

IDENTIFYING THE IMPACT OF CYSTIC FIBROSIS ON THE LIVES OF PARENTS

By

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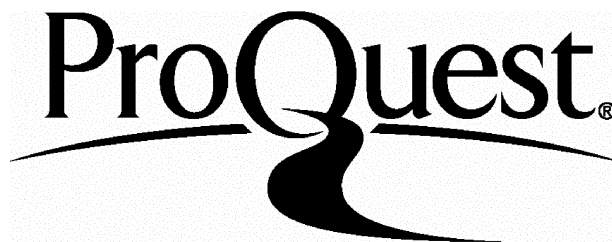
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She's already been up 3 hours getting him ready for school



The boy suffers from Cystic Fibrosis. Thick mucus collects in his lungs and has to be dislodged morning and night by vigorous physiotherapy. This is a daily routine and is essential to help prevent infection and lung damage. With your help we can fund workers in the community to help him and his family through each day.

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 **SMALL CHANGE MAKES A BIG DIFFERENCE**

‘Under present treatment methods (for cystic fibrosis), the child’s life depends on nothing less than the family’s ability to put him first as long as he lives’
(Travis, 1976)

Poster reproduced with permission from the Cystic Fibrosis Trust

Abstract

Background: Cystic Fibrosis is a multi-system disease with no cure. Treatment is largely home based and consists of a rigorous daily treatment regimen of chest physiotherapy, medication and diet. Clinical status should not be the only consideration for health professionals. The continuing ability of the parents to care for the child should be part of routine assessment. **Aim:** To identify the emotional and practical stressors parents associate through living with cystic fibrosis in the family. **Methods:** 124 parents of 284 children participated in this national, case controlled study. Validated measurements of psychopathology and questionnaires designed to identify the intrusion of disease were used, followed by interviews using a grounded theory approach.

Results: Parents of children with cystic fibrosis scored significantly higher in all measures: General Health Questionnaire: $p < 0.005$, Beck's Hopelessness Score: $p < 0.001$ and Impact on Family: $p < 0.001$ indicating a more negative response. Interviews identified key sources of stress: financial and employment, perception of disease status, knowledge and understanding of the disease and family relationships and support. **Conclusions:** Parents of children with cystic fibrosis identify greater mental and practical stress in caring for their child than parents of healthy children. Poor mental health can be explained by their perception of the practical burden of caring for a child with cystic fibrosis. Professionals must take into account the burden on parents when prescribing treatment and must be alert to indicators of stress rather than simply assessing psychopathology assumed to occur as a direct result of illness.

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CHAPTER 1

THE IMPACT OF CHRONIC DISEASE ON THE CARERS

1.1 Introduction

This study explores the impact chronic disease has on the lives of a small group of parents within the relatively large population of unrecognised individuals who informally care for their relatives. Governments universally accept, and in some countries expect, relatives to support the national healthcare systems by providing round the clock, free care to an enormous caseload of the sick, elderly and those with mental illness. There is very little practical or financial support offered to this group who take on a burden of care often to the detriment of their own and their families lives. Recognition of this undertaking has been studied in adult carers who look after their adult children or relatives. Society however expects parents to look after their children; unfortunately there is little recognition of the distinction made between parents as parents and parents as carers.

The purpose of this study is to identify some of the burden that a particular group of parents who have children with cystic fibrosis experience and whether variables such as gender of the carer, intrusion of disease, perception of disease status and employment status play a role in this experience. Cystic fibrosis is an

inherited, life long disease and as such could be used as a model for many other chronic diseases.

There can be no doubt that clinicians today, more than ever before, are starting to recognise and acknowledge the psychosocial impact that caring for a friend or relative with a chronic disease has on daily life. Time consuming and often technically demanding treatment regimens together with emotional concern and stress may overwhelm some carers. This constant pressure may result in sub-optimal and occasionally dangerous levels of care with poorer emotional and physical health for the carer. It may be that by trying to provide an ever-increasing level of treatment carers will fail to keep up with medical requests, resulting in a detrimental rather than improved impact on the patient's health. Patterson, McCubbin, and Warwick (1990) concluded however that when the family functions well the child's health will benefit, this theory has lent support to the need for an increasing focus on the ability of the family to manage treatment demands.

1.2 The Caregiver Burden

What is caregiver burden? The literature has provided us with a multidimensional view suggesting many aspects of burden experienced by caregivers. These definitions of burden have been described by informal caregivers looking after children, the mentally ill and the elderly and include

physical, psychological, emotional and financial problems (Dowdell 1995, Stuckey, Neundorfer, and Smyth 1996).

Research has indicated that stressors such as adaptation, anxiety or acceptance of the diagnosis may not individually have an impact on the daily life of the carer (Kazak, Reber and Snitzer 1988). However, taking all stressors together and examining the complexity of caring may allow the concept of burden to be applicable in the context of chronic disease. This concept must be acknowledged by clinicians to ensure a constant quality of care for their patients; health professionals prescribing any form of treatment must acknowledge the level of treatment complexity they are demanding. Professionals must also be aware of their responsibility for assessing the ability, psychological well being and personal health of the main carer and consider the potential burden they are imposing. Currently the patient's health status and treatment regimen are routinely followed up in outpatients. However the treatment related burden associated with caring for a friend or relative, as well as an acknowledgment of the psychological implications of caring for someone with a life threatening disease, must also be part of a clinicians interview.

Informal carers represent a major but hidden burden on the healthcare system, however the bulk of literature examining the impact of caring for a sick relative appears to have been focused mainly in the field of adult mental health and the care of the elderly. Such research has mostly been concerned with the coping mechanisms of relatives caring for someone with a psychiatric illness,

from schizophrenia to Alzheimer's disease, showing how caring can adversely affect the relationship between the carer and the patient often depending on the individual carers characteristics.

In 1996 Caserta, Lund, and Wright in a large study of 160 cases examined burden both quantitatively and qualitatively and extended the problems described above to five dimensions of burden - time demands, emotional, developmental, physical and social. Platt (1985) has provided us with a definition of burden as a disruption to family and household life that is potentially verifiable and observable. Hoenig and Hamilton (1966), further described burden as anything that occurs as a disruption factor in family life owing to the patient's illness, following a study of sixty-two carers of schizophrenia over a six year time period. Newman (1997) concludes, following a review of the literature, that it is clear that caregivers are subject to demands beyond their daily responsibilities and it is these demands that can be described as a burden that disrupts all aspects of daily life, emotionally, in employment and financial matters, social life and leisure activities and in the general interactions we all get involved in, within and beyond the family.

These definitions indicate that perhaps burden of care is a relative concept. Each individual's perception of burden is made in relation to their definition of normal lifestyle at any particular stage of their life. However, the caring role, whatever stage of life it is adopted can only be seen as an intrusion into an individual's lifestyle. Cuellar and Butts (1999), following a review of the

literature acknowledge that care giving may be a role assumed with little or no choice and may not be taken on willingly. Donaldson and Burns (1999), further describe the intrusion of a caring role. In a study that sought to identify the origins and management of caregiver burden the authors state that the caregiver's own characteristics play a major role in determining how burdensome and stressful they find this imposed role.

Burden described by caregivers therefore appears to be influenced not only by the degree of care required, but also by the characteristics of the individual involved and their relationship with the patient. For many however, providing informal care can dominate all aspects of an individual's life - whatever the level of care being identified. In recognition of this intrusion, care giving has been described as a solitary journey by Boland and Sims (1996). These authors, in a study using a Grounded Theory design, interviewed seventeen families caring for people between fourteen months and eighty-seven years with chronic, medical illnesses. Although this study used small numbers of cases with a wide variety of experience some common themes were identified. The carers interviewed reported overwhelming feelings of responsibility, isolation and commitment with little or no perceived support from friends, family, social or health services. It is the contribution of emotions such as these that play a major role in our understanding of the degree of burden experienced by caregivers.

Burns (2000) in a review of the burden that informal carers face, described feelings of strain, obligation and social isolation. Burns further commented that

the individual characteristics of the carer dictated carer burden. These characteristics included personality, gender, degree of formal and informal support, physical and mental health as well as attributional style (coping style) and expressed emotion (critical or hostile attitudes). The review concluded that as informal caregivers play such a crucial role in maintaining patients in the community, it is essential that appropriate management strategies incorporating interventions that address the specific burdens of the individual carers are essential.

Research investigating the effects of burden on the caregiver has been mainly centred in the field of adult medicine, with a particular focus on the elderly and those with mental illness using a methodology largely based on self-report questionnaire measurements. Much of this research has examined the effects of care-giving on individuals including their quality of life, the coping mechanisms developed and used, and which particular family members are carrying out the caring role (Weitzner, Jacobsen, Wagner, Friedland and Cox 1999). The literature suggests that there is an obvious relationship between the effects of objective burden on the caregiver and their subsequent quality of life. This relationship has been demonstrated in a large study of 1594 veterans investigating the relationship between caregiver burden (caring for a variety of mental and physical disabilities) and health related quality of life. Hughes, Giobbie-Hurder, Weaver, Kubal and Henderson (1999) reported that emotional distress, deteriorating marital and family relationships, and problems with finances played a significant role in the caregivers lives.

Further investigations into burden have used study designs that include telephone interviews schedules or face-to-face interview schedules over periods a few months to many years. These studies have been designed to examine methods of coping, adapting to burden and the complex association between objective and subjective burden. Researchers have concluded that the concepts of objective and subjective burden can be differentiated. Objective burden has been described as those tasks required to care for the patient, whereas subjective burden indicates how the carer feels about undertaking those tasks (Jones 1996, Budd, Oles and Hughes 1998, Faison, Faria and Frank 1999).

What further factors in the lives of carers can help us to understand objective and subjective burden and the impact of becoming an informal carer? Webb, Pfeffer, Muesar, Gladis, Mensch, DeGirolamo et al (1998) in a study examining the relationship between factors mediating the impact of caring and subjective burden reported that, although the carer may accept the many tasks presented, it is the relationship between the patient and carer that influences how successfully those tasks are carried out. There is evidence in this study indicating that it is conflict in the carer/dependent relationship that leads to an increase in reported subjective burden.

The conscious or unconscious development of a coping style also appears to be a factor that influences subjective burden. In a study looking at the burden and well being of caregivers for the severely mentally ill (Webb et al 1998), positive and negative symptom behaviour had a direct effect on the carer's ability

to use a problem-solving approach to coping. To illustrate this theory further Budd, Oles and Hughes 1998, in a study investigating coping developed a questionnaire designed to measure coping styles. The questionnaire was used with ninety-one carers of patients with schizophrenia. Results indicated that where carers employed collusion, criticism/coercion, overprotectedness, emotional over involvement and resignation as coping styles, higher levels of burden were also reported.

1.3 Gender

Special attention should be made to a more specific factor influencing the perception of burden an individual recognises. Although it is often not considered, gender is an important issue and has frequently been described as playing a role in the way caregivers deal with burden. The differences between male and female carers are significant features that influence coping and should be acknowledged therefore by health professionals when planning care in the home. The male as a caregiver has been described as a non-traditional role although it is acknowledged that this is changing as fathers, partners and husbands increasingly contribute to family healthcare in the community.

Men are reporting to researchers that care giving is a problem. Mays and Lund (1999) informally interviewed ten men caring for the severely mentally ill, addressing psychosocial, physical, financial and crisis management issues.

Results were reported in three themes; burden, duration and depth of commitment, and role affirmation. The investigators concluded that men as carers were identifying two main areas of concern - needing more support (than women) and recognition from professionals that they were carrying out a non-traditional role. Similarly, Gilbar (1999), in a study interviewing forty-five elderly carers of a spouse with cancer reported that husbands identified greater burden in their caring role than wives. Conversely however, although the male carer sometimes appears to request more support than female carers, it may not always be the case. Faison et al (1999) reported that in using interview schedules with eighty-eight caregivers of the elderly and chronically ill, sons as care givers reported less burden than daughters, wives or husbands.

It appears therefore that society's expectation continues to support the belief that women should traditionally carry out the care-giving role and have even been described as better suited to caring by both sexes – as patients and carers, often feeling a greater obligation to care. Collins and Jones (1997) used four questionnaires with forty-eight spouses to explore why women experienced more strain and distress in the care giving role than men and further explored what the factors underlying this sex difference may be. Women in the study reported significantly higher levels of strain and their morale was significantly lower than the men. Although women appeared to be carrying out the majority of care giving in the community there were many accounts of barriers to their ability to cope successfully. Women in the study described being frustrated and wanting

to give up their role as caregiver, but felt that attitudinal and societal factors were preventing them causing their higher levels of stress and distress.

It is emerging that women not only accept, but also expect to take on these caring role, however they perceive barriers that inhibit them carrying out this role successfully. Harrison and Neufled (1997) used a qualitative study to explore women's perceptions of the barriers that prevented them from using available support. Twenty carers of babies and twenty carers of the elderly described an obligation to care for relatives, preferring to be offered support rather than having to ask. The women felt too inhibited to ask; they felt that professionals would assume that as carers they would not need support. Women also identified various problems including feeling a loss of independence, lowered self esteem, a concern for others, a fear of what others may think if they refuse and, in practical terms, the time involved in providing the care. Societal norms suggest that family care giving is the responsibility of the women. It appears from studies therefore, that the high levels of strain and distress reported by women who take on the responsibility of family care giving may be attributed to both the expectation of society and of themselves as individuals (Collins et al. 1997).

1.4 Containing the Burden

Acknowledging the presence of burden in the lives of caregivers is only a part of the psychosocial support care givers require. Client involvement in all aspects of planning and implementation can aid management of the treatment regimens and improve coping. Winefield, Barlow and Harvey (1998) used both qualitative and quantitative data in a paper to advise professionals planning to run support groups. The recommendations were based on comments by carers of patients with schizophrenia. Carers described needing better healthcare services, accurate information, and respect from professionals. As with Burns (2000), the authors recommended that with the increasing focus on community based mental health services, family caregivers require and deserve professional support.

In adult medicine the concept of the treatment related burden of care for friends and relatives is considered to be an important concern for the professionals looking after patients. Expectations of responsibility placed on carers are accordingly modified with the contribution of user involvement in the development of interventions for the carers. Carer involvement in setting up appropriate services can be illustrated in a study by McGarry and Arthur (2001). These researchers interviewed ten female and four males aged seventy-five years and older who were informal carers and attended a General Practitioner Practice for regular health checks. Themes from the interviews included the organisational demands of caring and the structure of the caring relationship, the

need for informal support networks and formal services. The researchers concluded that the constancy of the carer and the need to be recognised could be met with involvement early on, from the time of their own health check to the development of strategies designed to assist the carer planned between the carer and the community nurse.

Dening and Lawton (1998) likewise supported the idea that carers should be involved in the development of services. A Medline search and a review of recent experiences in service planning, implementation and evaluation in the local service was carried out. The researchers were able to demonstrate that carers' perceptions and experience could contribute to the development and evaluation of specialist mental health services for the elderly. They concluded that there was considerable scope in involving carers in the development and evaluation of services, however warned that there could be problems as carers and users may have differing perspectives, and that there should be standardised assessments of carer satisfaction in place.

In order to allow health professionals to appropriately tailor the degree of treatment responsibility assigned to caregivers looking after patients with a chronic disease and additionally provide them with adequate support, 'burden' must be identified and measured in some way. Burden is however, a subjective concept that is interpreted differently by any individual at various stages of their life and the course of the illness. In order to be useful to the clinician, its definition and measurement therefore poses a huge problem.

1.5 Summary

It is clear that the concept of caregiver burden and the effect it has on an individual's emotional, physical, social and financial daily life has been recognised by professionals working in adult medicine. Recommendations for considering the needs of the individual providing healthcare at home for a chronically ill person are made throughout the literature. Recognising that parents may also be affected in a similar way is complicated by society's assumption that parents, by default, are automatically good carers and that mothers traditionally should carry out this role. This assumption however, denies their rights as parents and changes their role from parent to carer. The imposition of treatment and care may potentially alter the child - parent dynamic. The changed roles may affect the individual's relationships throughout childhood and have implications for the child's transition to adulthood.

Unfortunately there are potential problems with many of these study designs and care should be taken when interpreting the conclusions. Many of the studies described above have used face-to-face or telephone interview schedules, or questionnaires carried out on a local population of cases. Interviews alone may be limited to qualitative data collection therefore preventing measurable data being collected or, in the studies employing self-report questionnaires only, does not allow the individuals to confirm or elaborate on any of the issues raised. Although the cases are from a wide source of carers – those caring for children, the mentally ill and the elderly, the use of local populations

does not take in to account the effects of the regional variation of care or the support services available for the carers. Many of the cases were recruited from the general practitioner's surgery or outpatient clinic. This method of recruitment can bias the case population as it enrolls only those who attend the service, missing a population that do not attend a formal service and who will perhaps have differing needs and responses.

Despite the limitations, the various descriptions of burden discussed above have however provided recurring themes from a wide case source that can be summarised into three main categories - physical, emotional and social intrusion. These categories can be further sub-divided to encompass the aspects of a parent's life that the burden for caring for a child with a chronic disease can influence - marital relationships, family relationships, employment and financial problems, perception of disease status and gender of the carer. The following chapter will further discuss in more detail the impact on the lives of parents with a chronically ill child.

CHAPTER 2

THE IMPACT OF A CHRONICALLY ILL CHILD ON PARENTS

The expectation for a couple starting a family is to provide a caring and safe environment for their child, with reciprocal enjoyment and pleasure between themselves and their child. As parents, expectations in raising a growing and developing child is for a fairly trouble free time, while acknowledging that childhood illness and accidents do occur. The diagnosis of a life long, chronic disease is devastating for parents as their plans and dreams are immediately destroyed and they have to start learning how to become carers of a sick child, commonly being taught complicated medical techniques and concepts at this very difficult and emotional time (Eiser 1994).

In trying to identify the effects of the care-giving burden on the parents of a sick child we must take into consideration family lifestyle and the characteristics of the individuals. Different types of lifestyle may affect the degree of burden, particularly if all family members are involved in it. Perhaps families would be living a particular type of life, regardless of chronic illness therefore, how far could the effects of chronic illness on any given current lifestyle be blamed. Equally, when thinking about burden one could also consider the many aspects of care giving that in fact give pleasure and satisfaction. Could a sense of reward

from caring for someone with a chronic illness in some degree be thought to balance out the effect of the burden (Stuckey et al 1996)?

2.1 Parental Role

Although there is wide recognition of the needs of parents caring for adult children and partners caring for each other, there has been little or no recognition of the needs of parents caring for their chronically sick children. In September 1995, the British government recognised this need and indicated an interest in supporting parents as carers. The Department of Health, as part of the Policy Research Programme on health and personal social services invited research proposals on the availability and mobilisation of formal and informal support for parents. These issues were divided up into three specific areas of concern: firstly, the identification of families under stress, secondly, the difficulties parents face and the situations parents have to cope with and thirdly, specific parenting problems and the co-ordination and management of services with links to supportive networks.

This support from the Government has increasingly raised awareness in the UK and has highlighted areas where there is little knowledge. Although much research has been carried out examining the role of the parent as caregiver, it has predominantly been in North America. Researchers to date have commonly concentrated on the needs of the sick child. Coping, personal health and support

for parents with children who have a variety of chronic diseases have been examined in the context of the effect that parental coping has on the health of their children. It appears that despite the wealth of literature exploring the effects of the caregiver burden on carers of the mentally ill adult population and the elderly, parental responsibility as a caregiver is assumed. The impact of a parent becoming a carer rather than just a parent therefore has received little recognition.

Many of the studies that have been carried out exploring the care giving role have encountered problems. The methodological concerns with many of these studies have been recognised by Quittner, DiGirolamo, Michael and Eigen (1992). These researchers, in a study measuring specific stressors encountered by sixty-four parents of newly diagnosed children using structured interviews and standardised measures, noted that the main problem in many of the similar studies that they reviewed was the use of a non-categorical (different diseases) approach used in recruitment of cases. Most studies appear to combine diverse medical conditions with a limited case population; therefore the conclusions reached by such research identify a lack of situation specific causes of stress and coping. These comments highlight the need to accurately assess the role related changes and the need to measure the ongoing strain specific to the medical condition.

Initial research into the impact of childhood chronic disease on the parents assumed psychopathology and so studies were designed to identify and

measure this. It became apparent however that not all parents displayed these problems (Kazak et al. 1988). In this matched, controlled study of forty-five cases and forty-nine controls, five self-report questionnaires were used to identify parental psychological distress, marital satisfaction, parenting stress, family cohesion and adaptability, and child behaviour. Multivariate analyses failed to show significant group differences, although univariate analyses did indicate lower levels of adaptability and cohesion in the case families. These results indicate that perhaps the focus of research should change and concentrate more on the identification of problems if they exist and, if they do, the causes underpinning them.

The following sections review the literature to identify variables such as gender, employment status, perception of illness, and available support that may account for the impact that caring for a child with chronic illness on parents. These themes are the corner stones of a functioning society and appear to play an important role in the way that carers develop individual coping strategies and manage the demands placed upon them on a daily basis, both psychologically and practically.

2.2 Gender Difference

Although the idea of caregiver burden is a relatively new concept, the responsibility and ultimate consequences in caring for a family member has been

around for centuries. In a review article Hoffman and Mitchell (1998) report that despite increasing participation from husbands and sons much of the healthcare provision at home is based on the implicit assumption that women are available to provide homecare.

In a study investigating a sample of primetime network television advertisements in North America the researchers were able to demonstrate society's expectation of women as carers. The findings indicated that women were significantly more likely than men to appear as characters in advertisements for medicinal products and they were frequently portrayed as experts on home medical care, often as mothers caring for ill children (Craig 1992). Women in societies worldwide experience the burden of social policies reinforcing this value system. In Hong Kong, Ngan and Wong (1995) in a review article, described a 'caring trap' for female carers, especially unmarried daughters due to increasing longevity in their parents' generation and cultural expectations. The expectation that women will provide care can be further illustrated by Sterritt and Pokorny (1998). In a study to explore the meaning of care giving, researchers used a series of open-ended questions to nine African-American caregivers. The conclusions drawn from this study indicated that although care giving is a traditional family value and an act of love it continues to remain a female role.

Many authors have recognised and commented on the multifunctional role of the female, describing women as mothers, wives, sisters, daughters,

employees and housekeepers - often all at the same time. Stephens and Townsend (1997) in a study of 296 female primary care givers found a complex relationship often existing between a woman's multiple role experience and her psychological well-being, with carers fulfilling at least three other roles – mother, wife and employee. These extra roles increased the stress experienced by the women except for the role of employee, where it was found that going out to work actually became a stress buffer. As professionals, are we conscious of our presumption that the mother will give up work to carry out the majority of the treatment for the child? Should we perhaps ask ourselves whether our assumption that caring for a sick child is merely an extension of maternal duties and therefore not an issue, or is treatment related caring a physical and emotional burden no matter what the familial relationship between carer and cared for?

Higher levels of psychological distress have been identified in mothers than fathers, a result also found by Mastroyannopoulou, Stallard, Lewis and Lenton (1997), who, in a study of ninety-three women and seventy-eight men looked at the impact of childhood life threatening illness on parents. The researchers found significant differences in coping between mothers and fathers characterised by increased levels of psychological distress, significant effects on employment and relationships and a family environment characterised by low expressiveness, low cohesion and increased conflict. There is no doubt therefore that women are taking on the burden of care giving, willingly or unwillingly. When recommending new therapies and regimens, professionals should perhaps aim

to discover who the main carer would be and what other roles they would simultaneously have to undertake. Home care could then be planned through negotiation between carers and professionals with some give and take on both sides.

2.3 The Influence of Character

Studies discussed previously (Donaldson et al. 1999) have identified that the individual carer's character plays an important role in the level of identified burden. In two studies (Canning, Harris and Kelleher 1996, Luescher, Dede, Gitten, Fennell and Maria 1999) care givers of children with chronic illness completed self-report questionnaires to identify their perceptions of burden. Both these studies used local populations with 116 and forty-nine cases respectively. The carer's character and outlook appeared to influence their perception of the child's health status. This in turn emerged as important as parental attitude seemed to have a more direct relationship to burden than clinical measures of health status. Both of these character traits are interrelated as character and personality will indisputably influence perception of disease status.

These conclusions are further supported by Markiewicz, Reis and Gold (1997) in a study exploring attachment styles and personality traits in 126 caregiver-dependent pairs. Results indicated that the less anxious-ambivalent

care givers reported larger social support networks and more satisfaction with the support received from professionals than those scoring lower on this scale.

Again, further research concurs with other work examining character. Following a study to identify caregiver characteristics influencing childhood nutritional status in healthy children, carers of ninety-eight children in rural Chad were interviewed. Information on demographics such as income and number of pregnancies to social support and overall feelings of satisfaction with life were collected (Begin, Frongillo and Delisle 1999). The study concluded that caregiver characteristics influenced children' nutritional status, even when controlling for the socio-economic status of the household. It is perhaps not surprising that the personal characteristics of parents influence the way they bring up their child even when the child does not have a chronic disease. It is therefore important for professionals to be aware of individual character differences when discussing home care with parents of newly diagnosed children.

Character extremes also appear to influence parenting as Patrick and Hayden (1999) reported. In a study of 596 women who were caring for an adult child with a chronic disease, neuroticism appeared to exert both a direct and indirect negative effect on the well being of mothers. They discovered further that this characteristic had a direct effect on coping and the stresses experienced by the mother. Similarly, Miles, Holditch-Davis, Burchinal and Nelson (1999) collected data on sixty-seven mothers of neonates with a serious life threatening illness for a period up to sixteen months after birth. These data included personal

characteristics, parental role attainment, infant illness characteristics and maternal illness distress using a personal developmental impact rating scale. The researchers concluded that the level of distress experienced was influenced by maternal characteristics and advised that personal characteristics should be a major consideration when planning home care interventions with mothers.

Although it would appear to be common sense that an individual's manner of care giving would be influenced by their personal characteristics, professionals when planning care have paid scant attention to this issue. Research has however indicated that the character of the carer is as important a consideration as the treatment details when dealing with parents.

2.4 Disease Perception

As reported earlier, perception of disease status appears to play a greater role in caregiver reported burden than actual disease status. Luescher et al (1999) reports that the degree of parental burden depends more on the parent's coping skills and the level of family functioning than on the degree of the child's disease severity. In a study of children with Batten's disease, thirty-two primary care givers were compared to eleven primary care givers of children with chronic and less severe medical illnesses. This study examined the emotional state and perceived family functioning of the care givers and it was noted that the more severe the condition was believed to be, the higher the levels of anxiety,

depression and negative effects on daily life (Labbe 1996). Likewise, in a study assessing the long-term behavioural outcome of fifty-one children following brain injury. Kinsella, Ong, Murtagh, Prior and Sawyer (1999) discovered that the acute emotional reaction of the parent to the original injury and parental coping resources were predictive of the outcome.

Researchers examining both the carers and the adolescent/young adult have supported this theory. Abbott, Dodd and Webb (1995) studied sixty adults with cystic fibrosis and Leung, Steinbeck, Morris, Kohn, Towns and Bennett (1997) studied forty-eight adolescents with cystic fibrosis or insulin dependent diabetes mellitus. Both research groups used standardised measures to assess perception of disease severity compared to their physician's assessment of disease status. The results indicated that for adolescents and young adults with a chronic illness, their perception of illness severity was an important indicator of psychosocial well being. This was reflected in adherence to treatment regimens where the severity of illness was not found to influence the rate of adherence. Adolescents and young adults reported different rates of adherence for different aspects of treatment, with neither reflecting the importance of the regimen to their ongoing health. If these young people felt well relative to when they felt unwell, they regarded themselves as not requiring treatment and therefore not sick, thus maintaining an optimistic coping strategy.

This emotional rationalisation for adherence to treatment demands can equally be likened to the parent's understanding and perception of disease

severity following diagnosis. Parents perceiving a negative impact are more likely to feel burden and cope poorly, which may well play a contributory role to an overall worsening health status for the child.

2.5 Knowledge and Understanding of the Medical Condition

Being given the diagnosis of a serious, chronic illness in their child can only be regarded as a major, stressful event in the lives of parents. The diagnosis itself may be difficult to deal with, but parents are often then taught complicated medical treatment procedures that are necessary to maintain optimum health. The impact of disease management on daily life creates both a practical and emotional interruption that may become life long.

The changing emphasis of care for the chronically ill child from hospital to home, along with the expectation of parental involvement and participation will continue to expand. The burden of caring for a sick child at home may increase rather than improve, as developments in medicine are producing increasingly complex treatment interventions and novel treatment options rather than cures for previously untreatable diseases. The delivery of high quality care by parents to their children will only be realised if there are suitable training programmes and liaison between hospital and home to support the parents (Vidler 1999).

Petit de Mange (1998) reports a parent stating that had she known beforehand how difficult, demanding, time consuming and exhausting having to care for her child at home would be, she would never have agreed to take her home. This statement should serve as a reminder to all health professionals that parents may have to assume caregiver roles that professional health care providers have chosen and taken years to develop. Parents often feel under pressure, consciously or unconsciously, from professionals, family, friends and the sick child to take on demanding procedures and techniques in an effort to expedite discharge from hospital, sometimes against their better judgement. Not all parents are natural carers or even want to become carers and yet they are forced to accept the role for the good of their child and because they feel that they would be seen as bad parents if they refused to take it on.

As healthcare professionals our judgement is based – perhaps unfairly – on our own experience, if a parent is not seen to be providing treatment to the standard that we have been taught, then they are assumed to be negligent. This judgement of negligence is often made without considering the knowledge level or understanding of the parent or the demands being placed on them away from the hospital.

Professionals find themselves in a difficult position when a parent refuses to take on treatment demands at the outset or more covertly through non-adherence to the treatment regimens at home. It is often at this stage that legal intervention is occasionally considered, however this is hardly a fair way of

recognising or acknowledging the burden, perceived or otherwise, that parents feel.

As professionals we have to ask ourselves whether this subliminal or overt intimidation, however well meaning, could influence the way a parent copes with the burden of care in the home. Although it would appear obvious that continuous liaison, education and support (both psychological and practical) could help to ameliorate some of the stresses of the care burden, research has provided us with mixed reports. Studies examining the role of education for carers and patients have shown that the provision of information and education programmes have the potential to improve clinical outcome in many specialties, however it is the provision of practical self-management teaching programmes that report most success, although some of these conclusions should be accepted with caution as the provision of education may not always be successful.

Sholten, Brodowicz, Graninger, Gardavsky, Pils, Pesau et al. (1999) in a study of sixty-eight patients with rheumatoid arthritis assessed the outcome of a five year training programme and found that a multidisciplinary approach to patient education led to significant and sustained improvement in clinical outcome. Anthony, Paxton, Bines and Phelan (1999) again confirmed this finding in a study of parents of twenty-five children with cystic fibrosis using self-report questionnaires that measured specific knowledge of nutrition and more general knowledge of the disease, coping strategies, and attitudes and beliefs about

cystic fibrosis. The authors concluded that maternal knowledge improved adherence to diet leading to an expected improvement in growth.

