consideration of the links of the theory generated to attachment theory. This may be most effectively achieved through interviews with parents in addition to adolescents. Attempts were made, within this study, to delve more into family issues. Though this could potentially have been more successful, with revision of the questions asked, there was resistance on the part of some sufferers to discuss the relevant issues. This may be a facet of adolescent egocentrism.

Similarly, the interface of relationships within the family and those with the medical care team could potentially have been more fully conceptualised. This could potentially have been achieved by sampling from various members of the team and focusing on their perceptions of relationships with the sufferers and the perceived impact of these relationships upon adherence. Nevertheless it seems reasonable to suggest that enough conceptual density has been achieved to make the theory proposed useful and applicable to CF sufferers, their families and the clinical team responsible for their care.

It could be argued that the whole premise of saturation of categories is questionable. If the constructivist position is based upon the notion of people constructing their own realities within social, cultural, historical and political contexts (Henwood & Nicholson 1995), then the range of possibilities are surely infinite. This

means that it could be strongly argued that to ever consider a category to be complete is to make a false assumption. If the researcher searches hard enough, is able to keep an open mind and maintain enough theoretical sensitivity, then within the constructivist framework it is inevitable that more properties, dimensions, complexities and intricacies pertaining to the phenomenon under investigation will arise. If this stance is taken then it seems feasible to suggest that the concept of percentage of saturation proposed by Glaser & Strauss (1967) is a hark back to the positivistic roots of psychology. Interestingly Charmaz (1990) talks only of conceptual density, not saturation of categories.

### **Respondent validity**

Respondent validity has been proposed as one way of checking the validity of qualitative research (Reason & Rowan 1981). It has been suggested in order to enhance validity, and improve the quality of research accordingly, the findings should be taken back to the respondents and refined in accordance with their responses to the theory proposed.

This method was not employed in this study, partly due to time constraints, but more importantly because this could, within the context of this study, be considered to be extremely unethical. The research was conducted within a clinical context and the presenting back of the findings to those who took part would, in the light of the interpretations made, be extremely confrontational and possibly very painful for those concerned. The addressing of the issues raised would necessarily in itself constitute a clinical intervention, which would require any number of therapeutic sessions, in order to convey the essence of the theory proposed in a manner which would minimise the risk to the mental health of those concerned.

Silverman (1994) takes the view that the findings of studies such as this one are written for a professional audience, in this case in the first instance for clinical psychologists, and in the second instance for members of the medical care team. The aim of the analysis is to add a layer of understanding which makes sense to those puzzling with the phenomenon under investigation, but involves interpretation beyond the descriptions of the respondents themselves. Also, as Henwood & Pidgeon (1995) point out, from the constructivist position reality cannot be mirrored no matter how grounded the account given is in the experiences of the respondents. Validity therefore cannot be established simply by presenting back to the respondents, as this process too is subject to interpretation.

In a sense, validation of this theory could be achieved more ethically, by presentation to the medical care team responsible for the participants and to a group of clinical psychologists working in the paediatric setting. It is intended that this be carried out, at least informally but again practicalities did not allow for this within the time scale of this project. The application of the findings in the clinical setting on a day to day basis, by a clinical psychologist, and through other members of the team, will in itself provide some verification or disqualification of the theory proposed.

### **Reflexivity**

Depending upon the stance taken, this study could be criticised as not being as rigorously inductive as Glaser & Strauss (1967) claimed a grounded theory study should be. However, as Strauss & Corbin (1994) point out this stance has since been revised, and the grounded theory method has been modified to view the researchers training, reading, experience and theoretical stance as potentially useful, if constantly and systematically compared against the gathered data and the theoretical perspectives emerging from the data.

The findings of this study were clearly influenced in this way by the experiences of the researcher, through reading of the relevant literature from the outset, as presented in the introduction. This reading then continued throughout the analysis as seemingly important issues emerged and data collection and the interpretation of this were influenced accordingly. As the researcher was also working closely within the Cystic Fibrosis clinic from which the sample of participants was obtained data collection and analysis was also influenced by experiences within the clinic setting and issues raised as potentially important by the clinic team.