Education and the provision of information may result in an improvement in psychosocial function. Results from interview based studies in multiple sclerosis suggested that relevant, current and specific information helped people with multiple sclerosis retain their independence and empowered them to make informed decisions about their treatment however, professionals are warned that this is a continuing need and should not be based on a 'one off' teaching session (Baker 1998). Siminerio, Charron-Prochownik, Banion and Schreiner (1999), found that using an education package in a paediatric diabetic outpatient setting improved adherence, improved family function and maintained coping in both the parents and children although they did not state how long this improvement lasted.

It does appear from the literature that the timely provision of the most appropriate information and the offer of support by a variety of health professionals helps individuals or care givers to manage. However, as professionals we must not be complacent as the provision of educational programmes does not always work, otherwise the assumption could be made that educating carers would reduce burden. A Cochrane Systematic Review into eleven trials of adult education programmes for asthma concluded that the use of such programmes do not appear to improve health outcomes (Gibson, Coughlan, Wilson, Hensley, Abramson et al 2000). These reviewers pointed out that in

these eleven studies the type of programme and perhaps the timing were problematic. When planning an educational programme it is important for the health professional to consider the type of information being given, by whom and how often, for carers to gain from the service.

2.6 Managing Chronic Disease in Daily Life

The relentless and daily burden of care for parents of children with a chronic disease and the way that they manage it cannot be underestimated. The ability for an individual to cope depends on a multitude of influences and therefore it is not an easy concept to define or measure, although studies are offering more clarity there is still disagreement on an absolute definition. In recognition of the problem of a clear identification more recent studies have acknowledged that there are a range of variables that play a role in coping and many of these variables have been incorporated into methodology. Stewart (1984) reviewing the chronicity of cystic fibrosis and the effect the disease has on parents stated that variables such as previous experience of illness, religion, ethnicity, social class, financial situation and the expectations that parents had for their child, all play a role to a greater or lesser degree. Keren, Feldman and Tyano (1999) in a review, commented that assessment of the caregiver-child relationship has become an intrinsic part of the preschool child psychiatric evaluation. The authors point out that the caregiver-child relationship plays a fundamental role in the ability of the caregiver to cope. The developing

relationship between child and parent is influenced by a myriad of genetic, psychological, cultural and environmental factors in both the caregiver and the child. It is becoming evident that professionals planning a new treatment option must pay attention to the abundance of apparently secondary issues. It is not only the character and ability of the carer that needs to be evaluated but also the relationship between the carer and the child and the caregiver-child interaction.

Labbe, (1996) concluded in a review article discussed earlier, that different parents manage in different ways with illness and suggested that the nature of the specific illness had less influence on the child's adaptation than the quality of the parent-child relationship, the parental acceptance of the illness and the way that parents developed their coping techniques. Kazak et al (1988) in a matched, controlled study of forty-five cases and forty-nine controls, used five self-report questionnaires to identify parental psychological distress, marital satisfaction, parenting stress, family cohesion and adaptability, and child behaviour. They found that parents of children with cystic fibrosis displayed two major coping strategies associated with positive family functioning. These were described firstly as an ability to share the burden with others both within and outside the family, and secondly the way in which parents endowed the illness with meaning, referring to how the parents defined or reframed their situation.

An individual's development of coping strategies aids in their successful management of the treatment requirements. However adapting to these changes can cause problems. Canam (1993) in a paper identifying the common adaptive

tasks facing parents of children with chronic conditions has suggested eight adaptive tasks that parents have to work through as they learn to live with chronic illness (*table 1*). Although some parents, to a degree, successfully manage to accommodate the tasks involved, others experience more problems and develop a multitude of approaches to coping with not only the treatment regimens but also their emotional investment.

Table 1 - Parents Adaptive Tasks (Canam 1993)

Tasks	
1	Accept the child's condition
2	Manage the child's condition on a day to day basis
3	Meet the child's normal development needs
4	Meet the developmental needs of other family members
5	Cope with ongoing stress and family crisis
6	Assist family members to manage their feelings
7	Educate others about the child's condition
8	Establish a support system

Although there is no doubt that these adaptive tasks are important in the development of the parents as carers, they are limited. These tasks highlight aspects of the caregiver burden within the limitations of the home and family. However, as a parent caring for a child with a life long, chronic disease, daily life does not only centre around the home but extends to social networks including friends, neighbours, school and the work place. It is this complete intrusion into all aspects of life that could perhaps be identified as the caregiver's burden.

Watson (1997) in a collaborative project between two paediatric centres used self-report questionnaires to examine the long-term outcome in families with

children commencing renal replacement therapy over a two-year period. A 'burden of care' assessment was used and findings indicate that stress, anxiety and depression scores were higher in parents of children over 10 years of age and that parents who reported less burden had children with an improved health status. Researchers appear to be unanimous in their agreement that coping with chronic disease causes psychological distress, depression, stress and anxiety. However in an interview based study of fifty-four families in Northern Ireland, Burton (1975) found that despite many negative and conflicting emotions, most parents of children with cystic fibrosis managed to transcend their distress and cope with life in a positive way.

2.7 Summary

The majority of the studies discussed in this chapter again, as in the previous chapter, used self-report questionnaire or interview. Although a majority of the questionnaires used were recognised and validated with evidence of reliability, in using them cases were unable to add their own comments therefore contributing to the richness of the data. Many of the interviews were unstructured and open-ended questions can be difficult to analyse objectively, particularly if there are no quantitative data to support the findings. Often sample populations were not randomised and, as in the studies described in the previous chapter, were selected from local populations leading to bias in the population studied.

Although design flaws can be identified, common themes do emerge from the literature. Even in today's changing social structure, mothers continue to assume the role of primary care because of their own and society's expectations (Hoffman et al 1998, Sterritt et al 1998). However, this role appears to cause higher levels of stress for the mother (Mastroyannopoulou et al 1997, Stephens et al 1997). Researchers agree that the character of the carer plays a major role in coping and in the care that they are able to offer (Canning et al 1996, Patrick et al 1999, Miles et al 1999). They conclude that the personal characteristics of the carer should be a major consideration when planning home care. Perception of disease status and previous experience of illness also appears to have a greater influence on caregiver reported burden than actual disease status and researchers found that the degree of parental burden depended more on the parent's coping skills than on the child's disease status (Leuscher et al 1999, Labbe 1996, Kinsella et al 1999).

These studies conclude that different parents appear to manage illness in different ways depending on the nature of the illness, the parental acceptance of the illness and the way they develop their coping techniques. Professionals should not assume that parents can cope or are willing to accept the role of care giver, therefore management of the child's disease should be a partnership between parent and professional, individualised to each carers needs.

The following chapter will review the pathophysiology of cystic fibrosis, the requirements of treatment, the treatment regimens and the problems of adherence to therapies that both children and adults experience

CHAPTER 3

CYSTIC FIBROSIS

Cystic fibrosis is a complex, multi-system disease mainly affecting the respiratory system and the gastrointestinal tract although the liver, joints and reproductive systems may also become involved. The underlying gene mutation found on the long arm of chromosome seven, leads to defective production of the cystic fibrosis transmembrane conductance regulator (CFTR) protein, a cAMP-regulated chloride channel, resulting in abnormal salt and water transport at the apical surface of epithelial cells. Cystic fibrosis mainly affects the lungs where secretions are viscous and difficult to clear and recurrent cycles of bacterial infection and inflammation leads to irreversible lung disease, and digestive system causing malnutrition and malabsorption (Wine 1999).

Currently over 1000 mutations of the gene have been discovered and although there are poor genotype phenotype correlation some mutations indicate a more severe disease progression than others. Unfortunately, this information is not sensitive enough to accurately predict mortality or morbidity. Research is presently exploring the link of moderating genes on the CFTR protein and the effect these may have on disease outcome and future therapies.

3.1 The Requirements of Treatment

There are many aspects of care that must be carried out daily at home to allow the child with cystic fibrosis to live a 'normal' life, and studies have shown that the way in which a parent copes with treatment demands appears to correlate strongly with the health of a child with cystic fibrosis. Reed (1990), in a theoretical study exploring the use of nursing diagnosis to identify families of children with cystic fibrosis with coping difficulties, found that children of families who were not coping well tend to be hospitalised more than those of families who were. The families identified appeared to use hospitalisation as a rest period, again identifying their inability to manage the child's needs at home. In a two-year study comprising ninety-one families attending the same cystic fibrosis centre and using clinical scores (chest x-ray, pulmonary function and nutritional status), telephone interview and self-report questionnaires Patterson, Budd, Goetz and Warwick (1993) correlated disease status and family function. The researchers found that when both parents coped well the children were healthier (seen as an improvement in lung function trend) and recommended that families balance their resources between the child's health needs and the needs of the family as a whole. A similar study carried out by Patterson et al (1990), looked at family stress and parental coping and found that the way in which a family functioned had an indirect effect on critical indices (height, weight and lung function) of the child's health. These findings highlight the importance of considering family variables in order to aim for optimum health status in the child with cystic fibrosis.

The perceived demands of managing treatment regimens for a chronically ill child affect the psychological health of the carer. In a study based on semi-structured interviews with forty-five sets of parents looking at the effect of the burden of caring for cystic fibrosis problems with marital relationships, difficulties in accepting the illness, and inattention to other children in the family were identified (Phillips, Bohannon, Gayton and Friedman 1985). To investigate parental response to cystic fibrosis sixty-four parents were given two hour long, individual, structured interviews and completed five questionnaires. Researchers concluded that mothers reported significantly greater depression, strain in the care-giving role, than fathers, although both parents reported high levels of situation specific and global parenting stress (Quittner et al., 1992). These studies conclude that it is consistently the main caregiver, mother or father, who is reported as having the major problems. One of these problems, poor mental health, has been largely blamed for reduced proficiency of care leading to a poorer health status for the child.

The daily treatment regimens required for maintenance of health comprise aggressive antibiotic therapy coupled with chest physiotherapy for the lung disease, and a high-energy diet with pancreatic enzyme supplementation to combat the intestinal and digestive problems. Despite treatment advances, the disease remains progressive and in over 90% of patients, death is due to respiratory failure, while some patients die from associated complications such as liver disease (Lewis, Morrison, Dodge, Geddes, Coles, Russell et al 1999).

Although there is no cure for cystic fibrosis, conventional treatment for patients has advanced greatly over the last few decades, with consequent improvements in life expectancy to a current median of around 30 years in developed countries (Lewis et al 1999). However, it must be remembered that many patients still die in childhood (Lewis et al 1999). Despite medical advances treatment remains time consuming, complex and life-long and, despite several recent advances such as enteric-coated pancreatic enzyme supplements, little improvement in life expectancy has been seen in the last five years.

3.2 Treatment Regimens

The majority of children are diagnosed within the first few years of life and parents are given a great deal of information about the disease and how to carry out daily treatment regimens at this initial stage. Parents tend to focus almost exclusively on the knowledge that they will see their child die and, especially in the early years, find it difficult not to concentrate on the disease almost to the point of excluding some of their normal daily activities and the rest of the family.

The child with cystic fibrosis is required to attend appointments at the hospital as an outpatient on a two to three monthly basis throughout his or her life so that all aspects of the disease can be monitored and changes in management planned and discussed. Ideally cystic fibrosis care is managed between a specialist centre and a local hospital, when organised well this method

of shared care can contribute significantly to a reduction of the total treatment related burden on parents. Occasionally however, conflicting advice between specialist centre and local hospital does occur causing an increase in anxiety as parents may be left to sort out problems for themselves. Gradually, as the child's condition deteriorates, inpatient admissions for two to three weeks at a time are required, often several times a year. Either one or both parents take the child to outpatient appointments and many remain resident in the hospital during admissions (Dinwiddie 1990). Supported and supervised homecare is an option offered by many Specialist Cystic Fibrosis Centres and may help to reduce the stress associated with repeated hospitalisation. However, taking the hospital into the home may have longer-term implications for the parent-child relationship.

The treatment of cystic fibrosis has been reported by parents and patients alike as monotonous and repetitive. Daily treatment regimens mainly revolve around careful diet planning with administration of pancreatic enzyme supplements, chest physiotherapy, and the administration of both inhaled and oral medication. Patients suffering from exacerbations of their chest condition may also require intravenous antibiotic therapy, with this need increasing as their condition deteriorates. Those severely affected may eventually require continuous nutritional support via a gastrostomy, oxygen therapy, some ventilatory support and become wheelchair bound due to breathlessness and decreasing exercise ability.

Chest physiotherapy is an integral part of the management of cystic fibrosis (Prasad and Main 1998) as it prevents and reduces pulmonary complications by removing broncho-pulmonary secretions. This treatment regimen is introduced at diagnosis and has to be carried out daily for twenty to thirty minutes, increasing up to four or more times a day as the disease progresses. In the recent past much work by professionals and parents has been put into teaching and supporting children with cystic fibrosis to become as independent as possible with their physiotherapy. Many new treatment modalities have been devised and developed to facilitate independence, from breathing exercises such as the Active Cycle of Breathing Techniques and Autogenic Drainage to the use of various pieces of equipment such as the positive expiratory pressure face mask or the high frequency chest wall oscillation vest. Exercise is also encouraged, even when oxygen is required. The psychological benefits of exercise – a feeling of well being and self-confidence with improved body image, and the physical benefits – improved muscle strength and mobility with an increased fitness of the heart and lungs are important in the treatment of cystic fibrosis.

It has been acknowledged more recently that encouraging independence with treatment regimens can be difficult for both the child to accept responsibility and for the parents to 'let go'. Professionals working in cystic fibrosis now advocate a concept of inter-dependence between parents and child with support being given and received from either side. Anecdotal clinical reports indicate that

adherence to rigorous and complex treatment regimens appears to be improved where this philosophy has been adopted.

Recent research has shown that due to a high resting energy expenditure, poor intake and problems with malabsorption from the intestinal tract, people with cystic fibrosis require an intake of 120% - 150% more calories than their peers (Dodge 1998). Children with cystic fibrosis display many problems with maintaining this requirement. Reasons are multiple but may be clinical - abdominal pain due to malabsorption, or psychological - rebellion and peer group pressure. Parents report difficulties with their children's disinterest in food, slowness to eat, hiding food and refusal to swallow leading to mealtimes frequently becoming battlegrounds (Stark 1990). Adults with cystic fibrosis also report that the requirement to maintain an adequate nutritional intake becomes another part of their treatment regimen and not an enjoyable or social occasion.

Treatment for cystic fibrosis therefore is time consuming, intrusive, complicated, differentiating and restrictive. No immediate benefits are felt from most of the treatments as they are largely preventative. These regimens are far too complex for the child to carry out alone and even in adolescence and young adulthood some parental involvement is advocated.

3.3 Adherence

Both children and adults with cystic fibrosis are required to carry out chest physiotherapy, maintain an adequate diet and take a great deal of oral, inhaled and nebulised medication which can often be time consuming and intrusive. Parents find that administering the correct doses at the right times of the day an often overwhelming responsibility which can be made even harder when a child refuses to co-operate. Older children and adults report non-adherence to both treatment regimens and medication as it is often seen as time wasting, boring, and different from the normal life of their peers. These young adults are determined to live as normal a life as possible and acknowledge that they are often making an informed decision not to carry out their treatment. Abbott et al (1995) in a questionnaire based study of sixty adolescents and adults found that patients do not share their physician's perception concerning the severity of their disease and that adherence to treatment regimens were influenced by those perceptions. If patients thought they were well, regardless of their actual degree of illness, they did not feel the need to follow treatment advice. Increasingly the literature is reporting non-adherence to prescribed medical treatment in both children, despite parental supervision, and in adults (Czajkowski, and Koocher 1987). The consequences of poor adherence to treatment regimens can become devastating causing often irreparable damage to pulmonary and nutritional status. It is therefore important to understand factors influencing adherence, including the part burden plays, in promoting good health status.

Studies show varied but significant rates of non-adherence from 20% - 70%, however measurements are not standardised across the literature and rates vary according to the scale used, the content and the rater. Researchers have found adherence rates vary for different aspects of treatment with best rates to medication and poorest for diet and chest physiotherapy (Czajkowski and Koocher 1986). Patients and parents report that adhering to a dietary and physiotherapy regimen is time consuming, boring and interferes with daily activities. Despite understanding the reasons for carrying out treatment, there is an acknowledgement between patients and parents that normal daily life needs to continue.

Not only are parents required to give time to the administration of treatment, arguing with their child about carrying out treatment, dealing with other members of the family or coping with work commitments, but they also face a relentless reminder of the severity of the disease and the ultimate, inevitable outcome. It seems reasonable therefore that parents facing daily anxiety and concern may constantly question the quality and quantity of their treatment as this often becomes equated with the longevity of their child's life. The fact that cystic fibrosis takes up time and emotional energy cannot be denied. What we as healthcare professionals need to understand is, whether caring for a child with cystic fibrosis causes a burden to parents and if so, to what degree?

The following chapter will review the literature describing the impact that having a child with cystic fibrosis has on parents. Further, disease management

strategies and coping styles will also be discussed with the associated needs and family support that parents find helpful in managing their day-to-day life.

CHAPTER 4

THE IMPACT OF CYSTIC FIBROSIS ON PARENTS

Cystic fibrosis has profound influences on every aspect of family life, both practically in the day-to-day management of the disease, and psychosocially as the burden of caring for a child with a chronic, life threatening disease increases. The last few years have brought about a change in the medical management generally for people with chronic diseases. People with cystic fibrosis are now seeing the majority of their treatment regimes being carried out at home, from daily chest physiotherapy to the administration of intravenous antibiotics and ventilatory support. Finkelstein, Budd, Warwick, Kujawa, Wielinski and Ewing (1986), in a randomised, controlled study, used a home measurement monitoring system developed to assess progress and to plan changes in care for patients with cystic fibrosis. Through daily diary recordings of specified measurements 111 patients identified that, although they were unanimous in their acceptance of home management and the perceived benefit that it brought, this was usually because of the reduced inconvenience of hospital visits and interruption of home routines. However, treatment in the home means that the illness is more visible, the differences between the child with cystic fibrosis and the healthy peers or siblings are more apparent and the responsibility of the parent in a doctor or

nurse role increases. These changes beg the question - what effect does home treatments have on normal family functioning?

The role of the parents as caregivers in cystic fibrosis was recognised at an early stage. Cystic fibrosis is a relatively new disease and was first described just over 60 years ago as a separate disease entity by Anderson (1938) and later by Farber (1944). At that time, both authors gave a very gloomy prognosis of only a few years. By 1965, Holsclaw, Eckstein and Nixon estimated an increased survival rate of up to 11 years of age with parents being taught to carry out a basic, daily treatment regimen of chest physiotherapy, home nebuliser therapy and the administration of vitamins, pancreatic enzyme supplements and antibiotics to maintain health.

Unfortunately, despite the increasing involvement of the parents as caregivers interest has so far been directed towards the parents' role in helping their child. There has been relatively little interest in parental experiences or in planning interventions aimed at reducing their distress and promote coping, although their burden may have been identified.

The trend in medical care in the UK, as it is in many first world countries, is towards more home care, and in the future parents will be expected to carry out more technically demanding skills as their children and young adults are discharged into the community. There has however been sparse literature based on UK populations to support or evaluate this trend. In a review article Madge

and Khair (2000) recommended multidisciplinary team management in caring for children with cystic fibrosis due to the complexity of the disease, and in recognition of the multiple needs of the patient and the family. It is through this style of team management that the many psychosocial needs of parents have been recognised. However, acknowledgement of these issues continues to emphasise the needs of the patient, the effect of the disease and the consequent management of treatment.

4.1 Disease Management and Strategies

Parents are now playing a major role in the home management of cystic fibrosis but continue to lack adequate professional support and help to ameliorate some of their problems. Ievers and Drotar (1996) reviewed thirty-one articles describing the functioning of families and/or parents of children with cystic fibrosis in comparison with families of healthy children. The review identifies commonly cited concerns including the difficulty of the treatment regimen, the terminal nature of the disease and the disruption to family life. Parents experienced greater stress and recognised higher levels of burden than the parents of healthy children.

As mothers become more involved in the work force, fathers are becoming more involved with the care and upbringing of their children. Despite this however, it still continues to be predominantly mothers who are identifying major

areas of concern. Olsen (1994) reports the reflections of a mother who has two children with cystic fibrosis. This mother described issues including difficulty in managing treatment regimens, disruption of family relationships, guilt, stress and communication problems. Stewart, Ritchie, McGrath, Thompson and Bruce (1994) interviewed ninety mothers in their homes to examine the specific sources and types of social support they received. Virtually all of the mothers found that demands directly related to the child's care created the most concerns, such as physical care, healthcare in acute illness situations and the constant worry about their child's psychological and social development. Stewart et al (1994) also identified that mothers complained of secondary demands related to their own needs, their role within the family and activities outside the home. It seems apparent, yet unsurprising, that the rigors of caring for cystic fibrosis may appear to overwhelm the lives of parents.

As clinicians we instruct parents on a wide variety of medical procedures to carry out unsupervised at home. Anderson and Elfert (1989), caution us that it is a mother's expectation to cope with looking after her children and that as professionals we rely on that belief. Often the extra burden of caring for a child with cystic fibrosis is not recognised by friends, family or professionals especially as children with cystic fibrosis today do not look any different from their healthy peers. Parents attend outpatient appointments and rather than risk being labelled as an unfit or incapable parent, report that all treatments are being carried out properly and that there are no problems. As professionals we often collude in this

charade for fear of exposing greater problems that cannot be dealt with in the time allotted to a visit.

4.2 Coping Styles

Being unable to admit to any degree of failure or not having the facility to discuss concerns with anyone, leads many parents to report a feeling of isolation, especially in the early years following diagnosis. To explore the problem of loneliness Florian and Krulik (1991) interviewed mothers of children with chronic life-threatening disease, chronic disease and healthy children. The researchers noted that mothers caring for their child with chronic life-threatening disease and chronic disease experienced higher levels of loneliness as compared to the controls, this was despite greater support and an often wider social network. As a result of this work Florian et al have suggested that mothers of children with chronic life-threatening diseases may become subject to a higher existential loneliness.

Research has found however, that when parents acknowledge the challenges that cystic fibrosis presents and adapt to their new situation they develop a sense of control over the cystic fibrosis that leads to a notion of empowerment. In a fieldwork study undertaken to describe the concept of empowerment in mothers of chronically ill children, Gibson (1995) identified four areas of control that have helped parents. These are (i) a discovery of reality, (ii)

critical reflection, (iii) taking charge and (iv) holding on. Gibson concluded that empowerment was largely a personal process in which individuals developed and employed the necessary knowledge, competence and confidence required to enable them to cope with the care of their child. Likewise Cowen, Corey, Keenan, Simmons, Arndt and Levison (1985) confirm this theory in a study of fifty-one parents designed to explore family adaptation and psychosocial adjustment to cystic fibrosis in the pre-school child. By using self-report questionnaires researchers concluded that parents who have confronted the diagnosis of cystic fibrosis go on to minimise the normal stresses of the development period.

Mothers themselves have recognised their own abilities to cope with cystic fibrosis. In a study of fifty mothers and forty-four well siblings of children with cystic fibrosis by Foster, Bryon and Eiser (1998), postal questionnaires were used to identify correlates of maternal well being. In this study mothers did not rate their well being as any different to the normal population and felt that they did not experience any more stress except in times of acute illness. Thompson, Gustafson, Hamlett and Spock (1992) endorse this finding in a study of sixty-eight mothers of children with cystic fibrosis. The study concluded that maternal adjustment was associated with lower levels of perceived daily stress, less use of palliative coping mechanisms and a high level of family support.

It appears that developing effective coping strategies can ameliorate some of the problems associated with having a child with cystic fibrosis. In a paper examining the effect of chronic life threatening illness on families Coyne (1997)

identified three such strategies that parents reported as helping; assigning a meaning to the illness, sharing the burden and incorporating the treatment regimens into a schedule. Ray and Ritchie (1993) identified a similar group of strategies in twenty-nine parents completing two questionnaires, visual analogue scales and home interviews. Parents described many dimensions of care giving including; the use of family support, maintaining a positive outlook and ensuring that care was performed. Thirty-five mothers in a similar study also reported adopting avoidance strategies in an effort to cope, the most common of these being denial of the diagnosis, avoidance of stressful situations and non-adherence to treatment (Mullins, Olson, Reyes, Bernardy, Huszti and Volk 1991). The literature appears to be suggesting that although parents recognise the intrusion of cystic fibrosis into their lives, where positive coping styles are adopted, parents feel that they are managing.

4.3 Family Support

Family life has been found to be a predictor of both coping in the parents and consequent clinical status in the child. Macpherson, Redmond, Leavy and McMullan (1998) collected demographic data, clinical scores and a self-report questionnaire on twenty children with cystic fibrosis from single parents families. Results indicated that the young children of single or teenage mothers had a significantly worse clinical progress and consequently a higher demand for hospital services. In an exploration of the impact of parental relationships on the

home care of children with cystic fibrosis in thirty-seven families, Oppenheimer and Rucker (1980) found that a difference in family composition affected health status. Results indicated that children living at home with two natural parents received more optimal care and were in better physical condition than those with one natural parent. However this study used a population from a local source with no detail of socio-economic status. Many of the studies reported thus far have been carried out on parent pairs with results indicating that mothers in two parent families can and frequently do experience the burden associated with treatment.

Children in society today are increasingly living in non-traditional families (i.e. cohabiting parents, step-families or same sex parents) with a wide variety of cultural, religious and ethnic backgrounds. Single parent families are not unusual and parents no longer accept the tenet that they should stay together 'for the sake of the children'. Anecdotally, health care professionals feel that there are more single parents in their cystic fibrosis populations, although, Sabbeth and Leventhal (1984) in a review of thirty-four studies in the literature on family adjustment to chronic illness showed no significant difference in the reported divorce rate between parents of children with cystic fibrosis and those in the general population. As clinicians however, we must be aware of this vulnerable population of children and be vigilant as to their medical and social support.

4.4 Recognising the Needs of Parents

The age of the child and stage of the disease appear to be the main factors in identifying problems. Eiser, Zoritch, Hiller, Havermans and Billig (1995) while examining the routine stresses in caring for a child with cystic fibrosis found that mothers reported more difficulties in carrying out prescribed treatment regimens when their child was relatively healthy. This is understandable as most of the care is designed to prevent future disease-related problems with no immediate benefits perceived by either the child or the parent. In a study described earlier in parents of newly diagnosed children Quittner et al (1992) identified an increase in both situation specific and global parenting stress with greater strain in managing the care giving role and higher levels of depression in mothers. Walker, Ford and Donald (1987), exploring family stress in cystic fibrosis, used self-rating scales and clinical scores. Conclusions reached by this study indicated that although mothers were able to adapt to the presence of cystic fibrosis, during the stages of pre-school and adolescence they scored higher on a measure of depression than mothers of healthy children. Both these developmental stages are notorious as difficult ages in healthy children however; with the added stress of trying to maintain treatment regimens this finding is perhaps understandable.

Responsibility for treatment regimens and the psychological worry of their child's health create a constant level of stress and anxiety for parents that as clinicians we are not often aware of. Parents report that their basic concern,

which in turn impacts on every other aspect of their lives, is the terminal nature of the disease. Sharkey (1995) describes a chronic uncertainty in parents of children with life-long chronic disease, and Whyte (1992) in four case studies of families where there is a child with chronic disease, reports that the genetic aspects and life-threatening nature of the illness are seen to have a profound effect on the parents' lives. Parents continuously have to adapt their needs as a child with cystic fibrosis has a constantly changing health status. It must therefore be a constant source of strain for parents to adapt to new treatments, prepare and deal with inpatient episodes and learn about the change or deterioration in their child's health.

Recognising the needs of parents therefore, is the first stage in supporting them. To identify parents attitudes to the care provided by a paediatric hospital Baines, Rosenbaum and King (1995) posted a two-part questionnaire to eighty parents of children with diabetes mellitus and forty-five parents of children with cystic fibrosis, all children attended the same paediatric hospital. The results revealed that parents recognised that involvement in the health services - specifically at diagnosis, times of treatment, provision of information, continuity of care and the access and availability of health care professionals decreased their risk of mental health problems. Leonard, Brust and Nelson (1993) confirm this finding by using a standardised instrument to measure psychological distress in fifty-seven families and concluded that the type and amount of professional support taken up by the family affected the parent's level of distress. With the availability of innovative new treatments and an increase in life expectancy,

offering a more supportive role to both patients and their families is becoming increasingly important. It is only when the treatment of cystic fibrosis becomes truly holistic that positive adaptation to life with a chronic, progressive disease can be an expectation.

As with previously discussed studies, the limited area of study in many of the research populations does not allow for differences in healthcare provision and therefore does not recognise the potential variability in the reports of parents' experiences. However, despite the recurring problems with study design and the use of either questionnaires or interviews, using a homogeneous study population has revealed useful core themes in the identification of the impact of cystic fibrosis. Some of the studies discussed have used a random sample of cases and some have been case controlled with parents of healthy children. These type of robust study designs strengthen the conclusions that having a child with cystic fibrosis significantly contributes to the stresses parents encounter in their daily lives.

The following chapter explores past research into the recognition of the cystic fibrosis burden of care. The literature describes key areas that appear to play a role in the management of the disease by families, including acceptance and adaptation, knowledge of the disease and perception of its severity and the support networks available to parents.

CHAPTER 5

THE BURDEN OF CARING FOR CYSTIC FIBROSIS

5.1 Recognising the Burden

There has been little research on the burden of care in cystic fibrosis, specifically as studies have tended to concentrate on psychopathology. However, the concept of burden perhaps better represents what parents feel. A few studies have recognised the burden of cystic fibrosis treatment regimens with the added psychological impact of poor prognosis, however there have been some methodological limitations to these.

Turk (1964), using a questionnaire of eleven questions sought to highlight various aspects of the burden of care identified by twenty-eight parents of children with cystic fibrosis. The results from this study showed that mothers appeared to be carrying out most of the responsibility of care and that this frequently led to exhaustion, tiredness and stress with the consequence of instruction and information often being misunderstood or forgotten. There are however, limitations in this and similar studies, as many study designs used open-ended questions with no information about the reliability and validity of the questionnaires used.

Turk's early recognition that parents found caring for their child with cystic fibrosis tiring and stressful led to a series of other studies concentrating on psycho-social barriers to managing the medical care for a child with cystic fibrosis at home. Studies included interviews with parents (together or separately) or entire families, the use of validated and unvalidated questionnaires and the administration of various measures of psychopathology.

Holroyd and Guthrie (1986) compared sixteen families of children with cystic fibrosis not only with a control group (nominated by each set of parents to minimise demographic differences) but also with families of children who had chronic neuro-muscular disease or chronic renal disease. The design was based on the use of validated self-administered questionnaire. Parents of children with cystic fibrosis exhibited the least stress but did show a significant difference in their ability to cope with their child's management and feelings of hopelessness compared with the control group. As a result of this study the researchers postulated that parents of children with cystic fibrosis experienced less stress than had been previously thought. However a more accurate conclusion may be that parents of children with cystic fibrosis report less stress than parents of children with other diseases. This may be explained by their longer-term knowledge and recognition of cystic fibrosis and the improving prognosis as well as the established use a multi-disciplinary approach to support and care.