Finally, the whole research process was, in the opinion of the researcher, unavoidably influenced by the researcher's constructions of the phenomena of adherence, adolescence and chronic illness. These inevitably took account of past clinical experience and theoretical orientation. The analysis presented therefore represents one particular reading of the data, which in and of itself, is open to many different interpretations.

Criticism could be levelled that the researcher merely sets out to confirm existing preconceptions. The study aimed to avoid this pitfall and address this issue by taking the stance of Charmaz (1990) and maintaining an awareness of these

preconceptions from the beginning. Henwood & Pidgeon (1995) argue that if this stance is taken within grounded theory it encourages the researcher to remain on an analytic path, whilst retaining an acute awareness of the potential pitfall of simply reproducing his/her preconceived ideas in an unrevised format. Strauss & Corbin (1994) state that the rigorous coding procedures such as constant comparison, theoretical sampling and the development of concepts through the paradigm model, enable the researcher to continually question his/her own perspectives. Henwood & Pidgeon (1995) point out that although subjectivity and interpretation is in the forefront of qualitative research, whichever methodology is chosen, this an issue which pertains to any form of science in practice. Objectivity, in a sense, could be said to be a representation of deficits in knowledge and understanding.

### **Generalisability**

It could be argued that the findings of this study are not generalisable, as they were generated by theoretical sampling from one particular population of CF sufferers. However, Lincoln & Guba (1985) point out that the question of the transferability of the findings of any qualitative study should be judged according to the fit of the findings to other situations. They suggest the responsibility for establishing this fit lies with the person who wishes to use and interpret the findings. This seems a reasonable assertion to make in terms of this study, as the findings are described primarily for an audience of practising clinicians, who have extensive training and experience in deciding which theoretical viewpoint could effectively be applied in specific sets of circumstances. However, as Charmaz (1995) argues, grounded theory works do have greater generalisability when conducted at multiple sites, however the building of a theory on this scale was not feasible within the time frame of this study.

Glaser & Strauss (1967); Strauss & Corbin (1994) argue this point in terms of claiming that if the theory is adequately grounded in the data, it will have a close fit to the substantive area to which it applies and this fit will determine the usefulness of the theory. Strauss & Corbin (1994) do point out however that the shelf life of any grounded theory is inevitably time limited, as it is embedded in the history of the moments, eras and circumstances within which it is created. The theories thus require reformulation and revision in accordance with changing times. In the context of this study, for example, the issues may differ for the next generation of adolescent CF sufferers as they will derive benefit from advances in medical technology and, accordingly, have a longer life expectancy. Also, changes in service

provision, as a result of theorising about where improvements could be made, will inevitably impact upon their experiences.

Within this stance, providing an appropriate degree of caution is exercised which takes account of the limitations described, the findings of this study could potentially be applied by those grappling with issues pertaining to adherence to treatment in adolescent CF sufferers in other CF centres. The findings may also prove useful for those confronted with similar issues in dealing with adolescent chronic illness sufferers in general, perhaps particularly those illnesses which, like CF, are terminal and are genetic, or at least congenital in origin. Awareness of this latter point may have particular relevance in terms of using and interpreting the aspects of the theory proposed in this study which pertain to identity formation. It could be speculated that the implications for this may be very different for a chronic illness with a later onset and diagnosis and a lesser likelihood of premature death.

## **CLINICAL IMPLICATIONS**

The initial request for this study was driven by the desire to find out what the relevant issues may be for intervention in relation to adherence, prior to a clinical psychologist beginning specialist work within the clinic where the study was based. The theoretical perspective taken in this study has several potential implications for intervention, the outcome of which may serve to enhance or dispute the validity of the findings.

In terms of adherence, interventions made by staff could be enhanced by taking account of the need of the adolescent sufferer to bridge the gap between normality and difference effectively. The principle feature of this may be to keep respect for the desire for normality and the fundamental importance of this for the sufferers to the forefront, when presenting and dealing with issues relating to treatment, so as to avoid threatening their identities, and so jeopardising their balance upon the bridge, as far as is possible.

To a large extent, this is the style taken within the clinic at present, though the impact of different individual and family styles or attachments upon the sensitivity of the issue of difference, is perhaps not always fully understood and recognised. This is not surprising given the fluctuating needs and demands of some sufferers during adolescence. However, if there were more acknowledgement of the requirements for balancing and juggling on the part of sufferers and their families, as a result of the acquisition of a more in depth understanding, this may serve to enhance relationships with the clinical team and so increase adherence. The analogy of the bridge, and the links made between this and the theoretical framework offered by attachment theory, may facilitate this.

In view of the fluctuation in need, it may be beneficial to arrange for groups of adolescents to attend the same clinic to avoid the situation where the clinic team are having to switch very quickly between models of dealing with young children and their parents, to approaches more suited to dealing with adolescents, in particular those in late adolescence. The setting up of the adult service on the same premises, plans for which are already in progress, may somewhat aid the smoothness of the transition from child to adult services.

The suggestions made for linking the analogy of bridging the gap to attachment theory could perhaps explain the reasons for great success in dealing with some

sufferers and the difficulties experienced with others. At present it seems that the model within which the clinic operates works extremely well for 'secure' sufferers. It seems likely that a diagnosis of CF may precipitate the development of 'insecure' attachments in some families who otherwise might have managed very well. This suggests that facilitating the early adjustment of parents, in the face of diagnosis, may avert difficulties at a later stage. The modelling and suggesting, by staff, of a middle road to parents with regard to treatment, may lead to more effective coping with the illness. This should involve talking openly and honestly with parents and children in a containing way.

The model presently used by the CF nurses contributes to this as it involves early intervention and trouble shooting. However, the efficacy of such intervention may be enhanced by a greater understanding of the difficulties in taking on board and adjusting to the disease in those parents who perhaps have less secure internal working models of attachments. Intervention with these families and sufferers will inevitably take more time. It can be very tempting for the doctors and other key members of staff to try to tackle clearly avoidant sufferers and their families with scare tactics, in an attempt to save them from the harmful effects of poor adherence. However with the avoidant and ambivalent families and sufferers this is likely to backfire.

It may be helpful to draw up guidelines, relating to the different family attachments and approaches, for maintaining balance on the bridge, to enable members of the clinical team to recognise and deal with these patterns when they occur, and to enable them to help parents to change. This may make it easier to avoid the pitfall of either being drawn too far into the family's pattern of interaction, or trying too hard to counteract it to negative effect. In accordance with attachment theory the clinic team could be encouraged to act as a secure base for those tending towards avoidant and ambivalent attachments early on, and at points of crisis, by handling and containing the fears of the parents. This may enable the parents to realise that if they can confront and contain the issues on behalf of their children, this may provide a better and less frightening prognosis for the future.

In some cases, where there is not time for the issues to be dealt with satisfactorily, or they perhaps seem too complex, a clinical psychologist or trained therapist could provide the secure base through work with the family. Heard (1982) and Byng-Hall (1991) suggest that the family therapist, in the context of attachment theory, could be construed as an 'assuaging caregiver' for all family members, thereby reducing

anxiety and fear and enabling family members not to be drawn into patterns of interacting and dealing with difficulty, which they may use in situations outside of therapy. This in relation to CF, may allow the family to become more exploratory and containing in their approach to the disease and their associated fears.

In terms of those sufferers who are already experiencing difficulty, it may require a joint approach of all staff members in holding the fears for the person. When necessary, engaging the individual in therapy aimed at providing a temporary secure base, allowing the exploration of issues of identity and the development of more cognitive skills to enable more effective coping with the disease, may be beneficial. Pearce & Pezzot-Pearce (1994) describe a therapeutic process whereby the therapist forms a secure base for the child, and through the non-rejecting and sensitive responses to the child's emotions and dependency needs, enables the child to reconstruct his/her working model of attachment. This rationale, on the basis of the theory presented in this study, could be applied when necessary to the CF sufferer struggling to maintain his/her balance on the bridge.