Two further studies looked particularly at the coping strategies used by parents of children with cystic fibrosis. Andersson-Segsten and Plos (1986) used

the Chronicity Impact and Coping Instrument Parent Questionnaire to measure the needs, concerns and coping of eighty-five parents of children with cystic fibrosis. More than half the mothers expressed the need to discuss their child's health, diet and emotions. Over 70% were concerned with their child's future. The most common means of coping with these needs and concerns was to get in touch with a nurse or doctor and ask for help. Other reported coping mechanisms were avoidance strategies such as crying or shouting at others. Unfortunately, most of the families who were randomly selected from the Swedish Association for cystic fibrosis were upper and middle class families although the authors estimated that 90% of cystic fibrosis families were members of the association. Only forty-six (54%) responded to the questionnaires, perhaps suggesting that upper and middle class families are more likely to respond to this methodological approach. However, it cannot be denied that the sample is not representative of the cystic fibrosis population.

Again, in an examination of stress and coping, Gibson (1988) analysed parental coping skills. Fifty-six parents completed a questionnaire that was composed of open and closed questions while waiting in the out patient department. The analysis of their responses showed that all the parents were experiencing some cystic fibrosis related stress, the most common problems being related to treatment regimens. Three categories of coping strategies emerged from this study: (i) social support, (ii) problem solving skills and (iii) a belief in the efficacy of medical care.

5.2 Acceptance and Adaptation

As previously discussed, the age of the child and the stage of disease appear to be important factors in reporting stress. Acceptance and recognition of the times of particular vulnerability and that the demands created by illness fluctuate over time help parents adapt to their roles. To investigate levels of stress, McCollum and Gibson (1986) studied fifty-six families of sixty-five children with cystic fibrosis to discover whether these results would alter according to the child's stage of illness. In this study it was suggested that the adaptation process appeared to have four phases: (i) pre-diagnostic, (ii) confrontational, (iii) long-term adaptive and (iv) terminal. In the pre-diagnosis phase parents, especially mothers, felt guilt, despair and even hostility towards their child whom they were unable to nurture successfully. At the confrontational stage, when the diagnosis was confirmed, parents initially felt disbelief followed by an intense grief, anxiety and depression. This study was carried out with parents of children with cystic fibrosis, no control group of parents were used to discern the differences that cystic fibrosis might play in the more global parenting role.

Over time, as they were able to come to terms with the diagnosis, parents fluctuated between denial and optimism and grief and chronic anxiety. During this long-term adaptation phase problems with the management of treatment regimens, admitting repression and forgetting information were also reported. The implications of this study highlight the importance of professional sensitivity to the needs and well being of the carers at particular times of vulnerability.

Again in another study looking at adaptation to illness Cowen, Mok, Corey, MacMillan, Simmons and Levison (1986), studied forty-one parents of children (three - five years old) attending The Hospital for Sick Children, Toronto. The control group were parents of healthy children and were assessed using the Family Assessment Measure. Cowen's results implied that fathers of children with cystic fibrosis, as compared with control fathers, tended to distance themselves from involvement with the diagnosed child. The mothers thus assumed the major emotional and physical burden of caring for the child and used denial coping mechanisms more than the control group.

Although these results may have been confounded as the control group of mothers were mostly working whereas the mothers of children with cystic fibrosis were mostly at home, this study is consistent with the findings of both Turk (1964) and McCollum et al (1986).

5.3 Knowledge and Perception

Gayton, Friedman, Tavormina and Tucker (1977), recognised some of the methodological limitations of previous studies and in particular that most reports had been based on clinical impression rather than the use of objective personality and coping measurements. Their study therefore sought to overcome these problems. Forty-three families were used. Each parent was asked to complete the Family Concept Q Sort and the Minnesota Multiphasic Personality

Inventory. Their responses however, only partially confirmed previous findings. Gayton and colleagues' results showed that parents, unsurprisingly, identified that family satisfaction would be improved if their child did not have cystic fibrosis. Results also identified that fathers were less well adjusted to the situation than mothers, and, in accordance with previous research, 50% of mothers' revealed feelings of depression, hopelessness and futility.

The researchers endeavoured to control their study by using a group of parents of non cystic fibrosis children who had been rated by their teachers as either high or low in social and emotional development. Unfortunately, these control families and the use of different measuring instruments biased data making any statistical comparison difficult to validate. It is however interesting to note that the case group were sent a package of over 600 questions to answer at home and return and it is well recognised that lengthy questionnaires often result in poor compliance and poor quality response (Rust and Golombok 1989).

In a study by Tropauer, Neal Franz and Dilgard (1970) it was confirmed that cystic fibrosis had a considerable emotional effect on the parents. The researchers also noted that an intellectual understanding of the disease did not necessarily eliminate the development of psychological problems. A psychiatrist interviewed twenty-three mothers. No control population was used. Both over-protective and overtly rejecting behaviour was found, manifested by missing clinic appointments, neglecting important aspects of home care and allowing the child to resist treatment. This maladaptive behaviour was present regardless of the

degree of knowledge and understanding of cystic fibrosis. Unfortunately, although this study confirms earlier research, the authors do not describe any validation of the interview questions and the study group were selected purely on their availability.

Frydman (1980) examined the issue of how parents perceived their child's condition. It was hypothesised that those parents who showed a distorted perception would report more symptoms of stress. Eighty-one parents of children with cystic fibrosis were randomly selected from clinical records. Using a 227 item structured interview schedule, psychiatric symptomatology, parental perceptions of disease severity and their opinion of the prognosis were identified. The latter answers were compared with that of the child's paediatrician. The parents were then categorised as 'over-estimates', 'under-estimates' or 'accurate perceivers'. In all cases 'over-estimates' of the severity of their child's symptoms showed the highest level of stress symptomatology. However, no significant differences in psychological problems were found between 'over-estimates', 'under-estimates' or 'accurate perceivers' of prognosis. This study design presumes that the paediatricians' opinions are both objective and correct, but does not however take into account that parental stress may be a factor preventing an accurate perception of the situation.

In 1988, a study by Coren and Martinson looked at whether the uncertainty of having a child with a chronic illness impaired the parents' ability to appraise the health status of that child. Ten parents of children with cancer were

interviewed at annual intervals for a period of five years. Results conclude that once the diagnosis of cancer had been confirmed parents could no longer distinguish any normal variation in their child's behaviour from behaviours that were indicative of illness. The children in the study had different types of cancer, some carrying a poor prognosis, and some usually curable, which compares fairly well with the variability of cystic fibrosis. The authors suggest that the chronic uncertainty of an unknown outcome is responsible for much of the stress experienced by the parents which in turn determines their coping strategies.

In 1986 Nolan, Desmond, Herlich and Hardy studied the issue of parental knowledge of cystic fibrosis. The researchers hypothesised that doctors resist giving information to parents about the consequences of the condition. Consequently, there appears to be confusion amongst the staff in the multidisciplinary team as to whose role this is. Assuming that providing information gave parents a positive feeling of control, twenty-five parents were questioned about cystic fibrosis. Knowledge of the disease pathophysiology was found to be good but knowledge of the genetics involved in cystic fibrosis was only fair. The areas where knowledge was poor related to reproductive risks and male sterility. This could be explained by the age appropriate timing of information giving, reproductive issues are often not explained until the child can understand the implications, usually about eleven to twelve years. Parents who performed best were found to be those older mothers of a higher social class whose children were most ill.

This survey was performed in Canada, and although the relatively small sample contained no significant differences in age, sex or socio-economic status, two thirds of the cases were French speaking therefore the questionnaire had to be translated, potentially causing differences in understanding and answers.

5.4 Support

In 1989, Kulczycki, Robinson and Berg used interview schedules with twenty families and again found that managing treatment regimens successfully were often hampered by feelings of tiredness, hopelessness and difficulties in communicating stresses and fears with partner and child. These results confirmed the imposition that medical management at home places on the lives of parents; however, this was the first study to identify a strong link between overall adjustment and ability to cope with the level of support offered by the child's physician. For the first time, it was recognised that the ongoing health of the child with cystic fibrosis was a joint responsibility between parents and physicians.

Phillips et al (1985) also identified the support gained from professionals caring for families. This study followed up forty-three original cases from a previously mentioned study (Gayton et al 1977) by interviewing them to determine not only their specific problems, but also those areas in which they managed successfully and their means of information and support. Each parent

was interviewed separately for approximately two hours by two experienced interviewers using a semi-structured approach developed specifically for this study. Results indicated that uncertainty of outcome and communication between the parents was the most prevalent major concern. Approximately half (51%) of the parents reported a minor problem in following their child's treatment regimen but 78% denied any difficulty with accepting their child's illness. Most of the parents (72%) used the doctor to gain information about the condition and 43% found this the most valuable source of help.

Using social support, most commonly by a multi-disciplinary team, is an action oriented coping behaviour that enables parents to feel a sense of control. Problem solving skills and having a system of beliefs are described as intrapsychic methods of coping that gives parents a sense of personal control. In Gibson's (1988) study previously discussed, it was concluded that providing information about cystic fibrosis and allowing parents to talk about their feelings would facilitate a sense of control.

The majority of the studies described contain some flaw in study design or case recruitment. Turk (1964) as do many of the other studies, does not report any information about the validity or reliability of the questionnaires and it must be borne in mind that response to open-ended questions are commonly difficult to analyse objectively. McCollum et al (1986) primarily used multiple-choice questionnaires and although this design allows for a range of material to be

covered, it can be difficult to include a wide range of items effectively and gives the user an opportunity to guess the 'correct' answer (Rust et al 1989).

Many of the samples were not randomised and interviews were often semi-structured, a study design that does not always facilitate objective analysis. Phillips et al (1985) carried out a study based upon self-report measures but this study design is often liable to bias as it is found that the interviewee may respond in a socially desirable way. The interviewers also categorised responses into major or minor problems and their criteria were not tested for validity or reliability. 'No problem' was defined as no problems or difficulties having been stated which does not necessarily equate with no problem existing.

Despite the many research design flaws, these studies identified similar areas of concern for the health professionals, from recognition that the mother adopts the main caring role, to an acknowledgement that the clinician plays an important part in support and information giving.

Some of these studies were carried out in the early 1960s when the traditional and societal roles of mothers and fathers were very different from today. Fathers went out to work and mothers became the main child carer by default. It is perhaps no surprise therefore that in the majority of these studies mothers assumed the major caring role. As a consequence of this physical distancing studies (Cowen et al 1986, Gayton et al 1977 and Andersson-Segsten et al 1986) found that fathers distanced themselves emotionally, were less well

adjusted and were not able nor wanted to discuss their child's disease and the treatment. The burden of caring for the child with cystic fibrosis therefore fell on mothers who appeared to suffer both psychologically and physically as a result.

In all of the studies, mothers reported a variety of distressing emotions from guilt and denial to futility, depression and hopelessness. McCollum et al (1986) recognised that adaptation to their child's diagnosis fell into four phases and that in each phase different emotions predominated which either aided or hindered coping. Communication between partners was also reported to aid coping, although studies (Kulczycki et al 1989, Cowen et al 1986, Phillips et al 1985 and Andersson-Segsten et al 1986) found that parents reported difficulty in talking about their child and the disease, not only to each other but also to friends and health professionals.

Both mothers and fathers indicated that the heavy burden of treatment regimens caused them psychological stress and physical tiredness. Parents recognised that in turn this led to non-adherence, missed clinic appointments and forgotten information therefore led them to carry out sub-optimal or inadequate treatment and adopt avoidance strategies. Parents felt however, that their ability to cope was increased with the support of the health care professionals (Kulczycki et al 1989, Phillips et al 1985 and Andersson-Segsten et al 1986).

Many of the authors found that the very nature of cystic fibrosis compounded the psychological distress for parents. The overwhelming chronic

uncertainty of the future led to a sense of futility and hopelessness with all matters concerning their child. Unfortunately this problem still remains an issue in cystic fibrosis today as disease severity and life expectancy still remains unpredictable.

These studies identify a history of emotional distress in parents of children with cystic fibrosis. As clinicians, we must be aware of the impact that caring for a life threatening, chronic disease has on the carers. These authors have highlighted areas of concern that have remained constant over many years therefore to ensure optimum care for our patients we must acknowledge the psychological and physical burden that parents have identified.

The consensus of the literature suggests that the concept of burden appears to be a combination of many variables including the individual character of the carer, the age of the child, the perception of the severity of the disease, the intrusion of the disease felt by the carer into their daily lives and the family adaptation to the disease. Acknowledgement of these variables as part of daily life for the parents is a far more useful method of identifying the intrusion cystic fibrosis plays. The majority of studies have examined the caring role in terms of psychopathology. Cadman, Rosenbaum, Boyle and Offord (1991) however, cautioned against the clinical tendency to interpret stress as dysfunction and heralded a shift away from a pathological approach towards the assessment of psychosocial well being.

The following chapter will put forward the aims and hypothesis of the current study based on this review of the literature and the various study designs employed.

CHAPTER 6

AIMS OF THE CURRENT STUDY

6.1 Aims and Objectives

The literature has suggested that cystic fibrosis irrevocably alters the lives of parents caring for an affected child. This intrusion into people's lives has been identified as 'burden', which can be assessed by asking individuals about their quality of life, psychological health and the perceived impact that the disease has on their daily lives. Previous studies attempting to identify this intrusion have suffered from a number of study design problems including, little or no randomisation of the study population, selection availability with cases being taken from those attending outpatient clinics or general practitioner surgeries, no matched control group, little geographical or social variation and objective opinions from professionals.

Cystic fibrosis is a relatively rare disease and therefore numbers available for inclusion into any study are small. To be able to recognise the impact of a particular chronic disease on the carers and to make the results useful in the clinical situation however, researchers must use homogeneous samples.

The aim of the current study therefore is to identify the degree of intrusion into the lives of parents who care for a child with cystic fibrosis and the stressors they encounter and recognise as causing problems, not only in their day-to-day management but on their future plans. By recognising the limitations of previous studies, this investigation will be carried out by not only comparing measures of psychopathology but also, in acknowledgement of the issues of disease management, assessment will be made of the impact the illness has on the parents of a child with cystic fibrosis.

Family life without chronic disease is never trouble free and many parents encounter a variety of problems at different ages and stages of their children's development. To account for the differences in families that may be attributed to cystic fibrosis, the study was designed to use a comparison group of parents with no chronic disease in the family. This control group was matched for marital status, number and ages of children in the family and geographical area.

The majority of studies examining burden in parents of children with cystic fibrosis have used a population centralised in one area, for example those attending the same hospital. At present, funding for cystic fibrosis care in the United Kingdom is not centrally managed but varies from one health authority to another, therefore standards of treatment may also vary between one health authority and another. In recognition of this, cases were randomly selected from regions in England, Wales and Scotland. This not only allows for nationwide differences in the National Health Service provision and the differences in

individual cystic fibrosis centre management, but also the geographical differences in social and family life.

Previous studies have used interviews or validated questionnaires, however, to strengthen this study the design has been based around the use of standardised and well validated measures of psychopathology and intrusion of disease into family life. Interviews were carried out to explore the impact of an ill child on the family, the general psychological health of the individual and their pessimism about life. Three measures were therefore selected: the General Health Questionnaire 28, Beck's Hopelessness Score and The Impact on Family Scale. These three instruments have proven useful in the assessment of psychological and social adjustment of the disease into daily lives. Interviews were carried out using a Grounded Theory methodology.

Cystic fibrosis is a disease with no cure; despite optimum treatment the health of the individual slowly declines over time ultimately ending in premature death. To explore whether time plays a part in coming to terms with the intrusion a small sub-group of cases were asked to repeat the study again, two years on from the start. Demographic data has also been collected looking at the socio-economic status of the carers, number of children in the family - with and without cystic fibrosis and parental input into the treatment regimens.

The literature has suggested areas where problems may exist i.e. emotional, social, physical and financial areas. Problems in these areas have

implications for the clinical care of the child. Due to the use of specific and validated questionnaires, case recruitment from a broad geographical area, inclusion of a control group, and the use of longitudinal data, it is anticipated that further detail and insight will be highlighted by the current study. Following the study's findings it is anticipated that future studies should be undertaken to consider the options for support that could be made available by the cystic fibrosis multi-disciplinary team to enable parents to cope with the rigors of the long term care for their children.

6.2 Hypotheses

- Mothers caring for a child with a life limiting, chronic disease (cystic fibrosis), will experience decreased quality of life and decreased psycho-social function compared to mothers caring for well children.
- Fathers caring for a child with a life limiting, chronic disease (cystic fibrosis), will experience decreased quality of life and decreased psycho-social function compared to fathers caring for well children.
- Mothers caring for a child with a life limiting, chronic disease (cystic fibrosis), will leave employment to care for their child due to treatment requirements.
- A mother's perception of her child's health status will be identified as a stressor influencing her interactions in daily life both within and outside the family.
- A father's perception of his child's health status will be identified as a stressor influencing his interactions in daily life both within and outside the family.
- The age and position of the child with cystic fibrosis in the family will influence coping.

CHAPTER 7

METHODOLOGY

7:1 Design

The current investigation is based on a prospective survey of parents who have children with cystic fibrosis, with a matched, control group of parents who have no chronic disease in the family. Longitudinal data was also collected on a sub-group to examine any change over time. Cases have been randomly selected from regions in England, Wales and Scotland, in order to account for regional differences in health provision and individual hospital management.

The study is designed around the use of standardised and validated questionnaires, and interviews based on Grounded Theory to explore the impact of an ill child on the family, the general psychological health of the individual and their pessimism about life. Demographic data was also collected including employment status, number of children in the family - with and without cystic fibrosis and parental involvement into the treatment regimens.

7:2 Recruitment of Cases

To establish an equally distributed, and fair representation from parents throughout the United Kingdom recruitment of cases had to be national. The Cystic Fibrosis Trust is the sole charity in the United Kingdom devoted to cystic fibrosis and provides a focus for fund raising and support. It was decided therefore to approach the charity for assistance with recruitment.

The Cystic Fibrosis Trust is a national charity founded in 1964 to raise money for hospital and medical research into improved treatment options for cystic fibrosis. The Trust recognises the impact of cystic fibrosis on families and offers support and advice for people with cystic fibrosis and their families through the provision of a quarterly magazine that parents contribute to, literature on all aspects of the disease and through a network of support groups set up around the United Kingdom. The Cystic Fibrosis Trust's fund raising programmes reflect this recognition and have used examples of the burden of caring for a child with cystic fibrosis in their recent poster campaigns.

The support the Trust can offer is widely recognised by clinicians in the multi-disciplinary teams, therefore all individuals with cystic fibrosis and their families are encouraged to join at the time of diagnosis. The Trust consequently holds one of the largest databases of individuals with cystic fibrosis and their families in the country.

Contact was made with the director of the Family and Adult Support Services section of the Cystic Fibrosis Trust and the use of the database for the study was discussed. The Cystic Fibrosis Trust made the database available for the current study once the experimental design satisfactorily ensured total confidentiality and had been passed by the local research ethics committee of Great Ormond Street Hospital and the Institute of Child Health. Local Research Ethics Committee approval was gained in October 1996. (Please see appendix 1)

7:3 Sample Size

To calculate the sample size required for the current study, a power calculation was used on all three questionnaires (Wade, 1995).

$$n > \frac{2F\sigma^2}{d^2} + 2F \left(\frac{\sigma}{d} \right)^2$$

n = sample size

F = the constant required to detect a difference significant at 0.05 with a power of 80%

σ = standard deviation (based on preliminary data)

d = smallest difference considered to be of scientific importance (based on earlier work).

The difference in the mean for each questionnaire was calculated by deducting the total mean scores from each other, the standard deviation for each

questionnaire was taken from the scores. Using a significance level of 0.05 with a power of 80%, $F = 7.85$.

General Health Questionnaire:

$$15.7 \frac{(5.33)^2}{2.77} = 58$$

Becks Hopelessness Scale:

$$15.7 \frac{(3.98)^2}{2.63} = 36$$

Impact of Illness Score:

$$15.7 \frac{(7.63)^2}{9.57} = 10$$

Therefore to detect a significance level of 0.05 and a power of 80%, the study needed to recruit a total of 116 cases - 58 cases and 58 controls.

One hundred parents of children with cystic fibrosis were invited by post, to take part in the investigation. Information sheets with a tear off reply slip, plus a stamped, self-addressed envelope were put in un-addressed envelopes and sent to the Cystic Fibrosis Trust. 100 names were randomly selected from geographical regions around the United Kingdom from the database, address labels were printed and the envelopes sent off. This ensured that at no time would the investigator have any knowledge of to whom the information sheets

had been sent and confidentiality of the individual names on the database remained protected.

7:4 Information Sheets

The information sheets described the background, purpose of the study and what taking part would involve (*fig 1*). Individuals were invited to return the tear off reply slip if they wanted further information or wanted to take part in the study. At this point, if they had chosen to return the slips they were asked for their name, telephone number and address. It was hoped that by returning the slips only those individuals willing to take part would voluntarily give their names to the investigator and confidentiality of those not wishing to take part would be maintained. In this way consent for participation in the study was also assumed. When the reply slips were returned telephone contact was made and more detail about the study explained, individuals were then asked whether they would like to take part or not. Those individuals wishing to take part were entered into a computerised database held only by the investigator. *Table 2* illustrates the questionnaires used and data collected from each group.

Figure 1: Information Sheet

Parental quality of life when a child has cystic fibrosis.

Are you interested in taking part in a research study?

This is a study that is being carried out throughout the UK and aims to discover problems parents may have looking after a child (of any age) with cystic fibrosis (CF). As many of you will recognise the last few years have brought about a change in the management of people with CF with the majority of treatment regimes being carried out at home, from daily chest physiotherapy to the administration of intravenous antibiotics and nasal ventilation.

Although, as parents, you are willing to carry out varied treatment regimens at home, the demands made upon you in terms of practical skills and time is increasing. Despite the obvious benefits that treating your child at home can bring, we recognise that this regime of care can affect all aspects of your quality of life, personal and psychological health and social life.

The study is directed at parents and, with the aid of three self completing questionnaires and in some cases interviews, will allow parents to tell us how they feel about CF and the effect it has had on their lives.

The results of this study will allow us to identify areas that help and support could be offered in the future to make day to day coping easier and improve lives generally.

If you would like to take part in this study please return the slip below and you will be contacted by telephone to have further details explained to you.

Table 2: Data collection for each group

	Cases	Controls
Demographic data	✓	✓
Level of clinical service	✓	✗
Perception of child's health	✓	✗
Main carer	✓	✗
General Health Questionnaire	✓	✓
Becks Hopelessness Scale	✓	✓
Impact of Illness	✓	✓

7:5 Measures

To discover the effect, both psychologically and socially, that caring for a child with cystic fibrosis has on parental quality of life, three questionnaires were chosen. These were the (a) Impact on Family Scale, which highlighted the practical and social impact of having a chronically ill child on the parents and family, (b) Beck's Hopelessness Score, that identified pessimism and a feeling of hopelessness about the future, and (c) The General Health Questionnaire – 28 that identified the psychological status of the individual.

The publishers of the Beck's Hopelessness Score and the General Health Questionnaire were contacted for permission to use the measures and the author of the Impact of Family Scale was contacted for permission. In all cases, permission was granted for use in the current study. (Please see appendices 2, 3 & 4)

The Impact on Family Scale (Stein and Riessman 1980) (please see appendix 7)

This measurement tool has been used in many areas of chronic disease such as spina bifida, ventilator dependence, post-traumatic brain injury, behavioural disturbance and asthma (Loebig 1990, Quint, Chesterman, Crain, Winkleby and Bryce 1990, Sheeber and Johnson 1992, Su, Kemp, Varigos and Nolan 1997). Stein et al (1980) developed this modified version for use in studies of children with and without chronic illness conceptualised in various dimensions using factor analysis. These are - the financial situation, social interaction within

and outside the home, subjective distress felt by the parents, and a positive sense of mastery that may emerge from coping with the stress. An example would be 'Additional income is needed in order to cover my child's experiences.' Cases are asked to respond to statements with 'strongly agree', 'agree', 'disagree' and 'strongly disagree', at the same time they are asked to state whether this was 'fully', 'partly' or 'not at all' related to their child's health. Analysis is designed to elicit whether the behaviour was related to the child's health or not.

Scores are coded as to whether the answer 'not at all' related to the child's illness and then scored in reverse direction - eg 1=4, 2=3, 3=2, 4=1. Questions are grouped into five headings: financial impact, disruption of planning, caretaker burden, familial burden and coping. All the scores are added together to give a final score with the exception of coping that is kept as a separate score. There are no norms for this questionnaire; rather the authors suggest comparison with a control group.

The General Health Questionnaire - 28 item version (Goldberg and Hillier 1979)

(please see appendix 6)

This measure was designed to be a self administered, screening test and identifies four factors derived by factor analysis. These factors identify areas of symptomatology but are not necessarily diagnostic of psychiatric illness. They include somatic symptoms, anxiety and insomnia, social dysfunction and severe depression. An example of these questions would be 'Have you recently found

everything getting on top of you?' Cases are asked to answer each question using one of four given statements, eg. 'not at all', 'no more than usual', 'rather more than usual' and 'much more than usual'.

A score of 0 is assigned to the first two statements and 1 to the second two statements. Each sub-group score is totalled and then all scores are totalled. There is no agreement on the best threshold score for the GHQ - 28 but the modal value has been quoted as 4/5 (Goldberg et al 1979), therefore for the purposes of this study scores over 4 were identified as being abnormal.

Beck's Hopelessness Score (Beck and Steer 1993) (please see appendix 8)

This scale is a twenty-item measurement tool designed to identify the extent of negative attitudes (pessimism) about the future felt by adolescents and adults. Although originally designed to detect suicide patients considered to be at risk, it has since been used in normal adolescent and adult populations. The scale consists of twenty true/false statements that assess the extent of negative expectancies about the immediate and long-term future. An example of the statements given is 'I look forward to the future with hope and enthusiasm.' Nine of the statements are given a false value and eleven are given a true value, the higher scores then indicate a greater hopelessness.

Each statement is followed by two empty circles, one to be filled in if the case feels the statement to be true, and the other if it is false. Scoring is carried out using a template that lines up over the circles with a guide to the circles that

carry a score, these are then totalled. Guidelines indicate that scores of 9-14 are moderate and those greater than 14 are severe.

7:6 Interviews

In order to further explore the way that parents of children with cystic fibrosis cope with their unique situation, a series of interviews were carried out on a random sample of mothers and fathers using Grounded Theory. This methodology has been portrayed as a problem-solving endeavour concerned with understanding action from the perspective of the human agent (Haig 1996).

Grounded Theory was first described by (Glaser and Strauss 1967) as an emergent research methodology using observation, conversation and interview to understand what was happening within a given research situation. Grounded Theory is based on the premise of collecting and analysing qualitative data with the aim of discovering the theory within the given data rather than testing a hypothesis. This methodology therefore allows the study of human behaviour and interaction by generating theories about social and psychological phenomena.

Research is based traditionally on the premise of hypothesis testing; criteria universally accepted for judging the rigour of a study must therefore be abandoned when using an emergent methodology. Grounded Theory uses a different rigour, as this methodology is responsive to the situation in which the research is being carried out. Throughout the study period there will be a

continuing search for evidence driven by the accumulation of data that will eventually describe the final theory most likely to provide a good fit to the situation (Dick 2001).

The three main elements of Grounded Theory have been described as concepts, categories and propositions (Pandit 1996). Theories cannot be developed directly from raw data, rather from the conceptualisation and analysis of the data. This is not done sequentially; instead data collection and data analysis occur simultaneously allowing constant comparison. The second element of Grounded Theory is categories, equivalent to themes. As data are collected the researcher will be constantly reviewing and assessing, through categorising and sub-categorising the information. The final element – propositions, describe the links developing within and between categories that emerge through the study. In this way theory generation is inductive, with categories emerging from the data and becoming more focused as the research progresses. Coding of the categories allows theoretical propositions to become clear and as these links emerge and eventually become saturated, data is no longer added and analysis begins. Categories are described as saturated when no additional information can be found to develop them further.

Melia (1982) states that it is the researcher's responsibility to produce an account of the subjects' view of the situation with analysis of this viewpoint producing analytical concepts however, empathy with the case depends on the researcher's knowledge of the situation. Collection of quantitative data from

cases in the first part of this study allows a certain degree of knowledge therefore providing areas to explore in the interviews.

Finally, to illustrate the impact that cystic fibrosis has on the life of a parent; one of the personal accounts given by a case will be presented as a case study.

7:7 Demographic Data Collection

In addition to the standardised measures demographic data were also collected using a questionnaire. This included information about employment details both before and after the birth of their child from both parents, marital status, family composition including number of children with and without cystic fibrosis with their ages, treatment responsibility for the sick child within the family and the health status of the child at the time of completing the questionnaires, as perceived by the parent (Abbott et al 1995). As this study examines the psychosocial influences of cystic fibrosis, parental perceptions of health status rather than clinical scores were chosen as an evaluation. Studies have shown that both patients and carers view disease severity from a wider perspective, incorporating into their perceptions the social and mental health aspects of their life quality in addition to their child's physical limitations imposed on them by cystic fibrosis. It is suggested that this seeming discrepancy to the physicians

caring for them may afford emotional protection therefore influence the impact of the disease (Abbott et al 1995).

In recognition of the disparity of clinical services available throughout the UK cases were also asked to grade the service they received using the guidelines described by the Clinical Standards Advisory Group's report on Cystic Fibrosis Services in the United Kingdom (1993). Since the inception of this study however, the cystic fibrosis service provision throughout the United Kingdom has been re-evaluated and the recommendation now is that all patients attend a Specialist Cystic Fibrosis Centre (equivalent to a Level I or Level II Centre) at least twice a year.



Poster reproduced with permission from the Cystic Fibrosis Trust

7:8 Selection Of Controls

Controls were required to be matched as closely as possible for employment status, geographical area, marital status and number of children in the family. As cases were being recruited from all over the United Kingdom, it was felt that the only way to recruit matched controls was to ask the cases to nominate friends or colleagues who they felt were in a similar situation to themselves. The only criteria given were that the 'control' parents would not be caring for anyone with a chronic illness within the family. This process was explained to those volunteering to participate in the study during the initial telephone conversation, each individual was asked to consider two parents who they thought may fit the criteria and would not mind taking part; details were collected at a later date.

The names and addresses of the possible controls were asked for through completion of a form enclosed with the questionnaires. When these were returned the investigator chose one of the nominated names and contacted them using a similar information sheet as the cases were sent, inviting them, if they were interested, to take part in the study. If the reply slips were returned telephone contact was made, as in the case group.

7:9 Data Collection

After consent had been gained from the cases, packs containing the three questionnaires, the demographic data sheets and a form for information on possible control parents were sent out with a stamped, self-addressed envelope enclosed.

After six months, non-responders were sent an identical pack with a letter asking if they were still willing to take part in the study. If they were unwilling to participate, they were asked to return the pack in the stamped, self-address envelope provided.

To collect longitudinal data a sub-group of twenty-five were sent the same pack of questionnaires two years later. Sixteen cases responded. These results were matched and compared to data collected from the same cases at the beginning of the study.

When the controls had been identified they were also sent packs containing the three questionnaires and a demographic data sheet with the questions about children with cystic fibrosis, treatment responsibility, health status and clinical service grading omitted. Again, they were followed up after six months and asked if they would still like to take part in the study.

Following analysis of the quantitative data, interviews were then carried out with a random selection of mothers and fathers using the Grounded Theory approach. To allow parents a choice, interviews were carried out either Great Ormond Street Hospital for Children or at the family home.

7:10 Data Storage

As questionnaires were returned they were scored and data entered onto a database using the Statistical Package for the Social Sciences (SPSS) programme to allow for statistical analysis to take place. (License number - 30525 50782 41409 94745 74354 7091)

It was acknowledged that error might occur in scoring and data entry. Validation was therefore carried out by independently scoring all questionnaires, these scores were then compared and where discrepancies occurred scores were re-checked independently and compared again before entry into the database by two people. Identification numbers were assigned to each individual before entry onto the data, it was therefore impossible to identify any person from the data stored on the computer. The following figure (*fig 2*) demonstrates the areas that questionnaires were returned from around the United Kingdom.

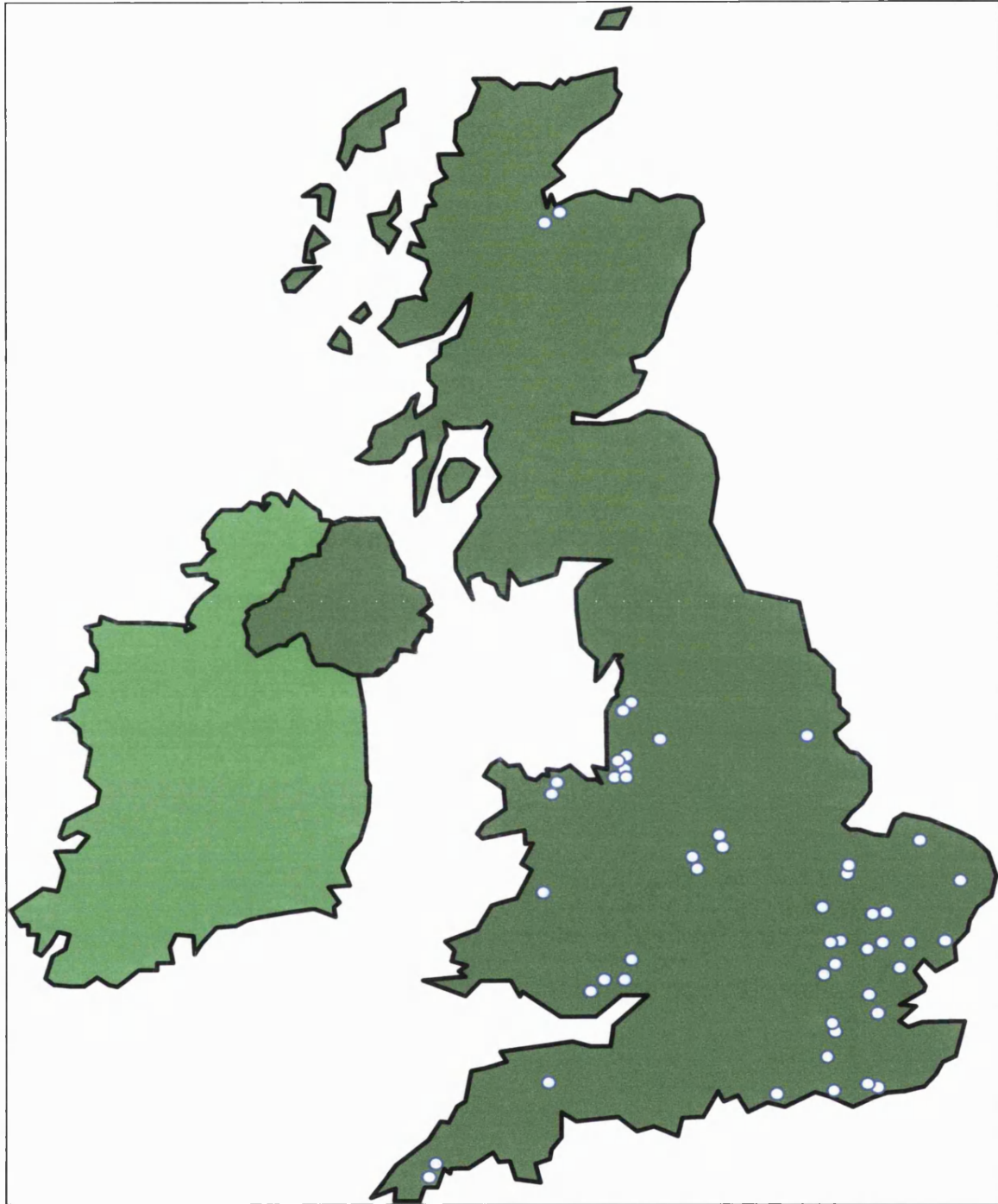


Figure 2: Areas of the United Kingdom that questionnaires were returned from

CHAPTER 8

RESULTS - 1

8.1 Study Population

One hundred invitations were anonymously posted to parents of children with cystic fibrosis, 80 individuals returned the tear off slip stating their interest in the study and asking for a telephone call to explain further, all 80 individuals agreed to take part in the study and were sent the pack of questionnaires. 65 (65%) of the questionnaires were returned from the case group (37 female) (*table 3*). Parents in the same case group were then asked to nominate two friends of the same sex, same marital status, with the same number of children in the family and from the same geographical area. One of these two names was then randomly chosen and the same process for recruitment was used – an invitation pack, 59 (91%) from the control group (37 female) (*table 6*). Longitudinal data were requested from 20 of the original case group, 15 (75%) responded.

Although packs were only sent to those people agreeing to take part after information was given over the telephone, fifteen of those who previously agreed did not respond - seven did not return the questionnaire packs. Those who did return the uncompleted packs (n=8) gave reasons such as: 'no time to fill in paperwork', 'it's a waste of time when I've got to look after my son', 'I refuse to

have anything to do with psycho questions'. The non-responders were from geographic areas throughout the United Kingdom.

Demographic data were collected from both groups (cases and controls), including number of children in each family, marital status, and employment history. Cases were additionally asked about the number of children with cystic fibrosis, perception of the health of their child with cystic fibrosis and asked to grade the level of clinical service they received for their child.

8.2 Demographic Data - Cases

All cases (28 male, 37 female, mean age 41 years) reported that their children with cystic fibrosis lived at home and that there had been no previous deaths due to cystic fibrosis. Parents were asked how old their child had been at diagnosis: 73% were diagnosed under one year, 16% before two years, 8% between two and five years and 3% between five and ten years of age. Data were analysed for demographic variables using descriptive statistics. The following table (*table 3*) illustrates the gender differences and mean age in the case group.

Table 3 Gender difference and mean age in case group

Total Group (n= 65)	Mean age (range)
Male = 28 (43%)	42 years (25 – 62)
Female = 37 (57%)	39 years (25 – 51)
Total mean age	41 years (25 – 62)

8.2.1 Family Constellation

Cases were asked how many children they had living at home (mean: 2.4, range: 1-6), with and without CF, their ages (mean age: 10.6 years, range: 1-27 years) (table 4). The following table illustrates the family constellation in the case group.

Table 4 Distribution of children in case families

Case Families (n=65)	
number of children with CF	74
number of children without CF	83
total number of children	157
mean number of children per family	2.4
mean age (range)	10.6 years (1 - 27)

8.2.2 Clinical Service

Cases were asked to grade the level of clinical service they received for their child/children with cystic fibrosis using the guidelines from the Clinical Services Advisory Group Report (1993). Centres were graded from Level 1 to Level 4; almost half the parents surveyed stated that the CF centre they attended was a

Level I centre (40%). The following table gives a description of each level as identified by the cases. (*table 5*)

Table 5 Level of clinical service

LEVEL OF CLINICAL SERVICE	Number = 65 (percentage)
1	26 (40%)
2	18 (28%)
3	9 (14%)
4	12 (18%)

8.3 Demographic Data - Controls

This first table (*table 6*) illustrates the gender differences and mean ages in the control group.

Table 6 Gender difference and mean age in control group

Total Group (n= 59)	Mean age (range)
Male = 22 (37%)	39 years (26 – 50)
Female = 37 (63%)	36 years (26 – 48)
Total mean age	39 years (26 – 50)

8.3.1 Family Constellation

Controls were asked how many children they had living at home (mean: 2.1, range 1-3), and their ages (mean age: 9.4 years, range: 1-21 years). The following table (*table 7*) illustrates the family constellation in the control group.

Table 7 Distribution of children in control families

Control Families (n=59)	
total number of children	127
mean number of children per family	2.1
mean age (range)	9.4 years (1-21)

In order to establish whether the control (n=59) and case (n=65) group differed in any of the matched variables (gender, marital status, current employment and number of children), independent t-tests were carried out (*table 8*). No significant differences were found for gender: $p=0.5$, marital status: $p=0.3$, and number of children in each family: $p=0.5$. The only variable on which they differed significantly was employment status (unemployed versus employed): $p<0.05$. The following table illustrates the comparison of the matched variables between the control and case groups

Table 8 Comparison of matched variables for cases and controls

Variables	cases vs controls
gender	$p=0.5$
marital status	$p=0.3$
current employment	$P<0.05$
number of children	$p=0.5$

CHAPTER 9

RESULTS - 2

9.1 QUALITY OF LIFE AND PSYCHOSOCIAL FUNCTION

Hypothesis : Mothers and fathers caring for a child with cystic fibrosis will experience decreased quality of life and decreased psychosocial function compared to mothers and fathers caring for well children

9.1.1 Comparisons - Cases Versus Controls

As the data were non-parametric all questionnaire scores were analysed using Mann Whitney U tests; cases versus controls.

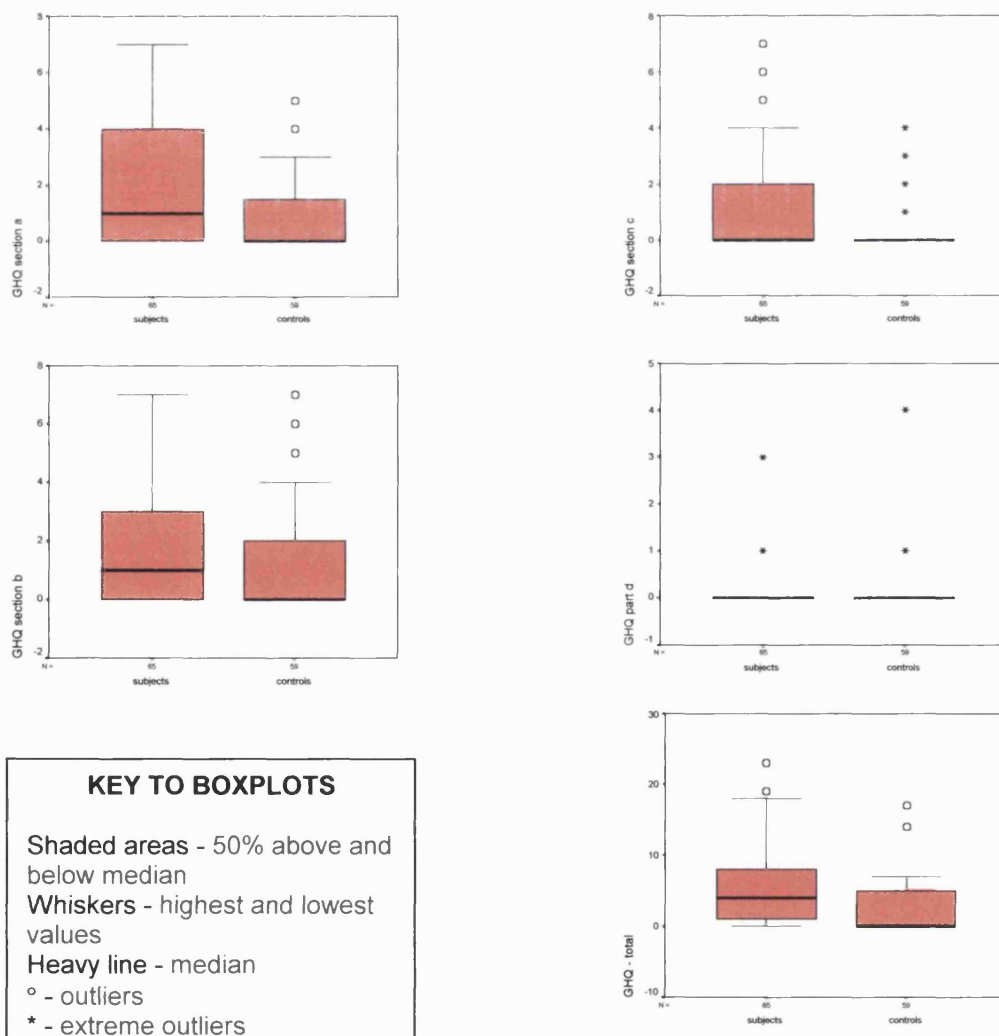
GENERAL HEALTH QUESTIONNAIRE There are significant differences between the case and control group in the three sections of the General Health Questionnaire - *somatic symptoms, anxiety and insomnia*, and *social dysfunction*. There were no significant differences in the section *severe depression*, with very low scores from both groups. The median total score for the case group showed significance (cases: 4, controls: 0). The inter-quartile range for the case group rose well above the threshold score quoted as 4/5. This table (*table 9*)

demonstrates the comparison of the General Health Questionnaire between the case and control groups.

Table 9 General Health Questionnaire - cases versus controls
median score (inter-quartile range)

	cases n=65	controls n=59	p
section a.somatic symptoms	1 (0-4)	0 (0-2)	<0.03
section b anxiety and insomnia	1 (0-3)	0 (0-2)	<0.003
section c social dysfunction	0 (0-2)	0 (0-0)	<0.01
section d severe depression	0 (0-0)	0 (0.0)	0.3
total score	4 (0.5-8)	0 (0-5)	<0.001

Figure 3 Boxplots for General Health Questionnaire Scores Cases versus Controls

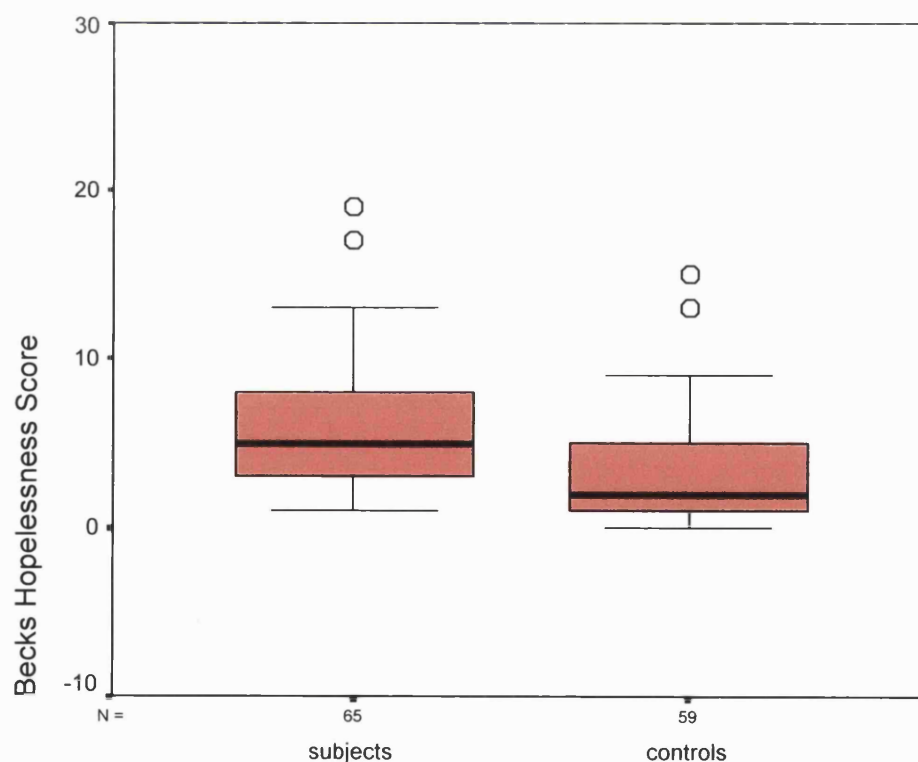


BECKS HOPELESSNESS SCORE A significant difference is shown in the Beck's Hopelessness Score with a difference in median score of 3 points (cases: 5, controls: 2). The table below (*table 10*) illustrates the comparison of this score between the case and control groups.

Table 10 Beck's Hopelessness Score - cases versus controls
median score (inter-quartile range)

	case n=65	control n=59	p
total score	5 (3-8)	2 (1-5)	<0.001

Figure 4 Boxplot of Becks Hopelessness Score Cases versus Controls

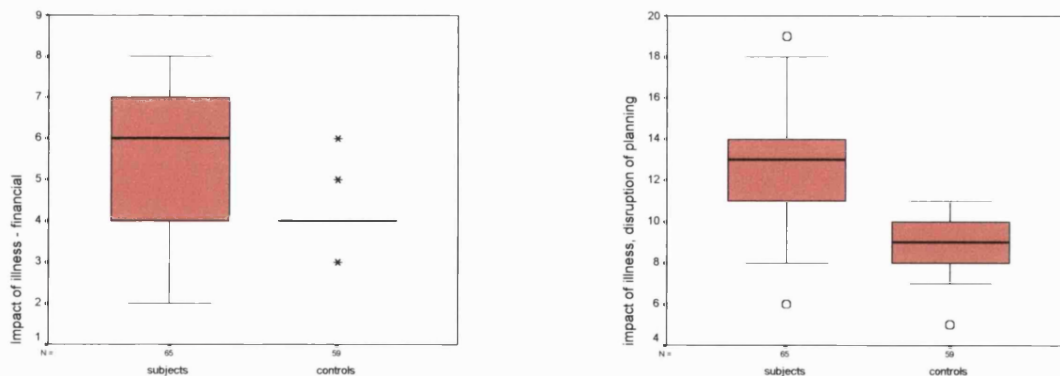


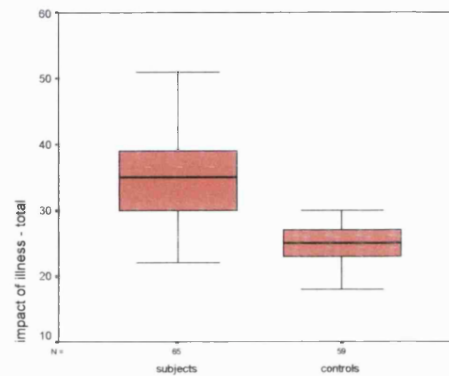
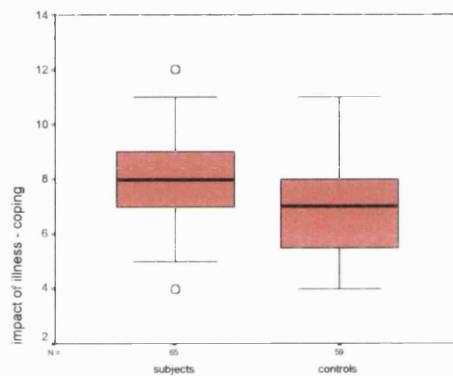
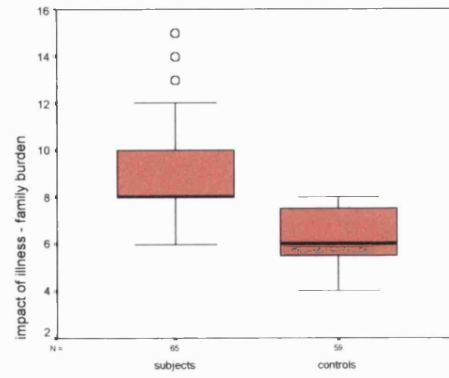
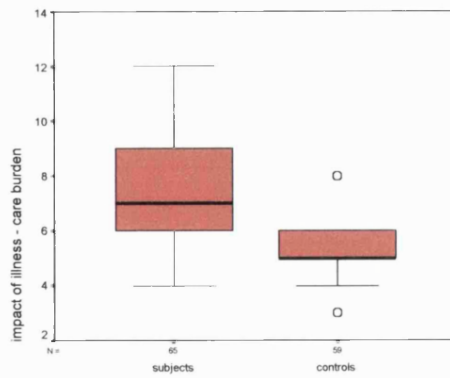
IMPACT ON FAMILY All sections of the Impact on Family Questionnaire demonstrate a highly significant difference between the two groups with a difference in the total median scores of 10 points. The table below (*table 11*) illustrates the comparison of the two scores between the case and control groups.

Table 11 Impact on Family - cases versus controls
median score (inter-quartile range)

	cases n=65	controls n=59	p
financial	6 (4-7)	4 (4-4)	<0.001
care burden	7 (6-9)	5 (5-6)	<0.001
disruption to planning	13 (11-14.5)	9 (8-10)	<0.001
family burden	8 (7.5-10)	6 (5-8)	<0.001
total score	35 (30-39)	25 (23-27)	<0.001
coping	8 (7-9)	7 (5-8)	0.004

Figure 5 Boxplot of Impact of Family Questionnaire Scores Cases versus controls





9.1.2 Gender

In the case group, although there are no significant differences found between men and women in the General Health Questionnaire, Impact on Family Questionnaire or the Beck's Hopelessness Score women consistently scored higher than the men in all the questionnaires. The following three tables (12, 13, 14) all demonstrate the comparison of mothers versus fathers with the General Health Questionnaire, the Impact on Family Scale and the Beck's Hopelessness Score using Mann Whitney U tests.

Table 12 Gender (mothers versus fathers) with General Health Questionnaire
(GHQ) score median score (inter-quartile range)

	mothers n=37	fathers n=28	p
GHQ section a	1 (0-4)	0 (0-3.75)	0.9
GHQ section b	2 (0-3.5)	0.5 (0-3)	0.2
GHQ section c	0 (0-2)	0.5 (0-2)	0.7
GHQ section d	0 (0-0)	0 (0-0)	0.1
GHQ total score	5 (1.5-8.5)	2 (0-7.5)	0.3

Figure 6 Boxplot for General Health Questionnaire total score and gender

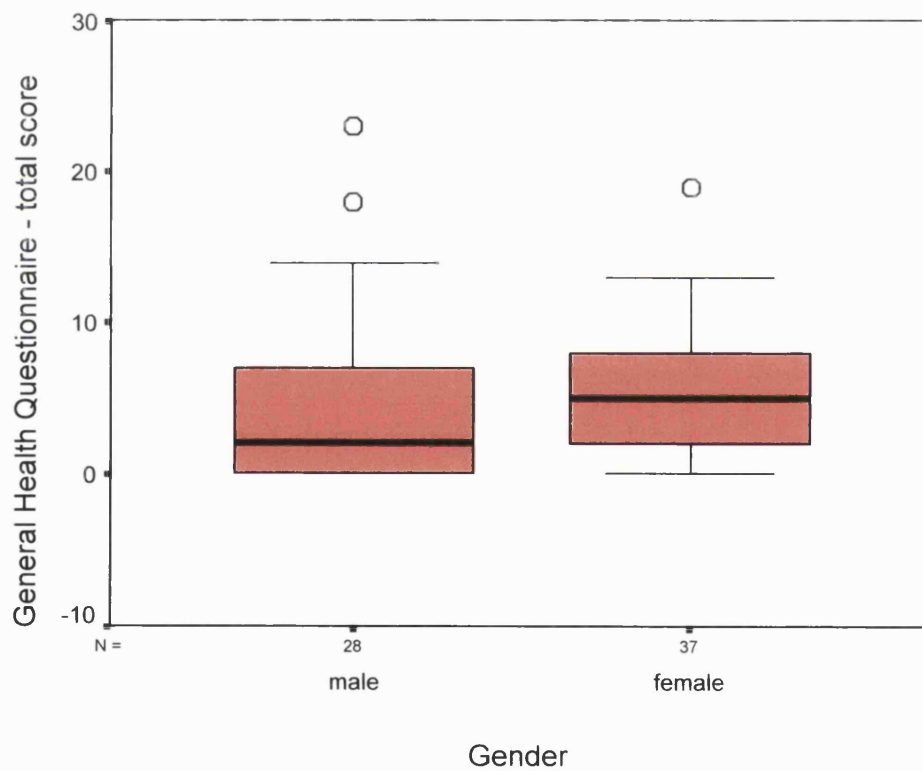


Table 13 Gender (mothers versus fathers) with Impact on Family (IoF)
Questionnaire score median score (inter-quartile range)

	mothers n=37	fathers n=28	p
IoF financial	6 (4-7)	6 (4-7)	0.9
IoF care burden	7 (7-9)	7 (6-8.75)	0.1
IoF disruption to planning	14 (11-15)	12.5 (11-14)	0.3
IoF family burden	9 (8-10)	8 (7-9.75)	0.1
IoF total score	37 (30-40)	34 (30-36.75)	0.3
IoF coping	9 (7-9.5)	8 (7-8)	0.1

Figure 7 Boxplot for Impact on Family total score and gender

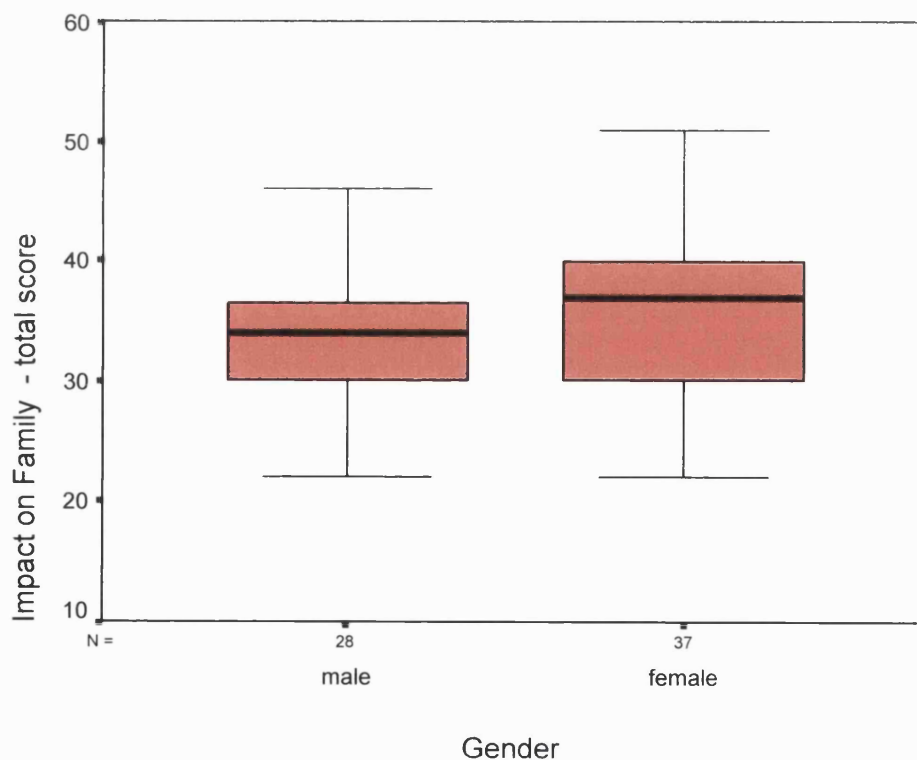
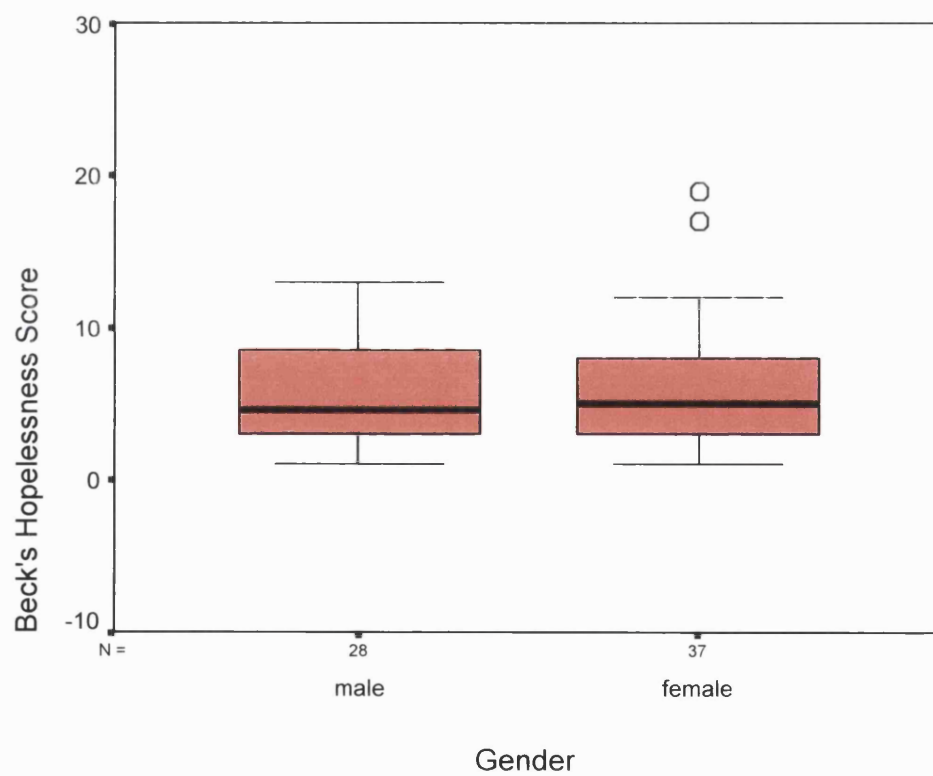


Table 14 Gender (mothers versus fathers) with Beck's Hopelessness Score
median (inter-quartile range)

	mothers n=37	fathers n=28	p
Beck's	5 (3-8)	4.5 (3-9.25)	0.8

Figure 8 Boxplot for Beck's Hopelessness total score and gender



9.2 EMPLOYMENT STATUS

Hypothesis : Mothers caring for a child with cystic fibrosis will leave employment to care for their child due to treatment requirements

Cases were asked about employment; whether they had been employed before the birth of their children, whether they were currently employed and whether their partner was employed. They were also asked about type of employment they were engaged in and whether it was full (f/t) or part time (p/t) (*table 15*). Whilst the majority of cases were employed full time (48%) around a quarter were in part time employment (26%) and a quarter unemployed (26%). This was also true for their partners. In comparison previous employment shows that only 6% of parents were in part time employment whereas 68% were in full time employment. The following table (*table 15*) illustrates the employment status of the cases, males and females together.