Clearly, the preferred approach would be to identify those who may have difficulties in adolescence prior to them entering this life stage and equip the sufferers and their families with the skills to cope proactively. Identification could be carried out by discussion with the clinical team, individual and family interviews, and possibly the routine administration of the Separation Anxiety Test (Klagsbrun & Bowlby 1976) in children approaching adolescence. Wright et al (1995) applied this test as a measure of internal working models of attachment in middle childhood to a clinical sample. Although it is suggested that further development and testing of this instrument be undertaken before it can be reliably used with this age range in clinical setting, the responses did demonstrate it to be a potentially valuable tool on a qualitative basis. This instrument could therefore be applied to good effect, within the context of CF, in combination with the findings of this study, to give further guidance as to which sufferers may experience particular difficulties in terms of treatment adherence in adolescence.

The current emphasis, amongst the clinic team, on early intervention may already be exerting positive influences upon adherence. If the depth of understanding driving this is developed, through use of the analogy of bridging the gap and the relationship of this to attachments, this may serve to further enhance any positive influences.

When considering those adolescents already facing difficulties, the ideas could be used to enable them to stabilise their position on the bridge. If a clinical psychologist were present in the clinic routinely, as is the proposed strategy, this may enable him/her to get to know the children, adolescents and their families, so enhancing the efficacy of the application of these findings. If the findings are applied systematically this may also facilitate the development of research in this area.

# **APPENDIX i**

## **Information sheet**

## **Living with Cystic Fibrosis: The Children and Teenagers' Perspective**

### **What is this study about?**

I am interested in finding out how young people who suffer from cystic fibrosis feel about and cope with the illness and it's treatment.

### **What do I have to do?**

If you agree to take part in the study you will be asked to go along to an informal talk with myself (the researcher) when you next visit the cystic fibrosis clinic at the Children's Hospital in Sheffield. The talk will last for at the most an hour and you will be asked some questions about cystic fibrosis, the treatment you have and how you feel about living with your illness. If you prefer not to talk at the clinic but would like to take part, arrangements can be made to meet at another time or place.

With your permission I would like to tape record the interview so that I will be able to remember what we have talked about correctly.

### **Will anyone else find out what I say?**

No, no one else but myself will have access to the tapes and no-one who takes part in the study will be identified. Nothing you say will be passed on to your family. The staff at the clinic who provide your care will not know what you have talked about and nothing you say will affect your care or treatment. All the tapes will be erased once the study is finished.

### **Can I change my mind?**

Yes. Even if you agree to take part you are free to change your mind at any time without saying why.

### **What happens afterwards?**

I will collect all the information from the interviews and then write a report which will be used by your medical care team to try to improve services for young people who suffer from cystic fibrosis. If you agree, I may also need to look at your medical notes kept by the doctors at the clinic to see how your treatment is going.

If at any time after our talk you feel you would like to talk some more to someone I would be happy to arrange this for you.

If you think you might like to take part but would like to know more about the study before you agree please give Sarah Longley a ring on 0114 271 7296.

## **APPENDIX ii**

### **Consent form for participants**

**CONSENT FORM**

**Living with Cystic Fibrosis: The Children and Teenagers Perspective**

**Please complete and sign this sheet.**

I have read the information sheet provided Yes/No

I have had the chance to ask questions about this study. Yes/No

I am happy with the information given to me about the study. Yes/No

I understand that I am free to change my mind at any time, without saying why and without this making any difference to my medical care. Yes/No

I agree to take part in this study Yes/No

Signed.....Date.....

NAME IN BLOCK LETTERS.....

**Thank you for your help**

## **APPENDIX iii**

### **Consent form for parents**

**CONSENT FORM**

**Living with Cystic Fibrosis: The Children and Teenagers Perspective**

**Please complete and sign this sheet.**

I have read the information sheet provided Yes/No

I have had the chance to ask questions about this study. Yes/No

I am happy with the information given to me about the study. Yes/No

I understand that myself and my child are free to change our minds at any time, without saying why and without this making any difference to my child's medical care. Yes/No

I agree to taking part in this study. Yes/No

I agree to my child taking part in this study Yes/No

Signed.....Date.....