Table 15 Employment history of cases

	previous employment			current employment			partner employed		
	yes		no	yes		no	yes		no
	f/t	p/t		f/t	p/t		f/t	p/t	
parents	44 (68%)	4 (6%)	17 (26%)	31 (48%)	17 (26%)	17 (26%)	39 (60%)	13 (20%)	13 (20%)

The data for mothers and fathers were then analysed separately, as illustrated below (*table 16*).

Table 16 Employment history of mothers and fathers - cases

	previous employment			current employment		
	yes		no	yes		no
	f/t	p/t		f/t	p/t	
mothers	19 (51%)	4 (11%)	14 (38%)	6 (16%)	14 (38%)	17 (46%)
fathers	25 (89%)	0	3 (11%)	25 (89%)	3 (11%)	0

9.2.1 Control group

Controls were asked about employment; whether they had been employed before the birth of their children, whether they were currently employed and whether their partner was employed. They were also asked about the type of employment they were engaged in and whether it was full (f/t) or part time (p/t) (*table 17*). All controls had previously been in full time employment, with the majority (83%) remaining in full time employment as did their partners (78%). The following table illustrates the employment history of the control group.

Table 17 Employment history of controls

	previous employment			current employment			partner employed		
	yes		no	yes		no	yes		no
	f/t	p/t		f/t	p/t		f/t	p/t	
controls	59 (100%)	0	0	49 (83%)	6 (10%)	4 (7%)	47 (80%)	5 (9%)	7 (11%)

As with the cases group the data for for mothers and fathers were then analysed seperately, as illustrated below (*table 18*).

Table 18 Employment history of mothers and fathers - controls

	previous employment			current employment		
	yes		no	yes		no
	f/t	p/t		f/t	p/t	
mothers	37 (100%)	0	0	27 (73%)	6 (16%)	4 (11%)
fathers	22 (100%)	0	0	22 (100%)	0	0

9.2.2 Comparisons within the case group

The following analysis will make comparisons between the cases and all the variables.

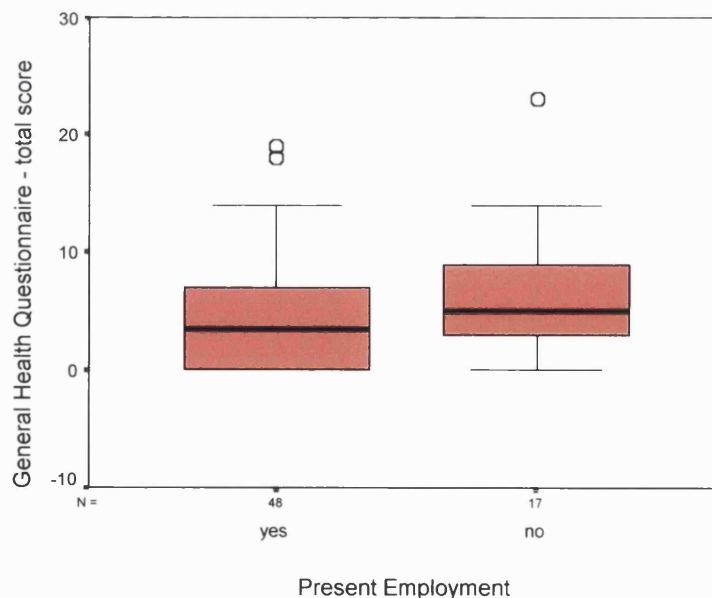
The effect of employment status on the questionnaire results of the cases (*tables 19, 20, 21*), was compared the using Mann Whitney U test.

Although there is no significant difference seen in the General Health Questionnaire between the employed cases and the unemployed cases, the unemployed group scored consistently higher. The table below (*table 19*) demonstrates the comparison of employment (employed versus unemployed) with the General Health Questionnaire.

Table 19 Employment (employed versus unemployed) with General Health Questionnaire median score (inter-quartile range)

	employed n=48	unemployed n=17	p
GHQ section a	0 (0-3)	2 (0-4)	0.2
GHQ section b	1 (0-3)	2 (0.5-5)	0.2
GHQ section c	0 (0-2)	0 (0-1.5)	0.9
GHQ section d	0 (0-0)	0 (0-0)	0.8
GHQ total score	3.5 (0-7)	5 (2.5-9.5)	0.2

Figure 9 Boxplot for the General Health Questionnaire and employment status

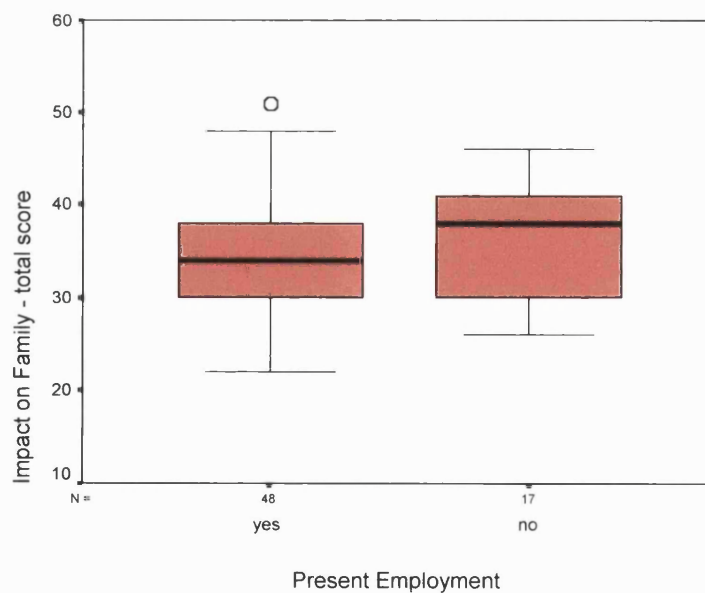


Again there was no significance found between the employed and unemployed groups on the Impact on Family Score, although the unemployed group continued to score higher. This is demonstrated in the table below (*table 20*).

Table 20 Employment (employed versus unemployed) with Impact on Family Questionnaire median score (inter-quartile range)

	employed n=48	unemployed n=17	p
loF financial	6 (4-7)	4 (4-6.5)	0.3
loF care burden	7 (6-9)	7 (6.5-10)	0.2
loF disruption to planning	12.5 (11-14)	14 (11-15)	0.4
loF family burden	8 (7-10)	9 (8-11.5)	0.2
loF total score	34 (30-38)	38 (29.5-41.5)	0.4
loF coping	8 (8-9)	7 (5.5-9)	0.1

Figure 10 Boxplot for the Impact on Family total score and employment status



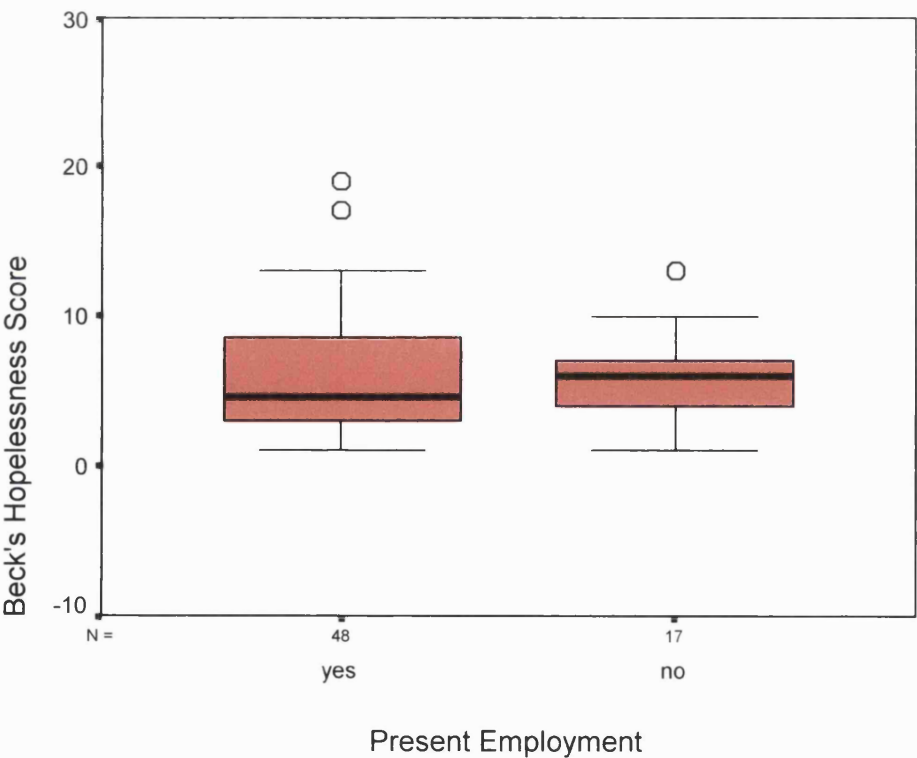
As with the previous two analyses there is no significant difference in employment status seen between the two groups in the Beck's Hopelessness

Score, although the unemployed group do score higher. This can be seen in the table below (table 21).

Table 21 Employment (employed versus unemployed) with Becks Hopelessness median score (inter-quartile range)

	employed n=48	unemployed n=17	p
Beck's	4.5 (3-8.75)	6 (3- 5.7)	0.5

Figure 11 Boxplot for Beck’s Hopelessness Score and employment status



9.3 HEALTH STATUS

Hypothesis : Mothers' and fathers' perception of their child's health status will be identified as a stressor influencing their interactions in daily life both within and outside the family

Cases were asked how well they thought their child with cystic fibrosis had been in the two weeks previous to completing the questionnaire. Options given were: 'well', 'slightly unwell' and 'requiring treatment'. (*table 22*) The majority of parents reported their child as being well at the time, just under a quarter perceived their child as being slightly unwell or requiring treatment. The following table illustrates the cases' perception of health at the time of the study.

Table 22 Parents health perception of child with CF

PARENTAL PERCEPTION OF HEALTH	Number n=65 (percentage)
well	40 (62%)
slightly unwell	12 (18%)
requiring treatment	13 (20%)

9.3.1 Comparisons within case group

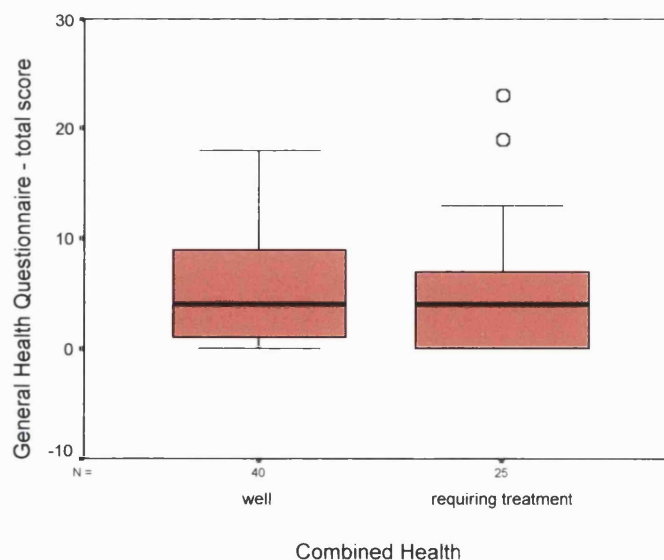
Case variables - perceptions of health (*tables 23,24,25*), were compared to the questionnaire results using Mann Whitney U tests. The group was divided into 'well' and 'those requiring treatment' to allow for analysis.

In the comparison of 'well' children versus the group 'requiring treatment', there was no significant difference in the General Health Questionnaire. The following table (*table 23*) demonstrates the comparison of the two variables – 'well' versus 'requiring treatment' - with the General Health Questionnaire.

Table 23 Perception of health ('well' versus 'requiring treatment') with General Health Questionnaire median score (inter-quartile range)

	well n=52	requiring treatment n=15	p
GHQ section a	1 (0-4)	0 (0-2.5)	0.5
GHQ section b	1 (0-3)	1 (0-3.5)	0.8
GHQ section c	0 (0-2)	0 (0-3.5)	0.8
GHQ section d	0 (0-0)	0 (0-0)	0.6
GHQ total score	4 (1-9)	4 (0-7)	0.6

Figure 12 Boxplot for the General Health Questionnaire total score and health status



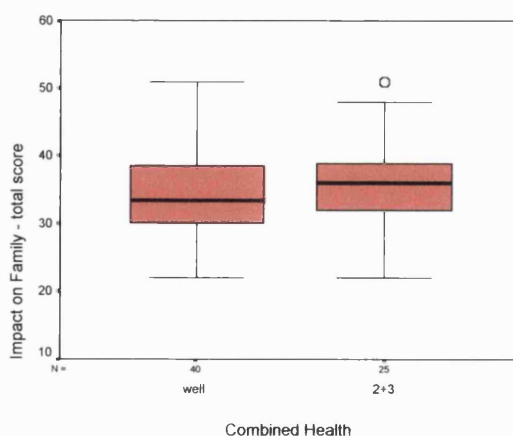
Again there was no significant difference seen in the Impact on Family questionnaire. However the total median score was higher in the group 'requiring treatment'. The table below (*table 24*) demonstrates the comparison of the two variables – 'well' versus 'requiring treatment' - with the Impact on Family Score.

Table 24 Perception of health ('well' versus 'requiring treatment') with Impact of Family Questionnaire median score (inter-quartile range)

	well n=52	requiring treatment n=15	p
loF financial	6 (4-6)	6 (4-7)	0.2
loF care burden	7 (6-9)	7 (6.5-9)	0.9
loF disruption to planning	12. 5 (11-15)	14 (11.5-14)	0.5
loF family burden	8 (7-10)	9 (8-10)	0.5
loF total score	33.5 (30-38.75)	36 (32-39.5)	0.3
loF coping	8 (7-9)	8 (7-10)	0.4

Figure 13 Boxplot for the Impact on Family total score and health status

(2+3 = combines slightly unwell with requiring treatment)

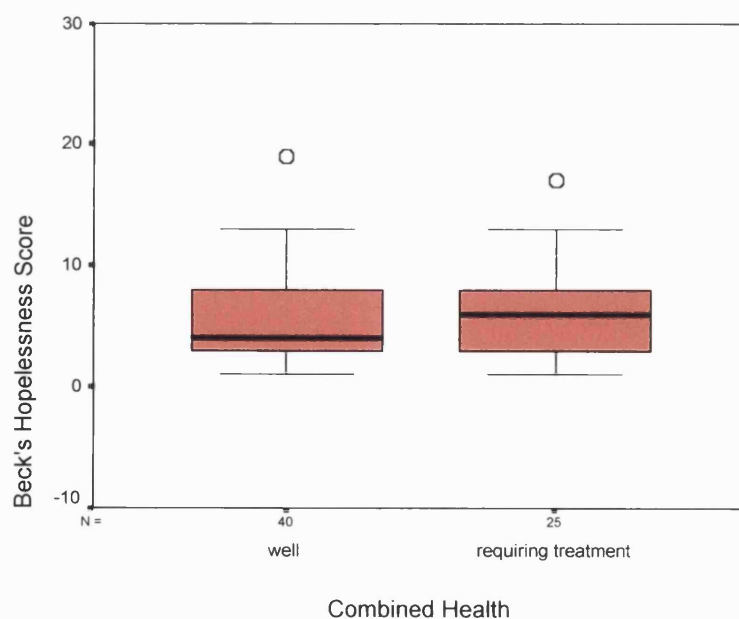


Again, as in the comparisons with the other questionnaires, the Beck's Hopelessness Score, although showing no significant difference demonstrated between the 'well' group and the group 'requiring treatment', both scored higher than the control group (well: 4, unwell: 6 - *table 25*, control: 2 - *table 10*). The comparison of these two variables with the Beck's Hopelessness Score is shown in the next table (*table 25*).

Table 25 Perception of health ('well' versus 'requiring treatment') with Beck's Hopelessness Score median (inter-quartile range)

	well n=52	requiring treatment n=15	p
Becks	4 (3-8)	6 (3-8)	0.6

Figure 14 Boxplot for the Beck's Hopelessness Score and health status



9.3.2 Carer status

As well as asking parents how well they perceived their child to be at the time of completing the questionnaires, they were also asked who they identified as the main carer (CF care) of the child/children with CF. Options given were: mother, father, child with CF, other (nanny or helper), or a combination of family members. (*table 26*) Neither the mothers or fathers identified fathers as the main carer; overall mothers were seen as the main carers for their child with CF (63%). The following table illustrates the division of treatment care as identified by cases.

Table 26 Distribution of main carers in case families

MAIN CARER	Number n=65 (percentage)
mother	41 (63%)
father	0
child	7 (11%)
other (eg nanny)	1 (1%)
combination of family members	16 (25%)

9.4 Position of child with cystic fibrosis in the family

Hypothesis: The age and position of the child with cystic fibrosis in the family will influence coping

Table 27 Position in family of child/children with Cystic Fibrosis

Position in Family	1 st child with CF	2 nd child with CF
only child	6 (9%)	-
first	31 (48%)	-
second	18 (28%)	1 (2%)
third	8 (12%)	4 (6%)
fourth	2 (3%)	4 (6%)

There are no significant differences seen in any of the questionnaire scores when comparing the oldest child having cystic fibrosis with the affected child being placed elsewhere in the family. The comparison with the position of child with the General Health Questionnaire (*table 27*), the Impact on Family Scale (*table 28*) and the Beck's Hopelessness Score (*table 29*) can be seen in the following three tables.

Table 28 Position of child (first versus other) with General Health Questionnaire (GHQ) median score (inter-quartile range)

	1st child n=37	other n=28	p
GHQ section a	1 (0-4)	1 (0-2.75)	0.7
GHQ section b	1 (0-4.5)	2 (0-3)	0.7
GHQ section c	0 (0-2)	0.5 (0-2)	0.9
GHQ section d	0 (0-0)	0 (0-0)	0.4
GHQ total score	4 (0-9)	3.5 (1-6)	0.8

Figure 15 Boxplot for the General Health Questionnaire and position of child in family

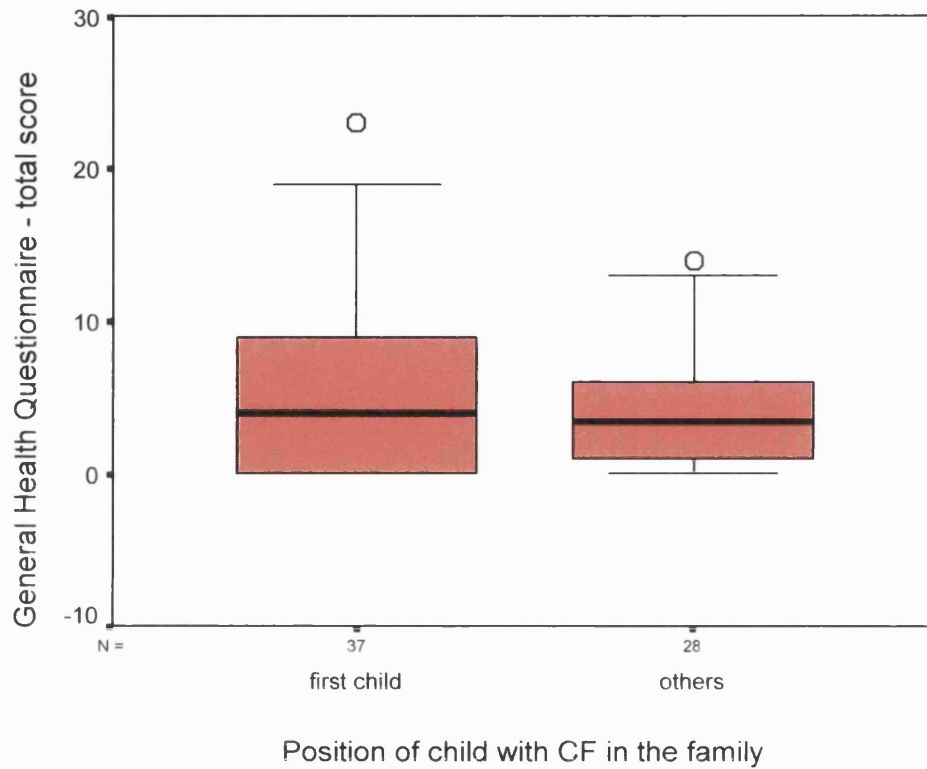


Table 29 Position of child (first versus other) with Impact on Family Questionnaire (IoF) median score (inter-quartile range)

	1st child n=37	other n=28	p
IoF financial	6 (4-7)	6 (4-7)	0.8
IoF care burden	7 (6.5-9)	7 (6-9)	0.5
IoF disruption to planning	13 (11-14.5)	12. 5 (11-14.75)	0.9
IoF family burden	9 (7.5-10)	8 (7.25-9)	0.1
IoF total score	36 (30-39)	33 (30-39.5)	0. 3
IoF coping	8 (7-9)	8 (6.25-9)	0.4

Figure 16 Boxplot for Impact on Family total score and position of child with CF
in the family

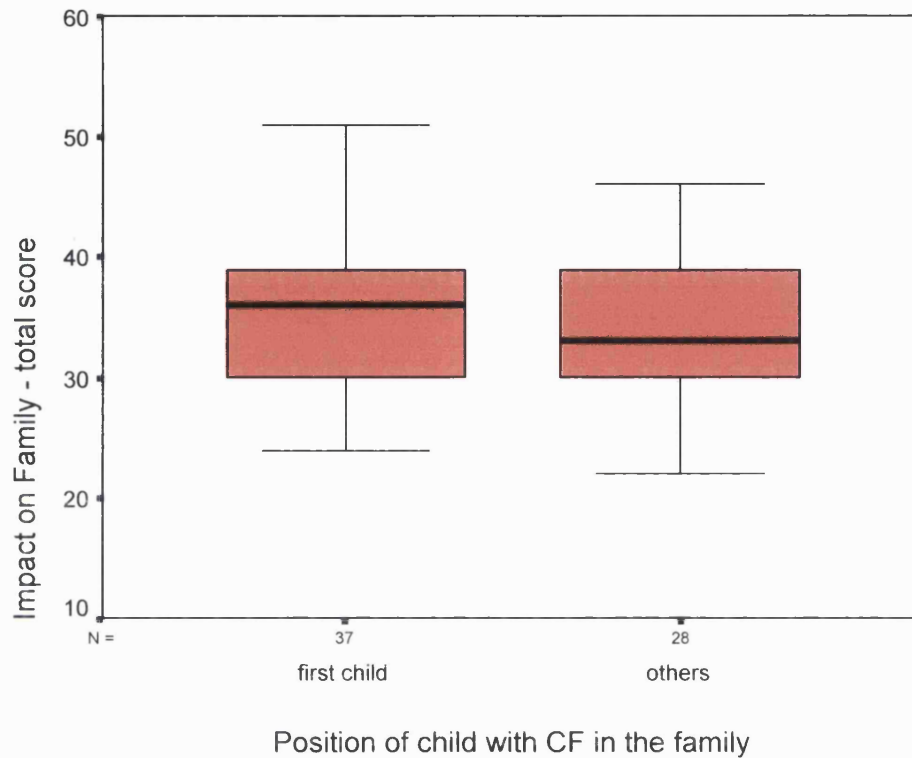
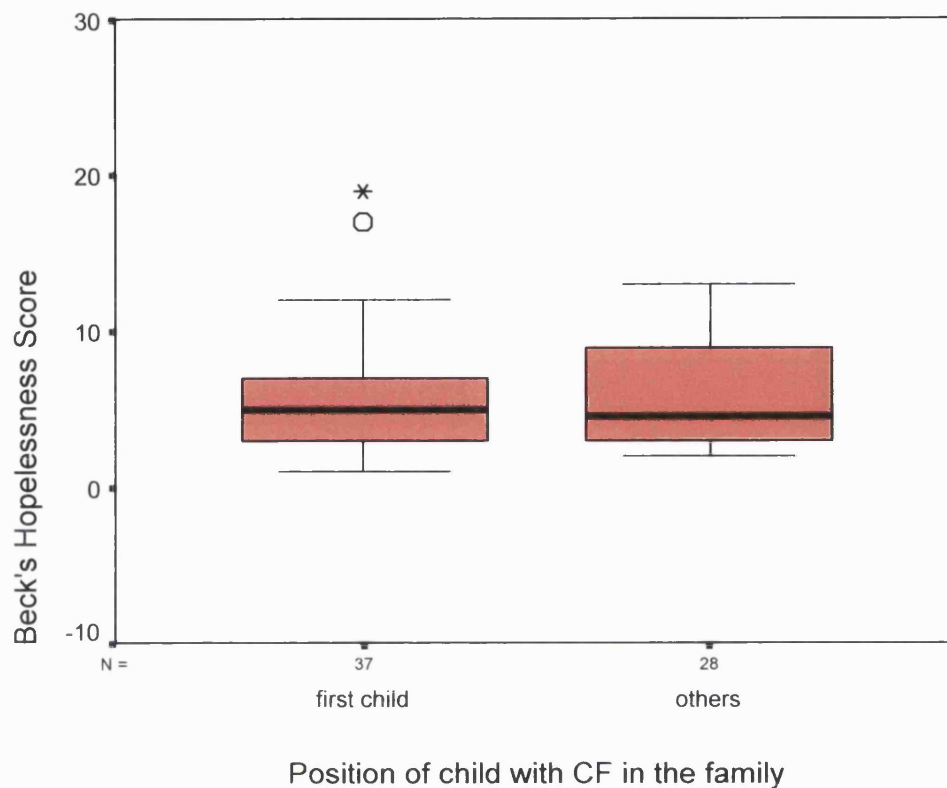


Table 30 Position of child (first versus other) with Beck's Hopelessness Score
median score (inter-quartile range)

	1st child n=37	other n=28	p
Beck's	5 (2.5-7.5)	4.5 (3-9.5)	0.5

Figure 17 Boxplot for Beck's Hopelessness Score and the position of the child with CF in the family



9.5 Exploratory Analysis

The aim of this analysis was to discover which co-variables had the greatest influence on the Beck's Hopelessness Score using multivariate analysis. To examine the association between the ages of the parents, the number of children with cystic fibrosis in each family, parental employment status, the perceived health status of the child with cystic fibrosis, the level of clinical service received

and the questionnaire scores a Pearson's Correlation analysis was therefore carried out (tables 31, 32).

For both men ($p=0.02$) and women ($p=0.01$) a link was found between increasing age and their perception that their child with cystic fibrosis was sicker. Men expressed an increasingly severe depression where they had more than one child with cystic fibrosis ($p=0.01$), if they felt an increasing burden of care ($p=0.04$) and if they were unemployed ($p=0.01$). Men also appeared to describe a negative correlation between financial burden and increased somatic symptoms ($p=0.01$) and social dysfunction ($p=0.01$) – in other words a decrease in somatic symptoms and social dysfunction led to an increase in financial burden. Women found it more difficult to cope with the burden of care when they were employed ($p=0.03$), when they reported an increase in anxiety and insomnia ($p=0.03$) and where they reported a poor clinical service ($p=0.04$).

Table 31 Correlation of demographic data and questionnaire scores – Male

	Pearson's Correlation (R)	2 tailed significance
Father's age with Perceived child's health status	0.44	0.02
GHQ section a with. IoF financial burden	-0.46	0.01
GHQ section c with. IoF financial burden	-0.47	0.01
GHQ section d with Number of children with CF	0.47	0.01
GHQ section d with Employment status	0.47	0.01
GHQ section d with IoF care burden	0.4	0.04

Table 32 Correlation of demographic data and questionnaire scores – Female

	Pearson's Correlation	2 tailed significance
Mother's age with Perceived child's health status	0.42	0.01
IoF coping with Employment status	-0.36	0.03
IoF care burden with Level of clinical service	0.34	0.04
GHQ section b with IoF care burden	0.34	0.034
GHQ total score with IoF care burden	0.38	0.02

9.5.1

Exploratory analysis of the questionnaire scores demonstrated a high correlation between the Beck's Hopelessness Score and the General Health Questionnaire 28. The General Health Questionnaire is a heterogeneous measure and in this study appears to be equally well expressed as a measure of hopelessness therefore, as the Beck's Hopelessness Score provided greater discrimination it was used as an indicator of mental health in further analysis (*table 33*).

Table 33 Correlation of Beck's Hopelessness Score with the General Health Questionnaire

	Beck's Hopelessness Score	
	Pearson's Correlation (R)	2 tailed significance
GHQ total	0.4	0.03

Variables were then analysed using linear stepwise regression with Becks' Hopelessness Score as the dependent variable, to search for either a variable that could predict a declining mental health (indicated by a high Beck's

Hopelessness Score) or a number of variables that could be used as a predictive index. (tables 34,35,36).

The previous analysis indicates a different relationship for men and women separately and together as a group, therefore the analysis was carried out on men, women and the group as a whole.

In all the analysis the variables entered in a stepwise regression included: number of children with cystic fibrosis, employment status, perceived health status of the child with cystic fibrosis, level of clinical service, the General Health Questionnaire (excluding total score) and the Impact on Family (excluding the total score).

Men described anxiety and insomnia (section b of the General Health Questionnaire) as the significant predictor of their poor mental health ($p=0.022$), women described social dysfunction ($p<0.001$) (section c of the General Health Questionnaire), and financial burden ($p=0.43$) (of the Impact on Family Scale) and together parents identified social dysfunction ($p<0.001$) and in increase in a disruption to planning as significant predictors of their poor mental health ($p=0.015$).

Table 34 Linear Regression with Beck's Hopelessness Score as dependent variable – Male

Model	Unstandardised Coefficients		Standardised Coefficients	t	p
	B	Std error	Beta		
(constant)	4.463	0.822		5.429	0.000
GHQ section b (Anxiety and insomnia)	0.724	0.298	0.430	2.429	0.022

Table 35 Linear Regression with Beck's Hopelessness Score as dependent variable – Female

Model	Unstandardised Coefficients		Standardised Coefficients	t	p
	B	Std error	Beta		
(constant)	0.317	2.049		0.155	0.878
GHQ section c (social dysfunction)	1.346	1.347	0.522	3.886	0.000
IoF financial burden	0.757	0.359	0.283	2.107	0.043

Table 36 Logistic Regression with Beck's Hopelessness Score as dependent variable – Combined Male and Female

Model	Unstandardised Coefficients		Standardised Coefficients	t	p
	B	Std error	Beta		
(constant)	-0.223	1.982		-0.112	0.911
GHQ section c (social dysfunction)	1.104	0.262	0.451	4.219	0.000
IoF – disruption of planning	0.380	0.151	0.268	2.509	0.015

The following chapter will present the longitudinal data from the follow-on part of the questionnaire study, comparing the matched pair data from the same cases at time zero and two years on.

CHAPTER 10

RESULTS - 3

10.1 Longitudinal Data From Sub-Group Of Cases

The same data were collected from a sub-group of cases (cases¹) two years following the initial completion of the original questionnaires (cases²). The proportion of men to women completing the study has stayed the same (previous study, male: 43%, female 57%). The table below (*table 37*) shows the gender difference in the case² group.