NAME IN BLOCK LETTERS.....

**Thank you for your help**

**APPENDIX iv**  
**Interview schedule**

## INTERVIEW SCHEDULE

Each child will be seen individually and before starting the setting will be explained and they will be reminded of the purpose of the study, confidentiality and their right to withdraw at any stage or opt not to complete any of the tasks or answer any particular questions.

### 1. Family Sculpt Task.

In order to complete this task the child will be given a wooden board divided into squares and a number of wooden figures, large ones to represent adults and small ones to represent children and some wooden blocks of various heights. They will then be asked to place the figures on the board so as 'to make a picture of their family'. They will be told that the picture should show how close everyone feels to each other and that the blocks can be used to elevate the powerful people in the family. the 'picture' will then be recorded on a standardised scoring sheet.

### 2. Bag of feelings.

This task involves the child drawing with the help of the researcher a picture of an imaginary bag which contains all their feelings. The task will be introduced in the following way: 'to help me to get to know you a little bit and to understand how you are feeling about things at the moment I wonder if you'd draw a picture with me (drawing a bag on a piece of paper). If this was a bag of all the feelings inside you I wonder what would be inside there.' if the child cannot generate any feelings to start with the researcher may prompt them by commenting on a feeling they have observed e.g. 'you're looking a bit fed up.' The child will then be asked to select a colour for each of the feelings as they are talked about and fill in the proportion of the bag that is full of that feeling. The wording and approach to the task will be varied to suit each individual child. The child is then prompted for each feeling, 'what kind of things make you feel X?; What/who is helpful to you when you feel X?'

This will then lead into a semi-structured interview designed to elicit how the child perceives and copes with their illness and its treatment and their view of how CF affects their life along the following lines.

Questions may be expanded by:- Why is that?

Could you tell me more?

What do you mean by that?

## **General feelings**

What are the three things that you spend most time thinking about at the moment?

Supplement with, any good things/any bad things if all three good or all bad

Space to comment on how much CF does/does not feature in the above

What is it like living with CF?

What does it mean to you to have CF?

Do you think anybody understands what it feels like to have CF? In what way do they or don't they understand?

What advice would you give to someone younger than you who has CF?

What are the worst things about having CF?

Are there any things you'd miss or think you might lose out on if you didn't have CF?

How do you think your life would be different if you didn't have CF? For family; friends, school work, interests, aspirations....

What do you think might make living with CF easier?

## **Other people's perceptions**

How do you think other people feel about you having CF?

Do you always let other people know you have CF? In what sort of situations /with which people do you try to hide it?

What do you think people might say/think if they knew you had CF?

Does having CF make a difference to the way people treat you/ the things they expect of you?

## **Treatment**

What treatments do you have/ does it involve? details re frequency, type etc. (fall back if struggling otherwise details not important)

What bits of the treatment cause you the most hassle?

" " " " " " " " least hassle?

Do you ever feel like missing bits of it?

When do you feel like that ?/ What sorts of things make you feel like that?

What do you do when you feel like that?

Who do you go to when you feel like that? What do they say/do?

Does it make any difference what they do or say?

Does anyone help you with the treatment?/ Make sure you take your tablets, have physio get plenty of exercise...?

What difference do you think it makes if you stick to the treatments as you are supposed to or not? Now and in the future?

What things do you think make a difference to how well/ill you are at different times?

### **Coping**

Can you think of a time in the past year when having CF has caused you a problem? Made you feel sad, scared, bad .... when things haven't gone the way you wanted them to?

How did that affect you? What did you do/think when that happened? Tell me all the things you thought/did? Why do you think things went that way?

What are the ways in which you tried to solve the problem? prompt for more than one way if possible

How did you think that would help?

What effect did it actually have, how did it or didn't it help?

What difference did it make to how you might handle a similar situation in the future?

Would you do anything the same or differently in a similar situation in the future?

What advice might you give to someone else facing a similar difficulty?

### **Clinic Visits**

How often do you usually come to the clinic? imposed by disease

How do you feel after you've been to the clinic if you are told things are going well/ if things are not going so well?