Table 37 Gender difference in case² group number=15

Cases ² (n=15)	
male	6 (40%)
female	9 (60%)

10.1.1

Cases² were asked how many children they had living at home, with and without CF, and their ages. The mean number of children per family in this sub-group has increased over the two year period from 2.3 children to 3.8 children. The table below shows the distribution of children in the case² families (*table 38*).

Table 38 Distribution of children in case² families

	Case² families
number of children with CF	17
number of children without CF	20
total number of children	37
mean number of children per family	3.8
mean age (range)	10 years (1 - 29)

10.1.2

Cases² were asked about employment; whether they or their partner continued to be employed or not two years on from answering the first questionnaires and whether it was full (f/t) or part time (p/t). The data has changed slightly over the two year period with unemployment in the sub-group increasing from 26% to 34% and from 20% in their partners to 26%. The following table illustrates this (*table 39*).

Table 39 Employment history of cases

	current employment			partner employed		
	yes		no	yes		no
	f/t	p/t		f/t	p/t	
cases	46%	20%	34%	60%	14%	26%

10.1.3

Cases² were asked which family member they identified as the main carer (CF care) of the child/children with CF. Options given were: mother, father, child with CF, other - nanny or helper, or a combination of family members. As in the main study, the father is reported to take no part in direct care as a single carer, although he is included when the family report a combination of family members taking part in care (cases¹: 27%, cases²: 46%). The table below illustrates the distribution of carers in the families (*table 40*).

Table 40 Distribution of main carers in case² families

MAIN CARER	
mother	40%
father	0%
child	14%
other (eg nanny)	0%
combination of family members	46%

10.1.4

Cases² were asked how well they thought their child with CF had been in the two weeks prior to completing the questionnaire. Options given were: well, slightly unwell and requiring treatment. Again, in this sub-group, both mothers and fathers perceived that their children were predominantly well at the time of completing the questionnaires (cases¹: 73%, cases²: 70%). The following table (*table 41*) demonstrates this.

Table 41 Parents health perception of child with CF

PARENTAL PERCEPTION OF HEALTH	
well	70%
slightly unwell	12%
requiring treatment	18%

10.2 Comparisons - Cases¹ versus Cases²

As the data was non-parametric Wilcoxon Signed Ranks tests were used to compare the data between the beginning of the study (cases¹) and the results from the same group two years later (cases²).

GENERAL HEALTH QUESTIONNAIRE Although there are no significant differences between the cases at the beginning of the study and two years on, median total scores were higher in the main study (*table 9*). The table below

(table 42) demonstrates the differences between the cases at the beginning and end of the study with the General Health Questionnaire.

Table 42 General Health Questionnaire - cases¹ versus cases²
median score (inter-quartile range)

	cases¹	cases²	p
section a somatic symptoms	3 (0-4)	3 (0-4)	0.62
section b anxiety and insomnia	2 (1-7)	1 (0-3)	0.3
section c social dysfunction	1 (0-2)	1 (0-3)	0.75
section d severe depression	0 (0-0)	0 (0-1)	0.41
total score	6 (2-13)	5 (3-11)	0.34

BECK'S HOPELESSNESS SCORE There are no significant differences seen in the questionnaire, either between the same cases two years apart, or the cases in the current analysis and those in the main study (table 10). The table below (table 43) demonstrates the differences in the scores.

Table 43 Beck's Hopelessness Score - cases¹ versus cases²
median score (inter-quartile range)

	cases¹	cases²	p
total score	4 (3-8)	5 (2-10)	0.57

IMPACT ON FAMILY The difference between cases¹ and cases² on the variable identifying *financial* problems in the family was significantly lower ($p < 0.01$) two years on for parents (cases¹: 6, cases²: 4) as was the median total score (cases¹: 36, cases²: 29). This is illustrated in the table below (*table 44*)

Table 44 Impact on Family - cases¹ versus cases²

median score (inter-quartile range)

	cases¹	cases²	p
financial	6 (4-7)	4 (4-6)	0.01
care burden	8 (7-9)	6 (6-9)	0.14
disruption to planning	14 (11-17)	11 (10-14)	0.1
family burden	9 (7-13)	8 (7-9)	0.44
total score	36 (32-46)	29 (27-36)	0.02
coping	8 (7-9)	8 (7-9)	0.9

The following chapter will present the data and discuss the results of the interviews carried out using Grounded Theory.

CHAPTER 11

RESULTS – 4

11.1 Interviews - Structure

The structure for the first interview was based on results from the questionnaires used in the first part of the study. Areas of concern identified from the analysis as highly significant included financial implications, employment, family burden and social dysfunction. The interviews were carried out using a grounded theory approach and then analysed using the concepts of coding, categorising and conceptualisation (Stern 1980). During each interview extensive notes were taken and following each one the information was coded before being grouped into categories. As the interviews progressed a constant comparison of categories arising from each subsequent interview was made. This process allowed the categories to be linked and refined and eventually theoretical labels were identified. Throughout the analysis memos were made which helped with the organisation of thoughts and ideas in relation to the data. A colleague was invited to review the notes and categories to allow for an unbiased approach.

11.2 Demographic Data

Data were collected from the group of parents being interviewed of whom 63% were females with a mean age of 36 years (range 29 – 49 years). The mean age of the children with cystic fibrosis was 10 years (range 2 – 17 years). The majority of mothers (92%) were in full time employment before the birth of their child with cystic fibrosis, with only 50% returning to part time work following the diagnosis. All fathers were in full time employment both before and after the birth and diagnosis of the child. The demographic data are presented in the table below (*table 45*).

Table 45 Demographic data of interviewed cases

Female / Male	12 / 7
Mean age of parent (range)	35 (29 – 49 years)
Mean age of CF child (range)	10 (2 – 17 years)
Employed pre diagnosis - mothers (full time / part time / not employed)	11 (11 / 0 / 1)
Employed post diagnosis - mothers (full time / part time / not employed)	6 (0 / 6 / 6)
Employed pre diagnosis - fathers (full time / part time)	7 (7 / 0)
Employed post diagnosis - fathers (full time / part time)	7 (7 / 0)

11.3 Interviews

Ten sub-categories were originally identified from the interviews and from those, four main categories were eventually settled on with agreement from the same colleague.

These categories were:

- Burden and intrusion
- Employment and financial implications
- Relationships
- Knowledge

Burden and intrusion

All the parents interviewed commented on the intrusion that treatment regimens made on their family life. It was felt that holidays, day trips and even the average school day had to be carefully timetabled so that physiotherapy, nebulisers or tablets could be organised to fit in to their arrangements. Parents commented on the fact that they often had to abandon their plans, as they felt that it was impossible to carry out both the treatment requirements and go out. Holidays abroad were universally thought to be too much trouble to contemplate, and although two of the interviewees had been abroad with their children they did not feel that it was something they could do again for a while.

Both mothers and fathers reflected that their choice of home, school, general practitioner, local hospital and even holidays had often been made with the child with cystic fibrosis in mind, although they admitted to not necessarily having realised this at the time. All parents were conscious that these decisions, such as moving to a new home chosen to be near a school that is well known for pastoral care or a general practitioner that accepts expensive patients such as those with cystic fibrosis, may well have been to the detriment of their other well children.

There were other issues that parents reported as less important, yet still felt guilty about, such as not allowing their child to attend children's parties as they did not trust the host to administer enzyme replacement therapy or planning their (parents) own social lives. One parent, who had to go away over night, commented that it took so long to prepare everything for the babysitter that she did not think that she could ever repeat the experience.

Both parents identified mothers as the main carers with mothers describing a feeling of an overwhelming psychological burden of responsibility for their child, not only during periods of sickness, but also when they were well. This burden was felt to be constantly in their lives and yet not shared by the father. One mother of two children with cystic fibrosis said that the disease had completely taken over her life and that it had become a significantly greater burden with the diagnosis of a second child.

Mothers also talked about feeling an extra emotional burden through being responsible for constantly monitoring their child's disease status. Again fathers were thought not to pay enough attention to the subtle changes in their child that would indicate a possible deterioration, and some were thought not to have any idea at all, relying totally on the mother to recognise any physical symptoms. Mothers described some resentment as they felt that this responsibility was expected of them, not only by their husbands but also by relatives, friends and the medical team caring for their child.

Whereas mothers said that they took each day at a time, fathers appeared to be more forward thinking and commented on the uncertainty of the future for their child with cystic fibrosis, as a father said "life can be very difficult living with the trauma of not knowing the future". Another father was worried that his son would never be able to leave home and that he and his wife would have to remain as carers for the rest of their lives. Other fathers were concerned about the issues of male infertility and that their sons would not be able to get married and have children. Interestingly, it was the fathers and not the mothers who appeared to be more concerned with this issue.

In retrospect, parents remember that although the thought of taking full responsibility for their child after being discharged for the first time was "scary", they concluded that by remaining "as optimistic as possible" and "taking each day as it comes", caring for their child was manageable and often rewarding.

Employment and financial implications

In the group of parents interviewed, all fathers were employed full time, although two were self-employed. Mothers however, had changed their employment status having had a child with cystic fibrosis. Many parents commented on the sudden drop in finances following the diagnosis when the mother gave up work and some felt angry that there were no state benefits to compensate for this change in status.

Parents talked about cystic fibrosis causing problems for them at work, for example they often had to take time off when their child was unwell or to attend hospital appointments. Many felt that they could not repeatedly ask their employer for time off from work, especially for hospital visits and therefore tended either to take their annual leave at these times or take unpaid leave. Mothers either did not return to work at all following the diagnosis or returned to work part time after their child had started school. These jobs tended to fit in with school hours (eg, taking in ironing), and in some instances were at their child's school (eg, as a midday supervisor) so that they were available if they were needed at school and at home when their child finished school. This type of employment was chosen so that they would have the time to be at home to carry out all the treatments, as one mother explained. Mothers who had previously been in professional positions felt that they had had to abandon their career and take on work that would fit around their caring role.

As a result of this change in employment status within the families, all parents felt that they had some financial difficulties. Many fathers described having to work longer hours as they were now the sole bread-winner and one father said that he felt that he had to work much harder as he did not expect his son to work, therefore he would have to support his son for ever. As a result of fathers working longer hours, mothers felt that they had to do much more for their children as they described their husbands as "never being around".

Relationships

All parents were aware of changes in their own marital relationship, their relationships with their healthy children and with their friends and relations. One mother said that she felt that her role had changed and she had now become the stronger one in the partnership often guiding her husband in decision making. Parents commented that for the first few years after diagnosis they had felt very emotional and wanted to hide their child away from friends and relatives. During that time their perception of life had changed and they described feeling very isolated. One mother explained it as "looking out at another world where healthy children lived". Mothers particularly described a special bond between them and the child with cystic fibrosis and one mother commented that there was a special bond between her mother, herself and her child. Another father said that his parents paid special attention to the well siblings in the family as the maternal grandparents spoilt the child with cystic fibrosis.

Parents acknowledged that their healthy children suffered as a result of the cystic fibrosis. Well siblings were described as 'bringing themselves up', or that the well siblings had 'to take a share in responsibility for treatment' – especially if they attended the same school. One older sibling was seeing a counsellor and several had had sessions with the psychologist at the Cystic Fibrosis Centre. Throughout the interviews parents commonly reported that their well children had often wished to have cystic fibrosis so that they too could have more time and attention from their parents.

Parents however appeared to be surprised and pleased about the relationships existing between the well siblings and children with cystic fibrosis. Many sibling relationships were described as very close, with in one instance two sisters often sharing a bed and two brothers sharing a bedroom, despite there being a spare room in the house. Only one mother raised some concern about the closeness of the relationship, commenting that the well sibling "...will not be able to cope when his sister dies".

Support networks, and help from relatives and friends were all thought of as important aids to coping, especially when treatment regimens and childcare were trusted to others. Some parents described the importance of The Cystic Fibrosis Trust and its role in providing information and arranging local educational or support meetings. The Cystic Fibrosis Teams at each hospital however, were seen as the primary source of support, both psychosocial and clinical. Parents admitted that in some instances, as a family, they saw more of

the Cystic Fibrosis Team than their own relatives and therefore felt very attached to team members. Parents felt that as individuals they had a great deal of experience and knowledge to offer and all said that they remember the time immediately prior to and after diagnosis as being particularly stressful and would especially like to be able to support parents of newly diagnosed children or parents who had children younger than their own.

Knowledge

Parents described knowledge of cystic fibrosis as an essential part of coping and that this knowledge was constantly being increased and challenged. Knowledge of cystic fibrosis was felt to be important from the beginning – as one parent commented, "knowledge of CF helps you organise your life". There were however warnings about the way knowledge can be gained with parents making comments such as: "Don't take on too much, too soon" or "Find out simple information early on", and "Don't read too much information, too quickly". One mother said that she found meeting older children with cystic fibrosis more helpful than reading information leaflets.

Parents are aware of the many sources of information and that some may come from dubious places such as some tabloid newspapers or the Internet. They were keen to caution other parents to find out the truth and only pay attention to the facts. Many parents discussed the role of the media and raising public awareness. Television programmes for example, medical documentaries or soap operas, were both avoided by parents as they were felt to either over

dramatise the facts or give false hope. Newspaper reports, although not avoided, were read with a degree of scepticism as "they always promise an imminent cure, but nothing ever happens". It was felt that fund raising tended to concentrate on the negative side of cystic fibrosis for example the treatment demands and life expectancy. Parents acknowledged the need to gain public sympathy in a dramatic way and, although recognising the importance of such campaigns, described the effect that it had on the family and their children with cystic fibrosis. One thirteen year old girl had seen a fund raising poster describing a life expectancy of thirty years, she now has stopped co-operating with treatment as she said to her father, "What's the point, I'm going to die soon anyway?"

Parents felt it was important that it was not only they who had an understanding of the disease but that their children, relatives and friends also showed an interest. Many parents were angry that there was not enough knowledge of the disease in the general public and thought that there should be far more information available.

Parents agreed that the provision of knowledge should not only be from professionals. Rather they felt that they should support and advise each other, either on a one-to-one basis or at meetings. One mother went into schools, clubs and offices to teach the general public about cystic fibrosis and all parents felt a strong sense of responsibility to take part in fund raising activities for The Cystic Fibrosis Trust, one father saying that he had raised over £100,000.

Cultural differences in dealing with cystic fibrosis, although not causing overwhelming problems, did however raise some issues not necessarily considered by professionals. A Spanish family commented that whenever they visited relatives in Spain they had problems with adhering to their son's treatment regimens, as the Spanish culture involves taking long rests in the afternoon and eating late in the evening. The parents said that they always had to leave early so that they could get home to do chest physiotherapy and give medication and they believed that this inevitably offended their relatives, consequently they avoided visiting their family. Some Asian parents, although not all, reported disagreements about treatment. Fathers tended to be more indulgent towards their sons and often felt that mothers were insisting on overdoing treatment, carrying out chest physiotherapy for too long for example. Mothers however, complained that fathers were undermining their responsibility and they were just doing what they had been told to do. The Greek and Italian parents described the close involvement of the extended family acknowledging both the practical and emotional support this offered them. All however, felt that they were not able to make decisions about their child's health or treatment without intervention from other family members and expressed a wish to be allowed to 'get on with looking after their child without interference'.

The next section of this chapter will highlight one individual's experience as a parent of a young child with cystic fibrosis and the impact the disease has had on her personal life and on the family.

11.4 Case Study – Valerie's Story

Hello, my name is Valerie. I am 37 years old. I have a husband and two daughters, Alison aged ten years and Anna aged 2½ years. Life was hard growing up in London with my Greek Orthodox parents, immigrants from Cyprus. I am the eldest of seven and I had my fair share of responsibility for all my siblings. When I was young I had an arranged marriage but it did not work out well so we divorced. I feel so lucky to have met Greg, the man of my dreams and have a family with him. Apart from a few medical problems such as asthma and osteochondritis of the hip there is really nothing that separates me from any other thirty something trying to bring up a family. Perhaps there is one exception, the miscarriages that I had between my daughters. I was very worried that there was something wrong with me, so when I eventually fell pregnant with Anna my husband and I were very happy.

That excitement did not last long however, at my 20 week scan they found an abnormality in the baby's bowel. Once investigated further nothing was found and I was assured it was nothing serious, however after this I was never quite settled. I did not feel pregnant. I had a sinking feeling about it. I sometimes mentioned to my husband that I thought there was an alien not a baby inside me. When Anna was born – two weeks early - she was grossly underweight and went to the Special Care Baby Unit for observation. However, after this initial worry she seemed to maintain her weight and we were allowed home.

The first few weeks of Anna's life were quite worrying for me. My beautiful baby had constant diarrhoea and looked like she was in a lot of discomfort. The milk was just running out of her as soon as she drank it. She also had nasty chest infections and cough. I took her to my GP a number of times and in a short space of time – under a month – she had been admitted to our local hospital twice. She was investigated there

for 'failure to thrive', diarrhoea and wheezing. I knew there was something wrong with my baby I could just feel it. I spent every night with her in the hospital, just in case anything went wrong. After doing loads of blood tests on her they found nothing. That made me even more worried. They decided to refer her to a specialist hospital for more investigations. I had not been home with my husband and daughter for ten days now and I was torn. I wanted to look after my family but I also could not leave my baby in case anything happened to her. She was in the specialist hospital in case anything happened to her. She was in there for a while so I was offered a room and as it was the summer holidays Alison came down to stay too. That was valuable because that was the first time that I had really had time to be with her since Anna was born.

They went through test after test and nothing was found. There was a strange kind of limbo feeling, wanting to find out what was wrong, but also not wanting to find anything too badly wrong with her.

One day a doctor told me that they had one more test to do. This was the sweat test for cystic fibrosis. The day the results came back I was sitting with Anna in my arms by her cot. Then, a team of doctors arrived and told me that she had cystic fibrosis. Well I just screamed, what was this all about. The name sounded so scary, how could they say that something like that could be wrong with my baby? I ordered them to go away and leave me in peace, they left and I just sobbed my heart out with this baby in my arms. I had no idea what cystic fibrosis was but I knew that it was something that my baby could not possibly have. I phoned my husband who had been working all this time; he could not accept it either. 'They must have made a mistake, there is nothing like that in our family', he said.

Soon after the diagnosis the staff on the ward started to teach me about cystic fibrosis and how to manage it. It was hard for me to learn the physiotherapy techniques,

I felt like I was beating her. I was finding it very difficult to accept the diagnosis and started to reason with myself that this in fact was not my real baby, she had died, so they had decided to give me this one instead as at least she was alive.

They had tried to introduce me to another mother of a child with cystic fibrosis - I didn't want to know. That would be like accepting that she had it. I just wanted to stay in the corner and hide my baby from everyone. Then one day the mother and I crossed paths on the ward. We looked at each other and burst into tears hugging each other. Watching me go through it all brought it back for her. That was a big point of support for me. Just to know I was not the only one.

They were worried about my personal health, they thought I needed a break but I would not leave Anna, who else was there to look after her, what would I do? But I couldn't leave, what if something happened, if something went wrong? Gradually however, I started to take breaks away from the bedside. I got to know the parents of other children; we became a source of support for each other. It almost felt like we had become normality and those outside with healthy children were in the minority.

A few weeks later Anna's condition had improved and we were allowed to go home. We had all sorts of people coming round to check up on us. In fact even though they were all there to help us adjust to home life, it all became overwhelming. We had two social workers, one health visitor, the community paediatric nurses, a mother who had a child with cystic fibrosis, the child disability team and a clinical nurse specialist. To be honest, I couldn't tell the difference between them all. Some came to offer advice; some came to help me out with household chores. I received money from a charity to get a dishwasher and some things for the home.

With them all coming in and out of the house every day I found it very difficult to cope with everything. Greg was at work and did not seem to want to learn how to do Anna's treatment. I had to look after my seven year old and my new baby who needed much more attention than a healthy baby. Anna needed three times the number of nappy changes and I had to get used to all the drugs I had to give her and do her physiotherapy. The dose of enzymes must be finely tuned to what food she is eating, otherwise it makes her diarrhoea worse and she loses weight. It takes experience to know how much to give.

Within a few days she was sickening again. I could not cope at all. Back to the local hospital she went, I kept asking them why they couldn't cure her; why not just wash all this badness away from her. We went back home only for her to become ill again. Our GP had to prescribe home nebulisers and stronger antibiotics if things became really bad. She remained extremely poorly.

So there was hospital admission after hospital admission. That was the story of her first year. Her breathing became particularly bad and every time she dipped I wondered if she would ever come round again. I used to prepare myself for the scenario that she might not make it to see the next day. When we had our cystic fibrosis reviews at the specialist centre I would look at the other children and compare them, they seemed much healthier than Anna. That worried me, perhaps she was a severe case and would die much younger than the others.

In that time my one focus was on Anna, my emotions were so tied up with her. As a result of this, Alison became very discontented. She started to say that she wished she had cystic fibrosis so that her mummy would pay attention to her. It broke my heart when I heard that. She became very defiant and that made things very difficult for me. This added stress was compounded by the lack of support I felt I was getting from my

husband. He was working hard to pay the mortgage but did not seem to want to help me out with Anna. At that time I had the nurse specialist from the specialist cystic fibrosis centre, the health visitor and the home care team visiting me and all I really wanted was my husband to help.

Then, as if nothing else could go wrong, when Anna was about ten months old, I woke up to find Greg in a complete sweat. He said it was indigestion but I knew something was wrong. I sent for an ambulance and he was rushed to hospital having a heart attack. I felt sure that someone somewhere had seen that finally something was going well for me and had decided to make everything bad again.

This must have been my lowest point. In fact one day I found myself on the phone to the practice nurse with a pile of multi-coloured pills in front of me asking her what I was doing. I'm sure I was about to take the lot. She came down the road as quick as a flash when she realised what was happening. She called the Crisis Team, the group of doctors and nurses who go into people's homes to assess their mental health. I was deeply depressed. I felt guilty smiling and just wanted to run away from my family. I thought that the way I could be happy was if they could cure cystic fibrosis. My GP put me on antidepressants to try and help me. I also started seeing the community psychiatric nurse.

Despite all this Anna's health was more stable in the latter part of her first year and improved over her second. Her growth and development was very much in the lower range of normal and she had a permanent chesty cough and wheeze, particularly when she caught an infection.

She remained on her routine of diet supplements, four hourly nebulisers, routine antibiotics etc. We both became more used to her physiotherapy. The main issue that

needed to be dealt with at that time was our own adjustment as a family to Anna's illness. I still couldn't fully accept her illness; it was so restricting on my life. Alison also found it difficult. She began to describe Anna and I as magnets, permanently stuck together and too difficult to drag apart. Anna was very attached to me; I had spent so much of her life with her that she felt unsettled when I was not there.

My husband kept his views quite quiet. I guess he does not like to worry me any more than I am. He is much more positive about things. He believes that when she gets older they will have a cure for it. Whereas I can't even plan her next birthday, he thinks that when Anna gets older they will have a cure for it, he can look into her teens. His ill health was also a worry. He has a list of medical conditions as long as your arm since he had that heart attack.

I also have my fair share of medical problems. I am being investigated for endometriosis as I have had pelvic pain and heavy periods for some time now. I am seeing a gynaecologist for that and late last year we asked if he could sterilise me. It was a hard decision to make because I always wanted more children, but the added workload of another baby, with cystic fibrosis or without, would just be too much for me.

We have all had help with the psychological adjustment that we have had to deal with. I have had a community psychiatric nurse visit me for support, Greg and I have embarked on a series of couples counselling sessions and Alison has had her own counselling. We also have additional support in this area from the specialist cystic fibrosis centre and our GP.

Unfortunately we had a reminder of just how ill Anna is last November. I found her lying motionless in her cot. They say it was some kind of febrile convulsion and I

thank the Lord that I got to her in time, I had to resuscitate her whilst the ambulance was on the way. Thankfully it was all okay and we all got through it, but I still prepare myself for the day when it might not be okay.

That said Anna is getting stronger. She has reached the toddler stage, where she hides everything and throws tantrums. I treat her as I would any other child who is naughty. I have had to learn to do that, in the beginning I would make excuses for her because she was sick, but it should not really matter. She is going through the phase where she does not co-operate with her physiotherapy; there are far too many interesting things to do. Anna is a bright sparky child. I value every moment with her. I look back on how close I came to not being here to see this and it makes me sad. I would have missed so much.

I feel much more on top of Anna's treatment now; I even go to surgery and tell the GP what needs to be changed in Anna's treatment if she is poorly. I guess I am still waiting deep down for someone to knock at the door and tell me that everything is okay, that Anna doesn't have cystic fibrosis at all and they were just testing me to see if I would cope.

As for the future, well I don't like to look ahead. If I do, then I guess I think negatively, then I won't be disappointed. If she makes it to her third birthday then at least I will have had three good years with her, if she makes it to five that is even better. All I want is to see my girls grow up to be happy. God willing, I will.

The following chapter will discuss the results of the questionnaires and the interviews in relation to the hypotheses presented at the start of the study.

CHAPTER 12

DISCUSSION

This thesis reports the results from the first national, case controlled study in the UK that explores whether having a child with cystic fibrosis has a greater impact on the lives of parents, compared to those parents with no extra care burden in the family to contend with. Are we as health professionals making assumptions about the lives of parents rather than seeking evidence to support our speculation? This study has shown that there can be no doubt that caring for a child with cystic fibrosis significantly intrudes into every aspect of daily life. However, despite identifying this intrusion parents appear to be coping with the day-to-day management of the disease and family life.

Cystic fibrosis is a life limiting, multi-system disease for which there is no cure. The current median survival in the United Kingdom is thirty years, however children and young adults continue to die despite improved medication and treatment regimens. Treatment is time consuming and life long, involving an ever increasing complexity of home management undertaken by parents and eventually the young adult. Although the original identification of the genetic mutation was thought to offer insight into disease severity and progression, more recent work has led to an understanding of the role that moderator genes play on the cystic fibrosis genetic mutation thus complicating prognosis even further.

Scientific research therefore has little impact on the daily life of families where there is a child with cystic fibrosis. Disease management is planned at a Specialist Cystic Fibrosis Centre, supported by the local hospital and general practitioner and carried out by the child and their parents. Although treatment is monotonous and time consuming there is both an emotional and practical burden associated with the disease that parents carry throughout their child's life.

Society assumes that all parents who have children will care for them; their ability, understanding and willingness are not considered. When this does not happen government agencies intervene and either offer support or remove the child from parental care. If parents give birth to a sick or disabled child this assumption does not change and many parents are forced into a medicalised role – often against their wishes – carrying out unsupervised, complex treatment regimens in the home that may have taken professionals years to become expert in. Parents are obliged to shoulder this burden, as there is usually no alternative. However they report feeling under pressure from both relatives and clinical staff, it is at this point that family dynamics can change and the parental role may become confused and indistinguishable from that of a clinician.

The impact of caring for a child with a life limiting disease cannot be under-estimated. Knowledge of the disease and eventual outcome coupled with the daily demands of treatment intrudes into every aspect of life; professionals caring for patients with cystic fibrosis and their families need to become more

aware of the treatment related burden of care that parents cope with every day of their child's life therefore allowing a more individualised approach to treatment.

This current study has sought, through questionnaire and interview, to discover areas of concern – both psychological and practical - identified by parents of children with cystic fibrosis and the impact these may have on their lives. Research has demonstrated that when the family functions well the child's health will benefit (Patterson et al 1990). Recognition of the problems associated with caring for a sick child and the timely offering of appropriate support may allow alleviation of stressors for the parents leading to an improved management of care.

Individually, stressors such as anxiety or acceptance of the disease may not have a direct impact on daily life, rather, it is more likely to be a collection of many stressors coupled with the complexity of the disease and its management that play a more important role. Parents in this study have highlighted several areas of concern leading to poorer mental health, with both the questionnaires and the interviews yielding similar results. The key areas described by parents include financial and employment problems, perception of disease health status and disease management, knowledge and understanding of the disease, family relationships and support.

12.1 Discussion – Summary of Study Population

One hundred parents – both mothers and fathers, were randomly selected from a national database held by the Cystic Fibrosis Trust, this was to ensure a wide geographical representation in the study. Invitations were sent out inviting recipients to take part in the study, further information about the study could be requested by returning a tear off slip to the researcher in a pre-paid envelope. Of the one hundred invitations sent out, eighty returned the slip requesting further information and of those eighty, sixty-five eventually returned completed questionnaires, giving an overall response rate of 65%. Although demographic data could not be collected on non-responders due to the anonymity promised in the study design, some gave their own reasons for not taking part. Most of these reasons were related to caring for their child with cystic fibrosis with parents commenting on the time it takes to carry out treatment regimens therefore not wanting to use their spare time filling in the questionnaires. A few non-responders admitted to being suspicious of the questionnaires describing them as 'psycho questions'.

Both mothers and fathers responded to the questionnaires and although there was not an equal number in each group, there was a fairly similar distribution of males to females (males - 43%, *table 3*). Inclusion criteria included all parents who cared for their child with cystic fibrosis at home irrespective of the child's age. This perhaps explains the wide age distribution of parents (25 – 62 years, *table 3*), as it is not uncommon for people with more severe disease to

return to the parental home for care. The median life expectancy for cystic fibrosis is 30 years therefore parents caring for their older child will still be relatively young when their child dies as can be seen by the upper age group of parents participating in the study.

Data were collected on the ages of children and size of the families. The mean size of family in the study was 2.4 children with an age range of 1 -27 years (mean age - 10.6 years, *table 4*), again reflecting the fact that older children with advancing disease often move back home. The position of the child with cystic fibrosis in the family was also recorded (*table 27*) with almost half the children with cystic fibrosis being the first-born child (48%) but not the only child, and nine families having more than one child with cystic fibrosis in the family. Treatment for cystic fibrosis has improved over the past fifteen years with the majority of children reaching adulthood. Today young children with cystic fibrosis look no different from their peers and usually remain relatively healthy well into their teen years with little medical intervention. Parents with young families therefore are distanced from the reality of the disease and base their family planning decisions on their personal experience and perception of the disease. This perception of disease status, coupled with increasingly easier access to antenatal testing, usually chorionic villus sampling, may account for parents continuing to have children after diagnosis of the first child.

Employment history (*table 15*) changed following the birth of their child with cystic fibrosis with fewer parents returning to full time employment (68%

down to 48%). Many cases who returned to part time employment were women (11% increasing to 38%), often leaving a professional position (lawyers, accountants, business managers) taking in preference a job with hours and holidays that allowed for treatment regimens to be carried out (attending to school dinners, taking in ironing, directing road crossing and class room helpers).

Society is starting to recognise that the traditional role of the mother as carer is changing as men take on a greater share of both childcare and caring for elderly or sick relatives. However, although this belief is held by men and women alike, it appears that on the whole lip service is paid to the idea. Both men and women in the study acknowledged that the mother shouldered the burden of cystic fibrosis care with 63% of mothers taking sole responsibility and 25% of care being provided on a shared family basis (*table 26*). This finding supports the literature where it has been found that there are significant differences in coping between mothers and fathers (Mastroyannopoulou, Stallard, Lewis and Lenton 1997). This information is reflected by data collected on employment where the return to part time work was mainly by mothers who gave up their profession (*table 16*). There is support for these findings in the literature, for example Stewart et al (1994) found that mothers complained of demands related to their role within the family and activities outside the home, and levers et al (1996) identified major areas of concern from mothers including difficulty in managing treatment regimens and disruption to family relationships.

In 1998, a government study on the provision of clinical services for cystic fibrosis graded the level of service available to patients and their families from Level IV up to Level I. This grading was made on the basis of the availability of services, with Level IV being a small general hospital service and Level I (only two in the country, both in London) providing an optimum service, including heart/lung transplantation. Data on the level of service being provided was collected, however parents were asked to assess the level of service themselves using the governments guidelines (*figure 21*), as satisfaction with clinical care is believed to impact on the individual's ability to cope with managing the disease himself or herself. Although cases had been recruited throughout the UK, 40% believed that they were receiving Level I care with a further 28% grading their centres as Level II (*table 5*). These data show that although centres providing cystic fibrosis care throughout the country may not all have met government criteria, many parents felt that that care they were receiving was equal to the current recommendations.

Given that previous research describes the perception of health status as playing a larger role in coping with illness (Abbott et al 1995), (Luescher et al 1999), than actual clinic status, data were therefore collected on parental perception of their child's health at the time of completing the questionnaires. 62% of cases assessed their children to be well at the time of the study and only 18% being slightly unwell. Although all children were receiving routine medication and undergoing a recommended daily treatment regimen, parents did not identify

these as extra problems rather, the majority felt their child to be well at the time of the study.

Figure 18: Grading of Clinical Services

'Parental quality of life when a child has cystic fibrosis'	
Clinical Service	
Please tick beside the category you think that your hospital falls into. <u>You do not need to put the name of your hospital.</u>	
Level 1	<input type="checkbox"/>
A level 1 hospital is regarded as a national resource centre such as The Royal Brompton Hospital or Great Ormond Street Hospital. It has a large collection of patients and is seen as a centre for national education, training and resources. It is also used by other hospitals in the country to refer patients with difficult problems to.	
Level 2	<input type="checkbox"/>
A level 2 hospital is regarded as a regional/major specialist centre such as Birmingham Children's Hospital. It has over 100 patients and a major specialist interest by doctors and paramedical staff who provide care for patients all over region. Staff at the hospital will act as a training resource and the hospital will be a research centre for the region.	
Level 3	<input type="checkbox"/>
A level 3 hospital is a local specialist centre such as City General, Stoke: It will have over 35-40 patients, including some adults and run special CF clinics. Some of the doctors and paramedical staff will have an interest in CF.	
Level 4	<input type="checkbox"/>
A level 4 hospital will have about 10 patients who are seen either in occasional special clinics or general clinics that provide day to day general medical care.	

12.2 Discussion – Summary of Control Population

The control population were recruited through the cases. After agreeing to take part in the study, cases were asked to discuss the study with two friends who were of a similar marital status, had a similar sized family and lived in the same area. Both sets of names and addresses were sent to the researcher. A similar procedure was then followed for recruitment of controls, one name was randomly selected and an invitation to take part in the study was posted with a reply slip and a pre-paid envelope. On receipt of the reply slip further information was sent and on agreement to take part in the study, the questionnaires were sent out. Of the 65 invitations sent out 59 eventually completed the study, a 91% response rate, this is a very good response rate for a questionnaire study of a control population and perhaps reflects this particular group of parents enthusiasm for supporting their friends. No data could be collected on the controls who did not take part, as the initial agreement stated that they would not be contacted again if they did not return the reply slip.

More women in the control group replied to the invitation to take part in the study, therefore there was not as equal a distribution of males to females (men - 37%, *table 6*) as in the case group. This could perhaps reflect the fact that women are generally more receptive to being questioned about their feelings. There was however a similar distribution of ages (26 – 50 years) with a similar number of children in each family (mean 2.1, *table 7*) and similar age range (mean 9.4 years, range 1-21 years).

The control group however had a significantly different employment status with many remaining in full time employment following the birth of their children and only 6% returning to part time employment (*table 17*), perhaps reflecting the imposed caring role of the case group of mothers.

12.3 Discussion of Methodology

12.3.1 Demographic data

In this national study 124 parents participated (age range: 25 - 62 years, 74 females) with 284 children between them (age range: 1 – 27 years, 74 with cystic fibrosis). A varied employment status was represented with many more in the control population being employed than the case population (cases - full time employment: 48%, part time employment: 26%, controls – full time employment: 83%, part time employment: 10%). Parents of children with cystic fibrosis however demonstrated a significant intrusion of cystic fibrosis into their lives compared to the matched control group. Having a child with this disease appears to impact heavily on parents at both a practical and emotional level. Nevertheless, having described many stressors including financial concerns, worry about other family relationships, their own marital relationship and the long term prognosis for their child, parents continued to manage their child's care and get on with their own daily lives. As one father said 'Cystic fibrosis produces another dimension to your life that helps you appreciate things more'. Parents acknowledge the impact of cystic fibrosis yet appear to assimilate into their lives.

12.3.2 Hypotheses

The hypotheses of this study suggested that parents caring for a child with a life limiting, chronic disease (cystic fibrosis), would experience decreased quality of life and decreased psycho-social function compared to parents without a child with cystic fibrosis. This would be affected by the gender of the carer, employment status, age and position of the child in the family, perception of the child's health status and the identification of the degree of intrusion into parents' lives. The results from both the questionnaires and interviews used in this study support these hypotheses as it can be seen that parents in the study have identified similar areas of concern, these will be discussed further in this chapter.

12.3.3 Matching the two groups

The control and case group were matched for gender, marital status, current employment and number of children in each family. To check for any significant variability of these points independent t-tests were carried out (*table 8*). Both groups appear to be well matched with no significant difference in marital status, gender and number of children in each family. There was however a significant difference in employment status ($p < 0.05$) with the parents of children with cystic fibrosis having a lower rate of employment. It appears that this variable cannot be matched as parents; especially mothers of children with cystic fibrosis change their employment patterns following the birth and diagnosis of their child.

12.3.4 Longitudinal group data

It may be reasonable to assume that parents become more confident in their abilities to provide care over time and parents two years on from completing the original questionnaire support this theory. Through the completion of the same battery of questionnaires a smaller group of parents show that they have further developed these skills.

Twenty parents – both mothers and fathers, were randomly selected from part one of the study, again a wide geographical representation was ensured. Invitations were again sent out inviting recipients to take part in a follow-up of the original study that involved completing the same selection of questionnaires as previously. As before, consent was made through returning a tear off slip to the researcher in a pre-paid envelope. Of the twenty invitations sent out, fifteen returned the slip requesting the pack of questionnaires, giving an overall response rate of 75%.

Both mothers and fathers responded to the questionnaires and although there were not an equal number in each group, it was more evenly distributed than Part One of the study (males - 47%, *table 37*). Data were collected again on the ages of children and size of the families, as two years on there may be more children in the families. The mean size of family in the cases that took part in the longitudinal follow up (*table 38*) had increased from 2.3 – 3.8 children with an age range of 3-29 years, mean age - 11.5 years (previous study: 1 - 27 years, mean age - 10.6 years, *table 4*), again reflecting the fact that older children with

advancing disease often move back home as described in the survey carried out by the Cystic Fibrosis Trust (Cystic Fibrosis Trust 2000).

Employment history (*table 39*) had changed over the two years with an increase in unemployment in the cases from 26% - 34% and in their partners from 13% - 33%. Whether this result is a reflection of today's society or personal choice cannot be deduced from this data. It could also perhaps be that as their children have got older, they have become sicker so that parents have to stay at home more as treatment regimens have become more demanding.

The role of mother as main carer had changed over the two-year period (from 63% - 33%), and although the father was still not being identified as a main carer there was more equity in the family as a whole contributing (from 27% - 46%) to treatment management (*table 40*).

Although larger numbers in this section of the study would make the study more robust, it is accepted that the longitudinal data were collected from a small number. Bearing in mind some of the comments made by parents not participating in the first part of the study – 'too busy looking after their child with CF', it is perhaps worth noting, if not the absolute figures, the trend over the two-year follow up period. Although not reaching significance, parents scored lower in all the questionnaires the second time around suggesting that they were gaining confidence in their daily lives. However, although the results support the hypothesis that parents learn to manage as time progresses, significance was

not reached and larger numbers would be required to confirm this. Unfortunately, many of the parents recruited into the first part of this study, although willing to complete the questionnaire packs once were reluctant to participate and complete the questionnaires a second time.

12.3.5 Interview group data

Nineteen interviews with parents (twelve mothers), took place using a grounded theory approach. Although almost all of the mothers had been in full time employment prior to the diagnosis of cystic fibrosis (eleven out of twelve), only half returned to work and this was part time – one mother described her ‘part time work’ as taking in ironing. All fathers had been and remained in full time employment following diagnosis (*table 45*). These interviews allowed parents to comment in more detail on areas identified as highly significant from the completed questionnaires, these included financial implications, employment, family burden and social dysfunction.

While these four main areas were used to inform the first interview, data collected eventually re-classified the areas of concern as; burden and intrusion, employment and financial implications, family relationships and knowledge of cystic fibrosis. Although, through interviewing the parents areas have been slightly re-defined, the interviews support the main findings from the questionnaire part of the study. Parents appear to be in no doubt that having a child with cystic fibrosis has far reaching implications on both parenting and their personal lives, both practically and psychologically.

12.4 Supporting the Hypotheses

12.4.1 Identifying the intrusion

In identifying the intrusion of cystic fibrosis into parents' lives both the psychological and practical elements of this intrusion must be investigated. The General Health Questionnaire has been designed to identify areas of symptomatology. In this study all but one ('severe depression') of the sub-sections (somatic symptoms: $p < 0.05$, anxiety and insomnia: $p < 0.005$, and social dysfunction: $p < 0.05$), comparison between the case and control groups showed a significant difference. With a total score for cases (mean score: 5.14) above the quoted threshold score of 4/5, parents in this study identified problems in psychopathology that do not appear to exist in the control population (median total score: 4, $p < 0.001$). These results may be understood more clearly in the light of the caring experience reported by parents where they face a future of uncertainty and often feel isolated as much of their time is taken up with carrying out treatment.

In caring for their child with cystic fibrosis, parents completing the General Health Questionnaire, have identified somatic symptoms as well as anxiety, insomnia and social dysfunction as significant problems. Living with the knowledge and anticipatory grief that your child is going to die before you, and that their quality of life depends on rigorous adherence to the recommended treatment regimens, will undoubtedly cause psychological distress for parents. In identifying these particular areas parents are acknowledging that the care burden

places great demands on their psychological well being. These issues appear to be all encompassing and bear little relationship to the gender of the parent, parental perception of their child's disease status and whether they are employed or not.

In fact parents highlighted the association between aspects of the General Health Questionnaire and particular areas of concern. Fathers recognised that an increase in burden of care ($p=0.04$), being unemployed ($p=0.01$), and having more than one child with cystic fibrosis ($p=0.01$) could lead to an increase in severe depression in the majority. They also associated an increase in financial burden with a decrease in somatic symptoms ($p=0.01$) and social dysfunction ($p=0.01$). This could reflect a bias in reporting – perhaps fathers were preoccupied with financial concerns, maybe working longer hours or overtime, such that in answering the questionnaire their financial anxiety predominated. Fathers also described an association between anxiety and insomnia and increasing feelings of hopelessness ($p=0.03$). Mothers however identified different problems, with anxiety and insomnia being associated with an increase in the care burden ($p=0.03$).

Two years on however both mothers and fathers identify fewer areas of psychological concern through completion of the General Health Questionnaire, perhaps reflecting that as they become more competent with managing the disease they gain greater control over their lives. However, mental health problems remain as their mean scores only drop from 6.93 in the first study to

6.2 in the second study, although this change does not reach significance it is evidence of the slight change. This score is still above the threshold level of 4/5.

To recognise the global impact that cystic fibrosis has on parents the Beck's Hopelessness Score was used. As in the other questionnaires, the parents highlighted significantly stronger feelings of hopelessness in their situation than the control group (mean total score – cases: 5, controls: 2, $p < 0.001$). In the comparison data (Mann Whitney U tests) this feeling of overwhelming bleakness for the future did not appear to be affected by gender, employment status or perception of illness. Rather, parents although reporting that they were coping on a day-to-day level did not see a positive outcome in the future.

In the longitudinal section of the study, although not reaching significance parents continue to identify a strong feeling of hopelessness as time progresses with a mean score increasing from 5.4 to 5.87. Although with time parents appear to be gaining competence and control, this conversely, does not appear to decrease feelings of hopelessness in the overall situation in either mothers or fathers. As their child gets older, issues of life expectancy become more pertinent to parents.

Scores from the Impact on Family Scale showed that, compared to the control group, parents in the study were in no doubt that cystic fibrosis made a

significant intrusion into their lives at both a practical and emotional level. All sub-sections (financial: $p < 0.001$, care burden: $p < 0.001$, disruption to planning: $p < 0.001$, family burden: $p < 0.001$ and coping: $p < 0.05$) of this questionnaire showed a significant difference between the case and control group indicating the degree of intrusion cystic fibrosis makes.

Both mothers and fathers identified many associations between aspects of the Impact on Family Scale and other problems. Mothers found that being employed decreased their levels of coping ($p = 0.03$) and that a low level of clinical service increased their burden of care ($p = 0.04$). Fathers as described above, associated many of the aspects of the Impact on Family Scale with the General Health Questionnaire including financial burden and burden of care.

Again, although not reaching significance in comparison to their earlier data, the follow-up group report similar scores, although they are lower (mean scores, care burden: 8.13 versus 7.4, disruption to planning: 13.53 versus 12.33, family burden: 9.53 versus 8.93, coping: 10.47 versus 8.07 and the total score: 34.27 versus 33.33).

All of the parents interviewed recognised the intrusion cystic fibrosis makes into every aspect of their lives. Both mothers and fathers identified that as they got older they perceived their child to be sicker. This may be because as the parents got older their children with cystic fibrosis were also getting older and consequently reaching a deteriorating stage in the disease process. Many talked

about the planning that is involved when organising everything from holidays to school days and evenings or days out, describing always arriving late or having to leave early to ensure treatment demands continue to be met. In interviews parents described the burden of carrying out a daily treatment regimen as clouded by the constant knowledge that their child has a shortened life expectancy. Treatment for cystic fibrosis is mainly home based and although on the whole parents are willing to carry out more complex treatment at home this increasing burden makes the illness more visible. With increasing home management, the differences between the child with cystic fibrosis and healthy peers become more apparent and the responsibility of the parent as doctor or nurse increases.

Parents at interview also identified that having a child with cystic fibrosis led to changes in marital relationships. Mothers felt that they had become stronger and for some, more dominant in their relationship, with husbands stepping back and letting their wives make more of the decisions, especially around treatment. Cultural issues occasionally contradicted these comments with some of the Asian mothers feeling undermined by fathers in their treatment decisions and Mediterranean mothers describing an intrusion from the extended family – a concern that professionals may not always consider when dealing with mothers in the outpatient environment.

Parents portrayed a changing relationship with family and friends following the diagnosis of their child, describing a change in their priorities and perceptions

of life, with a feeling of distance and even isolation from others. Although these emotions are understandable and may even be expected in parents who have a child requiring life long treatment with a limited life span, parents unanimously acknowledge the role a support network plays in their coping mechanism. Support was described as coming from many sources, from friends and relatives to charities and professionals. As well as receiving support, many parents felt that the experience they had gained through having a child with cystic fibrosis would be useful to others and were keen to share their knowledge with parents of newly diagnosed children and the wider forum of the general public.

Knowledge of cystic fibrosis - the disease, current treatment methods and potential therapies – was without a doubt seen as a key factor in the ability of parents to cope with the disease. The media was regarded generally as a negative and often untrustworthy source of information, although parents recognised and even demanded the education of the general public. Parents were also keen to warn about the amount of information parents of a newly diagnosed baby should deal with. Experience had shown them that information should be dealt with slowly and 'on a need to know' basis, too much information meant that 'You constantly worried about things before they happened'. Although parents described information sources as varied as the local library and the internet, most of the information about cystic fibrosis was mainly collected from the Cystic Fibrosis Trust and the professionals caring for their child. Both of these resources were felt to be up to date, correct and therefore trustworthy.

In recognition of the multi-system nature of the disease and the complexity of care new recommendations for the management of cystic fibrosis have been made (CF Trust 2001). These recommendations include a multidisciplinary approach to care. The results from the current study indicate that although parents recognise the many aspects of the intrusion that cystic fibrosis makes in their lives, they also indicate that psychosocial and practical support, plus some knowledge of the disease helps to lessen the burden. The multidisciplinary approach recommended by the Cystic Fibrosis Trust aims to help families in particular with issues of support and information, thus recognising the needs of parents. Gibson (1988) found that parents identified a lack of social support as a source of stress related to coping with cystic fibrosis treatment regimens. In completing the General Health Questionnaire the case group highlighted social dysfunction as a problem, especially when their child was requiring treatment. The first stage therefore, is often offering support to families. Baine et al (1995) found that parents reported involvement in the health services – specifically at diagnosis, times of treatment, provision of information, continuity of care and the access and availability of healthcare professionals decreased the risk of mental health problems experienced. Leonard et al (1993) supported this finding in a study that identified the type and amount of professional support affected the level of parental distress. Parents and children with cystic fibrosis build up a long-term relationship with the cystic fibrosis team, allowing the team to develop an understanding of the practical and emotional support each family needs in order to help them to develop effective coping strategies. The results from this study support the literature suggesting that although parents recognise the intrusion of

cystic fibrosis into their lives, where positive coping styles are adopted, parents feel that they are managing better (Coyne 1997, Ray et al 1993, Mullins et al 1991). The hypothesis for this study stated that mothers and fathers caring for a child with a life limiting disease, chronic disease (cystic fibrosis), will experience decreased quality of life and decreased psycho-social function compared to mothers and fathers caring for well children. The evidence from these results supports this hypothesis by providing a predictive index of causes in both men and women separately and together. Predictive causes of decreased quality of life and decreased psycho-social function include parents who complain of anxiety and insomnia, social dysfunction, financial burden in the family and a disruption to planning in the day to day issues of family life.

12.4.2 Gender of carer

The male caregiver has often been described as a non-traditional role, however as society changes so this belief is changing as fathers, sons and partners are increasingly contributing to healthcare in the community (Mays et al 1999). In this study however, both the qualitative and quantitative data revealed that, despite the changing role of the father in today's society, mothers were unanimously recognised as being the main carer, often giving up a profession to care for the child and, although literature reports that women appear to be carrying out the majority of care giving in the community (Craig, 1992, Ngan et al 1995, Sterritt et al 1998), they are reporting a loss of independence and a fear of what others may think if they refuse (Collins et al 1997, Harrison et al 1997). Mothers highlighted these findings during the interviews where comments

regarding responsibility of care and decision-making were discussed. As the main carer mothers were identified as carrying a constant, heavy psychological and practical burden. Although mothers accepted this role without question, they did describe it as an expectation of the family and society and one they felt that they could not reject. There was some obvious resentment of this amongst the mothers interviewed.

Women have a multifunctional role in society including being a mother, daughter, sister, employer, employee and housekeeper, often all at the same time. Again, through the interviews, mothers in this study have described the impact of cystic fibrosis on this complex role in regard to financial and employment constraints and their varied roles as wife, daughter and mother. In the study, both parents identified mothers as taking most of the responsibility for the treatment regimens and in Impact on Family Questionnaire, although not quite reaching significance, mothers also reported more problems with burden of care ($p < 0.1$), family burden ($p < 0.1$) and coping ($p < 0.1$). The interviews however expose a far greater dilemma with mothers describing a constant sense of accountability and responsibility for both the practical and emotional aspects of looking after their child. This mantle of caring was not taken on without some expression of resentment, mothers felt that their husbands, relatives and the professionals caring for their child expected them to embrace this role, giving little thought to the difference in roles between mother and carer. Although all mothers in the study stated that they wanted to care for their child, there was also a degree of guilt in expressing the feeling of having no choice.

The hypothesis stated that mothers and fathers would view the experience of parenting a child with cystic fibrosis very differently. Mothers described living each day at a time trying not to think or plan ahead. Being more closely involved with the daily management of cystic fibrosis, mothers preferred to carry out the present treatment requirements and not think about the complications of the disease in the future.

Fathers conversely appeared to give less thought to the present needs and were more concerned about long-term issues and their role in supporting their child as a 'sick' adult unable to live alone or be financially self-sufficient. Fathers also commented on the lack of control they feel over their lives expressing a fear of the future and a difficulty in living with the trauma of 'not knowing'. Again this evidence supports the hypothesis that gender plays a role in the way a parent copes with the impact of living with a child who has cystic fibrosis.

12.4.3 Employment status

Caring for a chronically sick child or adult places a certain financial burden on the carer in both the additional cost of non-prescribable requirements (e.g. high calorie diet or adaptation to the home) and through a loss of income due to time given up to caring (home care and time taken to go to hospital appointments etc).

Demographic data showed that a significant number of parents (mostly mothers) did not return to full time employment following the diagnosis of their child, with many saying that they had to leave their previous professions. Employment and the consequential financial implications were recognised as playing a significant part in the burden of caring for a child with cystic fibrosis. Mothers often gave up their careers and either left work completely or returned to part time work so that they could work hours suitable to maintaining the demanding treatment regimens and be available if their child became unwell.

When these results are examined in the light of both the demographic data and the interviews it becomes obvious that financial and employment issues would be highlighted by parents. Caring for a child with cystic fibrosis places great financial and time demands on parents. Routine outpatient visits, inpatient admissions and the cost of treatment not considered medical, such as provision of a high calorie diet, places a heavy financial demand on parents. As the disease progresses and the treatment needs increase employment suffers. Parents understandably both want to and have to spend more time with their sick child, time away from work can often lead to annual leave being used up, pay being reduced or jobs being lost. Several mothers described a culture of 'going without', saying that they tended to buy more expensive, high calorie food for their child with cystic fibrosis rather than spend money on themselves.

At interview fathers gave an account of having to work longer hours to compensate for the loss of their wives earnings. They also raised concerns about

having to work more as they felt that they not only had to support the sick child but that they would continue to have to support their child after it had reached adulthood. In reply to this mothers felt that they had been left to shoulder more of the caring burden and look after the sick child alone as their husbands worked late. These areas of stress related both directly and indirectly to financial worries impacting on the lives of parents. Mothers feel isolated with the care burden and fathers feel under pressure to work more while they are both trying to financially make ends meet with the added demands of a sick child. Fathers also associate many psychological problems – somatic symptoms, social dysfunction and overall poorer mental health when they perceive a higher financial burden. This supports the hypothesis that caring for a child with cystic fibrosis does have an impact on employment.

12.4.4 Age and position of the child with cystic fibrosis

At the beginning of the study, it was believed that the age and position of the child with cystic fibrosis in the family might play a role in the way that parents dealt with the impact of the disease on their lives. However, although having a child with cystic fibrosis in the family changes a family's life, the variables of age and position of the child with cystic fibrosis in the family compared to the questionnaire scores do not appear to cause any extra burden in the experience of parents. Fathers however do associate more than one child in the family with severe depression.

Almost half the children in this study were first born, comparison of the Impact on Family Questionnaire with the position of the child in the family showed that, although not reaching significance ($p < 0.1$), having a first born child with cystic fibrosis increased family burden compared to having the affected child placed elsewhere in the family. This could perhaps be related to problems young couples have with any first-born child, as many families went on to have further children, some with cystic fibrosis. Greater understanding of cystic fibrosis leading to improving treatment regimens have led to a generation of children with cystic fibrosis who are relatively healthy and look no different to their peers. Parents appear to be influenced by their personal experience of children with an improved health status and may perhaps be planning their reproductive decision making on this basis.

A wide age range of children were living at home (1 – 27 years) at the time of the study, supporting parents' suspicions that they would be caring for the 'sick' adult. In a study by the Cystic Fibrosis Trust (2000) exploring the needs of parents of children and adults with cystic fibrosis, it was found that most children continued to live at home well into their twenties, although this could be seen as not untypical of their generation. Most parents described feeling relieved about this decision as they could continue to be involved and oversee their child's treatment and general welfare. The researchers portray an unspoken collusion existing between the parent and child where the parent continues to manage cystic fibrosis (the abnormal part of the child's life), allowing the child to get on with leading a normal life, health permitting. Many of these young adults may

have left home for a while but returned with advancing disease status. Anecdotally, some parents reported that their older children often return home with increasing care demands accompanied by a partner for whom the care burden has become too great. Despite reporting a constant worry, most parents aspire to their children leading independent lives, marrying and having children. Some feel a true sense of relief when this happens and that they are no longer responsible, however most are also pragmatic about the possibility of their child returning home at some point in time.

12.4.5 Perception of disease status

Perception of disease status appears to play a greater role in the way that caregivers report burden than the actual disease status. Furthermore it has been found that the degree of parental burden depended more on the parent's coping skills and the level of family functioning than on the degree of the child's disease severity (Luescher et al 1999). Similarly, in a study investigating emotional state and perceived family functioning, researchers report that the more severe parents believed the condition to be, the higher their levels of anxiety, depression and negative effects on daily life (Labbe, 1996). Children in this current study ranged in age from one year to twenty-seven years, consequently there must have been a wide range of disease severity represented within the sample population. Parental report of their perception of whether their child was well or requiring treatment however indicated that the majority felt that at the time of participating in the study their children were well.

Having cystic fibrosis necessitates a level of daily treatment that includes medication, chest physiotherapy and nutritional management. It is perhaps an acceptance of normality or adaptation to the disease that parents did not describe all children as requiring treatment at the time of the study. Instead 80% of the children with cystic fibrosis were described by mothers or fathers as 'well' or 'slightly unwell', whereas only 20% were described as 'requiring treatment'. This is interesting in that the parents appear to have adapted to a way of life whereby the demanding daily treatments required for all people with cystic fibrosis (chest physiotherapy, pancreatic enzyme replacement therapy, oral, inhaled and intravenous medication and diet) are ignored. Parents only recognise the concept of treatment as that which is over and above this routine, treatment for a chest exacerbation for example. Studies exploring coping in chronic disease report that carers cope, maybe it is because perception of treatment is minimised and that the question asked should relate more to burden rather than coping. Thus, in adapting to cystic fibrosis and normalising the demands of daily treatment, it would be interesting to find a definition of treatment that parents would recognise.

Researchers investigating adherence to treatment in adolescents and young adults found that the perception of disease severity was an important indicator of psychosocial well being (Abbott et al 1995, Leung et al 1997). This was reflected in adherence to treatment regimens where the severity of the illness was not found to influence the rate of adherence. Despite clinical evidence to the contrary, if the adolescent felt well then they did not carry out

their treatment. Perhaps this emotional rationalisation can be likened to parents who perceive their child to be well and are therefore more able to manage the daily practical and emotional upheaval of living with a child with cystic fibrosis.

Although the majority of parents reported their child as not requiring treatment at the time of the study, in completing the Impact on Family Questionnaire parents identify an increase in financial burden ($p < 0.2$) and an increase in total median score from 33.5 – 36, although both scores do not reach significance. Many parents have described treatment regimens as time consuming and at times overwhelming, with increasing severity of disease these treatment regimens are increasingly monopolising their time during both the day and night. It is not surprising therefore that parents have highlighted illness as a problem, as this must be a time when they consider their child to have a deteriorating health status with increasing needs both practical and psychological.

12.5 Identifying the key issues

In this study questionnaire scores between the case and control group were compared using Mann Whitney U Tests and this analysis revealed a highly significant difference in all the variables. To identify which variables more accurately accounted for this difference exploratory analysis was undertaken and interviews with the case group of parents were carried out using a Grounded

Theory approach. This qualitative approach supported and clarified the findings of the more objective measures; this then enabled additional statistical investigation of key issues using exploratory analysis with the data from both the case and control group. This analysis showed an intercorrelation between the variables identified by parents from both the questionnaires and the interviews as having the most impact on their lives (caretaker burden, financial problems, and mental health problems). As discussed previously the literature suggests that parents caring for a child with a chronic disease describe the responsibility as burdensome complicated by the family's circumstances (financial circumstances, other dependants in the household) and the psychological burden of caring for a child with a life limiting disease.

The exploratory analysis of these variables demonstrated that the Beck's Hopelessness Score was highly correlated to the General Health Questionnaire total score ($p < 0.03$) (*table 33*). This questionnaire is a heterogeneous measure and in this study appears to be equally well expressed as a measure of hopelessness therefore, as the Beck's Hopelessness Score provided greater discrimination it was used as an indicator of mental health in further analysis.

Causal relationships between the variables that make parents of children with cystic fibrosis report increasing mental health problems were then identified using the data from both groups. The statistical test used was a regression stepwise analysis with the Beck's Hopelessness Score as the dependent variable. Anxiety and insomnia, social dysfunction, financial burden and a

disruption to planning produced a significant change providing a predictive index of causes.

It appears from this extended analysis that a sense of hopelessness in their roles as parents of children with cystic fibrosis distinguishes the two groups (cases and controls). Further, hopelessness (as measured by the Beck's Hopelessness Score) in this circumstance can be used as a proxy for mental health and can be predicted by social dysfunction, anxiety and insomnia, disruption to planning and financial problems.

Overall, the findings from the current study indicate an acknowledgment of the intrusion cystic fibrosis makes into every aspect of the daily lives of parents and their overwhelming sense of hopelessness with their situation. This feeling can be further aggravated by an increasing burden of care as their child's health deteriorates and treatments demands are intensified. It appears conclusive from these results that maintaining the challenging and demanding treatment regimens causes burden, which in turn leads to mental health deterioration as measured and indicated by feelings of hopelessness. This conclusion however appears to contradict the parents' perception of their child's disease status. Parents are describing their child as well despite maintaining a heavy treatment regimen and yet simultaneously appear to be identifying treatment burden as the primary cause of their poor mental health. This contradiction highlights the confusion that parents live with and the differences they feel about the 'here and now' and the future. Bluebond-Langner (1996) describes a strategy that she

refers to as Compartmentalisation of Information about Cystic Fibrosis and the Child's Condition. By using this strategy parent's process and sort information in a way that will protect them from the immediacy of cystic fibrosis.

Bluebond-Langner (1996) illustrates this theory as a filing cabinet where parents file the information they have on all aspects of cystic fibrosis, some folders are more accessible than others with the ones more useful to that part of their child's daily life to the front of the filing cabinet and those, perhaps to do with prognosis towards the back. As the child's condition deteriorates, so the files about transplant or prognosis move forward. This study has demonstrated how parents use this strategy of compartmentalisation through their accommodation of treatment into their daily lives while acknowledging the burden of cystic fibrosis as a long-term problem. Despite this, parents are successfully managing their lives, which suggests that they have been able to adapt to the intrusion of cystic fibrosis, both the treatment demands and the psychosocial burden of the disease. It is perhaps their perception of their child's disease severity that contributes to this daily coping.