What do you do afterwards if the outcome is good/bad?

What difference does this make to how you feel about the treatment?

### **Future**

How different was it living with CF when you were younger?

Why is that?

In what way do you think things might be different when you are older?

Why is that?

What has been the hardest time for you so far? Do you think it will get easier/harder as you get older?

What would you like to do in the future? job/education, where like to live, who with.

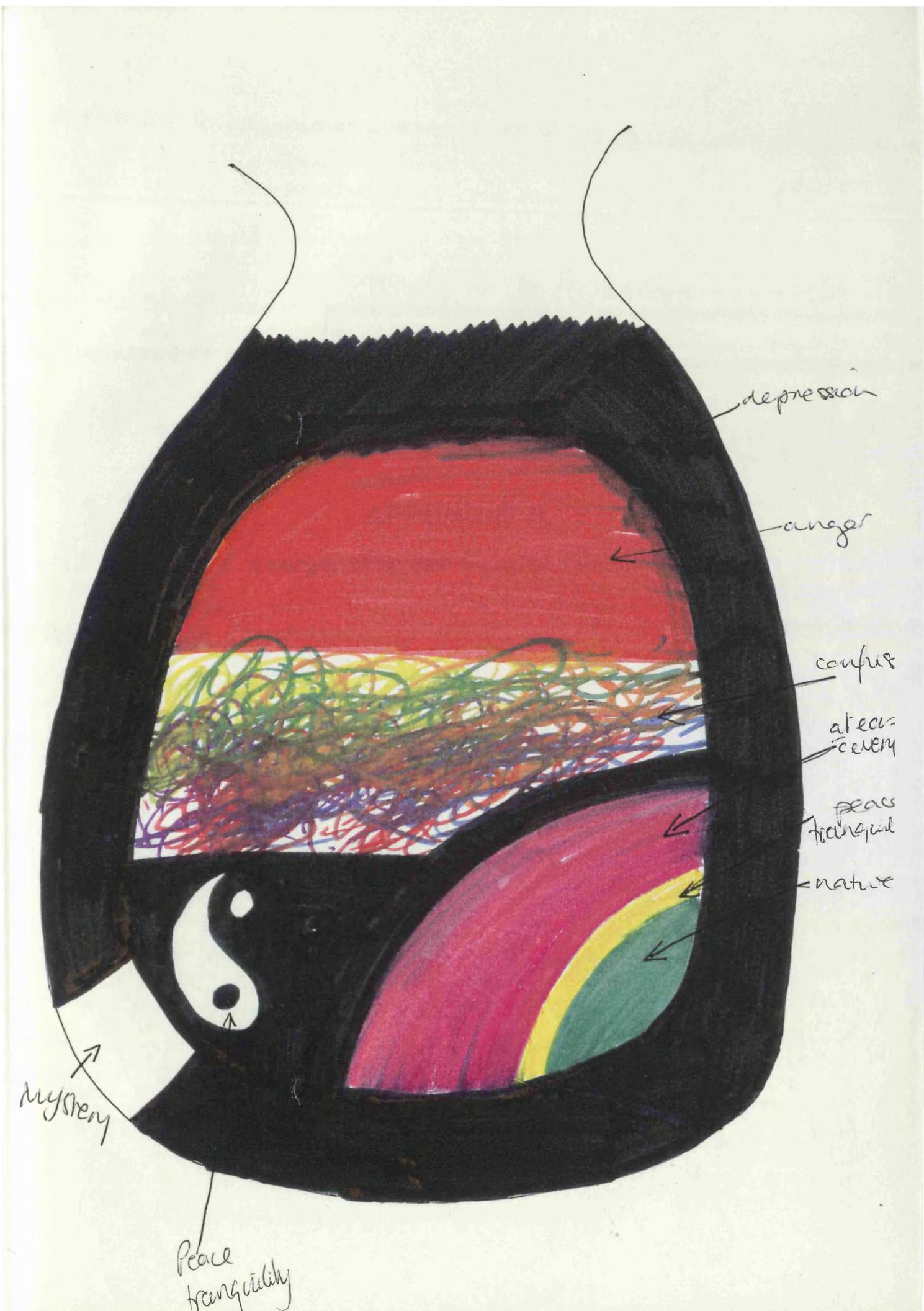
What would you like to see happen?

Do you ever worry about the future? What sorts of things do you worry about for the future?

Is there anything you would like to see change in the clinic/hospital/with your treatment?

## **APPENDIX v**

### **Example of completed bag of feelings**



depression

anger

confusion

anxiety

peace  
tranquility

nature

mystery

Peace  
tranquility

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## **APPENDIX vi**

### **Example of section of coded transcript**

22. R- the more you do...and its embarrassing as well if you want to get on a bus....you're sat on a bus and you start coughing like you've been smoking for 20 years or something.....and people look at you and you just think 'fuck off'..sorry

reception of others  
negative opps  
judged wrongly → anger/resentment

I- no, no

23. R-...because it just really gets on your nerves...so that as well and people who....you know think its really funny to say 'oh that's a bit of a smokers cough you've got there'..and you think oh yeah that's really funny.....its just ignorance, they don't know that its not but you just think 'piss off'.....its more social really..the way I feel...its not like the way I feel when I'm with my friends.....or like when I have to run for a bus..I don't I just wave and hope that he'll stop.....I wouldn't because I couldn't.....and you feel like a bit of an invalid sometimes.....urm so its just really the breathlessness but it wouldn't be a problem if I wasn't so concerned with that...the way I look and stuff like that...you know my image.....because you know like X said last you when I was really ill about I might have to go in a wheel chair and I said 'I'm not going in a wheel chair'...you know...I'm not going round in a wheel chair.....so it probably wouldn't be too bad if I wasn't too vain (laughs)

insensitivity / thoughtlessness  
hidden resent  
justification for insensitivity  
image maintenance  
worry about image  
judgement of self as harsh as possible  
judgement of others

Fear of further damage to image → motivation

I- show me a person who's not vain

R- Exactly

I- Do you think there's anything you'd miss out of or that you might lose if you didn't have CF?

24. R- ...urm...I don't know.....I've never thought of it like that.....I suppose in a funny way I'd sort of miss coming into hospital....because but only because of the people because you know Y and Z and the staff on the ward you know they're practically friends now.....urm I'd miss that I think...a little bit (laughs)

importance of relationships with medical team

I- Not enough to make it worthwhile

25. R- yeah.....urm.....I don't know I think it'd just be very strange not to have it.....not having the drugs would be really weird and not having the physio.....not having loads of tablets before I had like a chocolate bar or something.....I'd just think oh god...diarrhoea.....urm.....it'd be strange.....it'd be nice.....urm.....

integral part of life  
uncertain before positive decision making

I- Are there other ways your life would be different if you didn't have CF?

26. R- I think the way people would treat me as well like mum especially.....she's very very protective at the minute....well she always has been and I don't think she'd be half as protective if I didn't have CF.....

increased protectiveness by others  
allusion to how being a difficult time brushed over, minimized

I- Is she protective with your sister?

27. R- I think she's just protective anyway but she wasn't this bad with my sister....or good whatever way you look at it....urm.....

positive & negative side to differences in relationships

I- What about your dad would he treat you differently?

28. R- No and yes.....yes because he wouldn't....he wouldn't be 'oh are you alright ' sort of thing all the time.....but.....no because I think he tries to forget that I've got it anyway.....I don't know...I think he finds it pretty hard to deal with anyway.....urm I think really its just his defense dawned on him that I'm as bad as I am....I don't think he thought.....because he wasn't really very involved when I was little because he was always quite busy so I think its shocked him that I've got this bad.....so that's why he sort of....it seems very false to me that he's like he's really concerned all the time but I don't think it is false its just because I'm not used to it because he's always put on this pretence that I'm O.K.....I think anyway I don't know

dad's seek reassurance helpful  
denial by other  
his defense  
avoidance or denial  
he pretends o.k easier to do for self  
concern in his awareness of avoidant strategy or denial

I- Its something that's changed a bit and you're not quite sure why?

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