12.6 Summary

In all categories of the Impact on Family Scale the case group of parents scored significantly higher than the control group. Caring for a child with cystic fibrosis appears to intrude into all aspects of family life including financial and

employment status, the burden of care and the burden to the family, and the disruption that this caring imposes on family life. Treatment regimens are monotonous and time consuming. They intrude into family life by affecting daily functioning and planning, as well as disrupting longer term issues such as house buying, planning holidays and choosing schools. Parental choice can be influenced by the availability of a Specialist Cystic Fibrosis Centre, the helpfulness of a family doctor, the willingness of schools to allow treatment to be carried out during school hours and the support of family and friends. Parents in the current study have identified that having a child with cystic fibrosis in the family dominates all aspects of daily life and their planning for the future.

Although not statistically significant, gender also appears to play a part in the identification of this intrusion with mothers scoring higher than fathers in all categories of the Impact on Family Scale. Large sample sizes would have been necessary for this finding to achieve statistical significance. This is not an unexpected differential response given that mothers have been identified as carrying out the major burden of care for children with cystic fibrosis. There was also a trend towards higher scores from both parents when the child with cystic fibrosis was the first-born (median score: 36). Perhaps, as new parents, these scores reflect their inexperience of children that is further exacerbated by the diagnosis of cystic fibrosis. Mastery of the disease management programme can be identified from the follow-up data that shows, although not significant, a lower score in all categories from both mothers and fathers except the longer term outlook on life reflected in the Beck's Hopelessness Score.

By having a child with cystic fibrosis parents in the current study have identified an overwhelming feeling of hopelessness, irrespective of gender, employment status, age of child, perception of disease severity and burden of care. It is obvious from this study that the majority of parents foresee their future and the future of their child as bleak, with very little to look forward to or plan for. Data from the follow-up study further reflects this feeling as, although not reaching significance, scores the second time around are much higher. It appears that parents in this current study cope with the day-to-day management of the demands of treatment, adapting their lives so that cystic fibrosis becomes incorporated into every aspect of it. However, this adjustment goes only so far as parents report management and control but continue to acknowledge an uncertain future that increases as caretaker demands grow.

It emerges from the results of the current study that the hypotheses postulated at the beginning have been supported to some extent by the parents' own observations through both formal measurements and more definitely through interview. There is no doubt that having a child with cystic fibrosis has a profound influence on many aspects of parents lives including employment, gender, position of their child in the family, family relationships and their perception of their child's disease status. Although identifying the intrusion that the disease makes on their lives, parents also report that with knowledge and understanding of the disease and its management and a support network that includes family, friends and professionals, they can achieve daily functioning

competently and only succumb when certain stressors become acute i.e. hospital admissions.

Parents admit that there is a certain reality of life with cystic fibrosis that involves hope for a cure in the future as well as fears of losing their child who has become more precious as time has gone on. However, parents have described having to build a protective boundary around them to survive. Although they admit to physical and emotional exhaustion, they also depict a prevailing optimism keeps them going day to day although this does not extend to the future. This ever present feeling of hopelessness about the future can be used as a proxy for mental health and, as social dysfunction, problems with anxiety and sleeping, a disruption to home life and financial problems increase, so mental health appears to deteriorate.

The final chapter will discuss the implications of the findings of this study; the limitations of the study design and make some recommendations for the future.

CHAPTER 13

CONCLUSIONS

13.1 Conclusions

Through having a child with cystic fibrosis, the burden of care placed on parents appears from this study to influence the structure of family life. Parents have identified a significant burden in caring for their child with cystic fibrosis, both practically and psychologically, influencing all aspects of family life. It would appear from these results therefore that parents of children with cystic fibrosis are at increased risk for stress compared with families without a child with cystic fibrosis. However, this current study has conformed to recent thinking in the area of chronic illness within families that emphasises the need to consider factors affecting stress rather than simply assessing psychopathology that is assumed to occur as a direct result of illness.

From an examination of these data, one could ask why it is that parents do not appear to suffer from severe psychopathology? Eiser (1994) suggests that all parents caring for a sick child are not depressed all of the time. Instead parents

display features of ongoing stress rather than a common psychopathology. Cadman et al (1991) also cautions against the clinical tendency to interpret stress as dysfunctional and proposes a different approach from pathological to an assessment of psychological well being. This further leads to the question do parents employ specific protective factors to help them resist stress or their symptoms? Kazak et al (1988) suggests that despite seemingly overwhelming odds, some parents display competent behaviour when coping with a stressful life and, as indicated in some of these results (comparison of questionnaire scores with variables), may not indicate stress-provoked responses to chronic illness. Gibson (1995) identified four areas of control that have helped parents: (i) a discovery of reality, (ii) critical reflection, (iii) taking charge and (iv) holding on. Cowen et al (1985) agrees with this theory by suggesting that parents who have confronted the diagnosis are able to then go on to minimise the normal stresses of the development period. In a study by Foster et al (1998), mothers did not rate their well being as any different to the normal population and felt that they did not experience any more stress except in times of acute illness. A further study by Thompson et al (1992) confirms this with a study recognising that maternal adjustment was associated with lower levels of perceived daily stress, less use of coping mechanisms and a higher level of family support.

How do professionals therefore identify indicators of stress? Is there anything within families that makes them more at risk for burden and are professionals in a position to be able to lighten the burden? In the range of variables investigated in this study (gender, age and position of child, perception

of illness and employment status) none appear to directly affect burden, although there is no doubt that it is perceived. However, analysis shows that men and women identify various predictors of decreased quality of life and psychological impact including; anxiety and sleeplessness, financial burden, social dysfunction and a perceived disruption to family life. The understanding of stressors therefore needs to be individualised and assessed for each parent, especially when introducing new treatment regimens and explaining new concepts about the disease.

We are reminded by Eddy, Carter, Kronenberger, Conradson, Eid and Bourland, et al (1998) that key transition points throughout the child's illness will impact on coping, however, as professionals we must be conscious of the difference in influence that these transition points have on the carer and child. Professionals must be aware that a seemingly routine procedure, for example first hospitalisation, may have a devastating impact on a parent or patient, leading to a potential inability to cope with the situation, either in the short or long term. As professional we can however be reassured by Burton, (1975) who found that despite many negative and conflicting emotions, most parents of children with cystic fibrosis managed to transcend their distress and cope with life in a positive way.

The results of the current study provide useful information for healthcare professionals on the psychological and practical intrusion of long term chronic disease for parents caring for a child with cystic fibrosis. The highly significant

differences between the case and control groups indicates the degree of burden that parents identify in their lives. The overall comparison of the questionnaire scores with the variables however show that there are a few areas of stress that can be highlighted, instead parents appear to feel generally overwhelmed by the demands of caring. The interviews support the findings resulting from the questionnaires, highlighting both the emotional and physical demands cystic fibrosis places on parents however, through the interview process, parents were more able to identify clearly areas that appear to be common areas of stress such as employment, acute phases of illness, financial worries and the lack of a support network. Bearing in mind the correlation between mental health and caretaker burden, professionals can take practical steps in identifying individual burden and devising strategies that either remove aspects of burden or change the management requirements.

Clinical research in cystic fibrosis is increasing, as gene replacement therapy and chaperone therapy become more of a reality for patients and clinicians. Taking part in clinical trials has become a regular event in the life of most patients with cystic fibrosis. With the development of further treatment modalities we cannot help therefore but place an increased burden of care on patients and their carers. Investigators in clinical trials are starting to appreciate the treatment burden and often include some measure of quality of life for the research case as part of the study. Parents have shown that caring for a child with cystic fibrosis creates a physical and emotional burden, should we also perhaps be assessing the extra burden of treatment for the carers during

research trials? The impact of cystic fibrosis and the burden of care on individual parents therefore needs to be continually re-assessed, especially when discussing disease changes, introducing new treatment options or asking for participation in a clinical trial.

13.2 Limitations of the study

The two major limitations of this study are to do with sample size and choice of data collection. Cystic Fibrosis is however a relatively rare disease providing a comparatively small potential research sample, coupled with the psychological nature of the study (parents not willing to be involved with psychological research) and the enormous daily demands on their time, the small numbers are perhaps not such a surprise. Twenty cases were approached to participate in the longitudinal follow-up and only fifteen cases returned completed questionnaires. Further cases were not approached as those who had completed the second pack, had emphasised the imposition that a repeated approach had made on their lives. It is acknowledged that a larger sample size would have provided the study with a more significant conclusion however; an additional contribution to the parental burden did not seem justified.

This study was designed to identify stressors in chronic disease and the literature suggested that perception of disease status played a major role in this. It was decided therefore that parental perception of their child's disease at the

time of completing the questionnaires would provide more meaningful data. Retrospectively, it would have been interesting to compare perception of disease status with actual clinical scores (lung function, height and weight). Unfortunately this would have entailed application to many Local Research Ethics Committees around the UK, this would inevitably have meant limiting or changing the sample group.

Many of the studies reported from the literature used a large selection of validated measures (up to six or seven), thereby increasing the variety of data collected. Action research explores real lives and the factors that intrude into those lives; recruitment problems for this study were therefore anticipated. This study may have been enhanced with the use of other measures, however, as the study was designed to examine burden it was decided to reduce the time imposed on parents keeping participation to a minimum. This study design was chosen to be hypotheses driven rather than exploratory and the choice of questionnaires was based on a more specific approach. It is however accepted that burden is a difficult concept to define and measure – with no definitive, validated measure to use, potentially therefore the choice of measures could be very subjective.

13.3 Future Plans

There is no doubt that assessing the additional burden of caring for a child with cystic fibrosis should become part of clinical routine. How could the multidisciplinary team carry out this assessment? Support, both psychological and practical, helps carers to cope with ongoing anxieties and treatment issues. Recommendations have been made in a variety of chronic diseases (Madge et al 2000) that multidisciplinary teams are important in providing a holistic service to patients and their families. The provision of professionals other than medical staff, such as the clinical psychologist, social worker or nurse specialist allow long term relationships to be formed between carer and professional thereby improving the appropriateness and quality of support available.

Recognition of the need for support and offering support should primarily come from the Specialist Cystic Fibrosis Center with responsibility for care, however supervised shared care with accredited Cystic Fibrosis Clinics allows families to receive the support they need closer to home. Good two-way communication between carers and professionals is paramount therefore in helping parents to cope (Canam 1986). Communication may play a major role in the perception and understanding of the disease and the required treatment regimens. Assumption that a parent of an older child has a good understanding of the illness and the reasons for the treatments cannot be made. Explanations given at the time of diagnosis or at the introduction of a new therapy may not have been sufficient or may have been forgotten due to anxiety at the time. Communication

therefore must be continuous throughout the child's life to be effective. Ongoing two-way communication will highlight concerns from the carers or professionals that may be having a detrimental effect on coping at home. If recognised and dealt with promptly many problems may be alleviated early before they become a major issue.

Recent guidelines (Cystic Fibrosis Trust 2001) recommend that clinical annual reviews, including a psychosocial review, be carried out routinely in Specialist Cystic Fibrosis Centres. As clinicians how can we prevent and ameliorate burden for those more susceptible parents? Is there a place for pro-forma assessments, or individual semi-structured interviews to be done at this time? Although relatively easy for a multidisciplinary team to amalgamate this into a routine review there could be a potential problem with follow-up, as intervention based on assessment may be difficult to measure over a month or so. This problem however is not insurmountable as options such as qualitative interviews/questionnaires could be carried out either postal or face-to-face at a reasonable time - unrelated to clinical follow-up - therefore allowing an individually recommended intervention. Interventions could be usefully studied in the future to assess the most constructive method of helping parents of children with cystic fibrosis.

The conclusions reached from this study indicate that compared to the control population, parents of children with cystic fibrosis live with both a physical and emotional burden that intrude into every aspect of their lives. However, more

remarkable is the evidence to suggest that despite this responsibility parents are managing their family and personal lives successfully. During interviews many parents expressed a feeling that, through having a child with cystic fibrosis they have become better and stronger people. As shown by other investigators discussed here, it is perhaps this seeming acceptance of the disease and the associated problems that allows them to approach their lives in a more positive manner. Cystic fibrosis is a long term, chronic disease and as such could be used as a model for other chronic diseases. By acknowledging that parents recognise the impact that chronic disease has on their lives yet with self knowledge and a wide support network they are able to live with this burden we can, as professionals, offer more appropriate and timely advice and provision of support that will help parents manage this unwelcome intrusion into their lives.

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Abstracts and Presentations from Thesis

Presentations

- Invited speaker: Parents identifying the intrusion of cystic fibrosis
North American Cystic Fibrosis Conference, New Orleans, USA. 2002
- Invited speaker: Parental quality of life when a child has cystic fibrosis
The Child in Focus – Investing in the Future, Royal College of Nursing, Manchester, UK. 2001
- Invited speaker: Identifying the caregiver burden
13th International Cystic Fibrosis Congress, Stockholm, Sweden. 2000
- Invited speaker: Coping with cystic fibrosis, are we expecting too much of the families?
North American Cystic Fibrosis Conference, Seattle, USA. 1999
- Invited speaker: Cystic fibrosis – a family affair
Cystic Fibrosis Trust Annual Lecture, London, UK. 1998

Abstracts and posters

- Identifying the caregiver burden.
Abstract and poster at the 13th International Cystic Fibrosis Congress, Stockholm, Sweden. 2001
- Parental quality of life when their child has cystic fibrosis.
In Paediatric Pulmonology suppl 17.
Abstract and poster at the 12th North American Cystic Fibrosis Conference, Montreal, Canada. 1998

Appendices

Appendix 1 - Letter from Hospital Research Ethics Committee

Institute of Child Health
and Great Ormond Street Hospital for Children NHS Trust
UNIVERSITY COLLEGE LONDON MEDICAL SCHOOL

FILE COPY

11 December 1996

Ms SL Madge
Respiratory Unit
GOS Trust

Dear Ms Madge


96AR16 Parental quality of life and personal health when a child has cystic fibrosis.


Notification of ethical approval

The above research has been given ethical approval after review by the Great Ormond Street Hospital for Sick Children NHS Trust / Institute of Child Health Research Ethics Committee subject to the following conditions:

1. Your research must commence within twelve months of the date of this letter and ethical approval is given for a period of 48 months from the commencement of the project. If you wish to start the research more than twelve months from the date of this letter or extend the duration of your approval you should seek Chairman's approval.
2. You must seek Chairman's approval for of proposed amendments to the research for which this approval has been given. Ethical approval is specific to this project and must not be treated as applicable to research of a similar nature, ie. using the same procedure(s) or medicinal product(s). Each research project is reviewed separately and if there are significant changes to the research protocol, for example in response to a grant giving bodies requirements you should seek confirmation of continued ethical approval.
3. It is your responsibility to notify the Committee immediately of any information which would raise questions about the safety and continued conduct of the research.
4. Specific conditions pertaining to the approval of this project are:

Dean: Professor Roland Levinsky *rs.levinsky*
Director of Finance: Mr Mary Berry *ms.ber*
Director of Research Administration: Dr Penny Leach *drp*
Director of Administration: Mr Shane O'Brien *sho@ucl.ac.uk*





The child first and always

30 Guilford St
London WC1
Direct Line: 0171 813
Direct Fax: 0171 813

- The use of the enclosed standard consent forms for the research. A copy of the signed form must be kept by you with the research records as our insurers may demand access to them.

Yours sincerely

Anna Jenkins

Anna Jenkins
Secretary to the Research Ethics Committee

enc

cc Professor D Skuse
 Behavioural Sciences Unit
 ICH

Appendix 2 - Letter from Dr R Stein re: Impact on Family Scale

ALBERT EINSTEIN COLLEGE OF MEDICINE
OF YESHIVA UNIVERSITY

JACK AND PEARL RESNICK CAMPUS • 1300 MORRIS PARK AVENUE • BRONX, NEW YORK 10461

DEPARTMENT OF PEDIATRICS
Division of General Pediatrics
Office of the Vice Chairman
PHONE: (718) 918-5300
FAX: (718) 918-5007

Mailing Address:
Jacobi Medical Center, Room 817
Pelham Parkway So. & Eastchester Road
Bronx, New York 10461

June 16, 1997

Susan Madge, MSc RN RSCN
Clinical Nurse Specialist
for Cystic Fibrosis
Respiratory Unit, Level 6 Cardiac Wing
Great Ormond Street Hospital
for Children NHS Trust
Great Ormond Street
London WC1N 3JH
ENGLAND

Dear Ms. Madge:

Thank you for your request regarding the Impact on Family Scale used by our group in studying chronic illness in children. Because of the rising number of requests for the instrument and the need to supply psychometric data on the instrument as well, we have developed a standardized packet of information regarding the measure.


Unfortunately we do not have support to cover the costs of handling these requests and are therefore forced to charge a processing fee of \$25.00 for the Impact on Family Scale. This covers the cost of preparation, duplication, and mailing the information. The check for this fee must be in United States dollars and payable through a United States bank. It should be made out to "PACTS PAPERS/AECOM" and sent to Dr. Ruth Stein, Room 817 at the above mailing address.

Should you personally decide to use the instrument, we would be willing to have you do so with the following stipulations: we would like to be kept informed of your decision. We would also expect that the sources of the instrument be acknowledged in any products and that copies be sent to us. In an effort to keep track of the use of the instrument, we would expect you to refer any interested colleagues directly to us.

The packet has been prepared based on our experience with a particular population of inner-city children with chronic physical health problems and in some instances on other populations as well. While the instrument has also been used in still other populations, we cannot describe or guarantee their properties in those situations. We are sure you realize this would be a standard caution with the use of any instrument.

Many thanks for your interest.

Sincerely,


Ruth E.K. Stein, M.D.
Professor of Pediatrics

REKS/ltr

Montefiore Medical Center, Weiler and Moses Divisions • Jacobi Medical Center • North Central Bronx Hospital

Appendix 3 - Letter re: use of General Health Questionnaire (28)



NFER-NELSON
INFORMING YOUR DECISIONS

DARVILLE HOUSE
2 OXFORD ROAD EAST
WINDSOR
BERKSHIRE SL4 1DP
TEL: +44 (0)1753 858961
FAX: +44 (0)1753 856830

5th February 1997

Ms Susan Madge
143 Winston Road
Stoke Newington
LONDON N16 9LL

Dear Ms Madge

GENERAL HEALTH QUESTIONNAIRE 28

Thank you for your letter of 29th January. As you are using the GHQ as a single measure, I cannot give you permission to reproduce it rather you should purchase it direct by contacting our Customer Support department on the above number. Record Forms come in packs of 100 at £36.00 plus VAT per pack: please quote your supervisors qualification code when ordering.

I am sure you will be able to access the *User's Guide to the GHQ* through your department or library: if not it is available at £42.00.

Yours sincerely
NFER-NELSON

SUSAN THOMPSON
Rights & Royalties Coordinator

e-mail: susan.thompson@nfer-nelson.co.uk

Appendix 4 - Letter re: use of Beck's Hopelessness Score

The Psychological Corporation Limited

A DIVISION OF HARCOURT BRACE & COMPANY LIMITED



4 February 1996

Ms Susan Madge
143 Winston Road
Stoke Newington
London
N16 9LL

24 - 28 OVAL ROAD
LONDON NW1 7DX
U.K. Registered Office

Telephone: 0171 267 4466
Fax: 0171 482 2293 & 485 4732
Cable: ACADINC LONDON
Registered No. 493356 England

Dear Ms Madge

Beck Hopelessness Inventory

Thank you for your letter of 29 January.

The Beck Hopelessness Scale and Record Forms can be purchased from us and the prices are:



Kit (manual, scoring keys, 25 record forms)	£46
Record Forms (pack of 25)	£25.50

With the permission of your supervisor you can use her registration number when ordering. You will also need some method of payment e.g. trust/university account or credit card etc. You can order directly through our customer services on 0181 300 3322.

Yours sincerely

Paul McKeown
Manager
The Psychological Corporation Europe

Appendix 5 - letter from the Cystic Fibrosis Trust re: reproduction of posters

SGK/publications	
7 August 2001	
TO WHOM IT MAY CONCERN	
<u>Sister Susan Madge, Great Ormond Street Hospital for Children, London</u>	
This is to confirm that permission has been granted to the above-named to reproduce posters published by the Cystic Fibrosis Trust within work submitted in her PhD Thesis.	
The posters were supplied by the Cystic Fibrosis Trust.	
 Sandra Kennedy Publications Manager Cystic Fibrosis Trust	
	11 London Road Bromley Kent BR1 1BY Telephone 020 8464 7211 Facsimile 020 8313 0472 E-mail enquiries@cftrust.org.uk Website www.cftrust.org.uk
<small>Patron: HRH Princess Alexandra, the Hon. Lady Ogilvy GVO President: Sir John Batten KCVO MD FRCP Cystic Fibrosis Trust registered as a charity number 1079049 A company limited by guarantee registered in England and Wales number 3880213 Registered office: 11 London Road, Bromley, Kent BR1 1BY.</small>	

Appendix 6 - General Health Questionnaire (28)

THE GENERAL HEALTH QUESTIONNAIRE

GHQ 28

David Goldberg

Please read this carefully.

We should like to know if you have had any medical complaints and how your health has been in general, *over the past few weeks*. Please answer ALL the questions on the following pages simply by underlining the answer which you think most nearly applies to you. Remember that we want to know about present and recent complaints, not those that you had in the past.

It is important that you try to answer ALL the questions.

Thank you very much for your co-operation.

Have you recently

A1 – been feeling perfectly well and in good health?	Better than usual	Same as usual	Worse than usual	Much worse than usual
A2 – been feeling in need of a good tonic?	Not at all	No more than usual	Rather more than usual	Much more than usual
A3 – been feeling run down and out of sorts?	Not at all	No more than usual	Rather more than usual	Much more than usual
A4 – felt that you are ill?	Not at all	No more than usual	Rather more than usual	Much more than usual
A5 – been getting any pains in your head?	Not at all	No more than usual	Rather more than usual	Much more than usual
A6 – been getting a feeling of tightness or pressure in your head?	Not at all	No more than usual	Rather more than usual	Much more than usual
A7 – been having hot or cold spells?	Not at all	No more than usual	Rather more than usual	Much more than usual
B1 – lost much sleep over worry?	Not at all	No more than usual	Rather more than usual	Much more than usual
B2 – had difficulty in staying asleep once you are off?	Not at all	No more than usual	Rather more than usual	Much more than usual
B3 – felt constantly under strain?	Not at all	No more than usual	Rather more than usual	Much more than usual
B4 – been getting edgy and bad-tempered?	Not at all	No more than usual	Rather more than usual	Much more than usual
B5 – been getting scared or panicky for no good reason?	Not at all	No more than usual	Rather more than usual	Much more than usual
B6 – found everything getting on top of you?	Not at all	No more than usual	Rather more than usual	Much more than usual
B7 – been feeling nervous and strung-up all the time?	Not at all	No more than usual	Rather more than usual	Much more than usual

Appendix 6 - General Health Questionnaire (28) cont


Have you recently				
C1 – been managing to keep yourself busy and occupied?	More so than usual	Same as usual	Rather less than usual	Much less than usual
C2 – been taking longer over the things you do?	Quicker than usual	Same as usual	Longer than usual	Much longer than usual
C3 – felt on the whole you were doing things well?	Better than usual	About the same	Less well than usual	Much less well
C4 – been satisfied with the way you've carried out your task?	More satisfied	About same as usual	Less satisfied than usual	Much less satisfied
C5 – felt that you are playing a useful part in things?	More so than usual	Same as usual	Less useful than usual	Much less useful
C6 – felt capable of making decisions about things?	More so than usual	Same as usual	Less so than usual	Much less capable
C7 – been able to enjoy your normal day-to-day activities?	More so than usual	Same as usual	Less so than usual	Much less than usual
<hr/>				
D1 – been thinking of yourself as a worthless person?	Not at all	No more than usual	Rather more than usual	Much more than usual
D2 – felt that life is entirely hopeless?	Not at all	No more than usual	Rather more than usual	Much more than usual
D3 – felt that life isn't worth living?	Not at all	No more than usual	Rather more than usual	Much more than usual
D4 – thought of the possibility that you might make away with yourself?	Definitely not	I don't think so	Has crossed my mind	Definitely have
D5 – found at times you couldn't do anything because your nerves were too bad?	Not at all	No more than usual	Rather more than usual	Much more than usual
D6 – found yourself wishing you were dead and away from it all?	Not at all	No more than usual	Rather more than usual	Much more than usual
D7 – found that the idea of taking your own life kept coming into your mind?	Definitely not	I don't think so	Has crossed my mind	Definitely has
<hr/>				
A	B	C	D	TOTAL
<input type="text"/>	<input type="text"/>	<input type="text"/>	<input type="text"/>	<input type="text"/>
<hr/>				

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First published 1978


NFER-NELSON
INFORMING YOUR DECISIONS

Code 4075 02 4

Appendix 7 - Impact on Family Scale

IMPACT OF ILLNESS ON FAMILY SCALE

PLEASE READ THESE INSTRUCTIONS CAREFULLY BEFORE STARTING Having children can change peoples lives. Here are some statements that people have made about living with a child. For each statement, please circle whether, at the present time, you would 'strongly agree', 'agree', 'disagree', or 'strongly disagree'.

PLEASE ANSWER BOTH PART I & PART II OF THE STATEMENTS - THIS IS VERY IMPORTANT

PART I					PART II				Total Score
		Strongly Disagree	Disagree	Agree	Strongly Disagree	Fully Disagree	Partly Disagree	Yes, definitely	
a	Children cause financial problems for the family.	1	2	3	4	2	1	0	40-41
b	Additional income is needed in order to cover my child's expenses.	1	2	3	4	2	1	0	42-43
c	Because of my child we are not able to travel much.	1	2	3	4	2	1	0	44-45
d	We have little desire to go out because of my child.	1	2	3	4	2	1	0	46-47
e	It is hard to find a reliable person to take care of my child.	1	2	3	4	2	1	0	48-49
f	Sometimes we have to change plans at the last minute because of the children.	1	2	3	4	2	1	0	50-51
g	We see family and friends less because of my child.	1	2	3	4	2	1	0	52-53
h	Because of the experiences we've been through we are a closer family.	1	2	3	4	2	1	0	54-55
i	My relatives have been understanding and helpful with my child.	1	2	3	4	2	1	0	56-57
j	I think about not having more children.	1	2	3	4	2	1	0	58-59

PART I					PART II				
		strongly agree	agree	disagree	strongly disagree	Fully	Partly	Not at all	
		1	2	3	4	2	1	0	
k	My family and I discuss my child and his/her problems together.	1	2	3	4	2	1	0	60-61
l	I don't have much time left over for other family members after caring for my child.	1	2	3	4	2	1	0	62-63
m	Relatives interfere and think they know what's best for my child.	1	2	3	4	2	1	0	64-65
n	Our family gives up things because of my child.	1	2	3	4	2	1	0	66-67
o	Being tired is often a problem for me.	1	2	3	4	2	1	0	68-69
p	I live from day to day and don't plan for the future.	1	2	3	4	2	1	0	70-71
q	Nobody understands the burden I carry.	1	2	3	4	2	1	0	72-73
r	Travelling for medical care is a strain on me.	1	2	3	4	2	1	0	74-75
s	Learning to care for my child has made me feel better about myself.	1	2	3	4	2	1	0	76-77
t	I worry about what will happen to my child in the future (when I'm not around and he/she grows up).	1	2	3	4	2	1	0	78-79
u	Sometimes I feel like we live on a roller coaster, in crisis when my child has a problem, OK when things are stable.	1	2	3	4	2	1	0	80-81
STOP ANSWERING									
v	Did you or anyone else in your household stop working because of your child?	YES (2) NO (1)				2	1	0	82-83
w	Do you or anyone in your household now have a full or part time job?	YES (2) NO (1)				2	1	0	84-85

PART I						PART II			Total Score
		strongly agree	agree	disagree	strongly disagree	Is this related to your child's health?			
		1	2	3	4	Fully	Partly	Not at all	
If YES to 'w', would you 'strongly agree', 'agree', 'disagree', 'strongly disagree', with these statements:									
x	Time is lost from work because of appointments for health care.	1	2	3	4	2	1	0	16-17
y	We cut down the hours we work to care for my child.	1	2	3	4	3	1	0	18-19
CONTINUE ONLY IF YOUR CHILD HAS CYSTIC FIBROSIS. OTHERWISE STOP HERE.									
aa	People in the area treat us specially because of my child.	1	2	3	4	2	1	0	20-21
bb	Sometimes I wonder whether this child should be treated 'specially' or the same as any other child.	1	2	3	4	2	1	0	22-23
cc	We try to treat this child the same as any other child.	1	2	3	4	2	1	0	24-25
dd	It is hard to give much attention to the other children because of the needs of this child.	1	2	3	4	2	1	0	26-27
ee	I worry about my other children's health.	1	2	3	4	2	1	0	28-29
PLEASE ANSWER THESE ONLY IF THE OTHERS ARE OVER FOUR YEARS OLD									
ff	There is fighting between the children and my child with CF.	1	2	3	4	2	1	0	30-31
gg	My other children are frightened by my child with CF.	1	2	3	4	2	1	0	32-33
hh	My other children seem to have more illnesses, aches and pains than other children their age.	1	2	3	4	2	1	0	34-35
ii	The school grades of my other children suffer because of my child with CF.	1	2	3	4	2	1	0	36-37

© Ruth EK Stein MD, and Catherine K Riessman PhD.

Appendix 8 - Beck's Hopelessness score



Date: _____

Name: _____ Marital Status: _____ Age: _____ Sex: _____

Occupation: _____ Education: _____

This questionnaire consists of 20 statements. Please read the statements carefully one by one. If the statement describes your attitude for the **past week including today**, darken the circle with a 'T' indicating **TRUE** in the column next to the statement. If the statement does not describe your attitude, darken the circle with an 'F' indicating **FALSE** in the column next to this statement. **Please be sure to read each statement carefully.**

- | | |
|--|---|
| 1. I look forward to the future with hope and enthusiasm. | <input type="radio"/> T <input type="radio"/> F |
| 2. I might as well give up because there is nothing I can do about making things better for myself. | <input type="radio"/> T <input type="radio"/> F |
| 3. When things are going badly, I am helped by knowing that they cannot stay that way forever. | <input type="radio"/> T <input type="radio"/> F |
| 4. I can't imagine what my life would be like in ten years. | <input type="radio"/> T <input type="radio"/> F |
| 5. I have enough time to accomplish the things I want to do. | <input type="radio"/> T <input type="radio"/> F |
| 6. In the future, I expect to succeed in what concerns me most. | <input type="radio"/> T <input type="radio"/> F |
| 7. My future seems dark to me. | <input type="radio"/> T <input type="radio"/> F |
| 8. I happen to be particularly lucky, and I expect to get more of the good things in life than the average person. | <input type="radio"/> T <input type="radio"/> F |
| 9. I just can't get the breaks, and there's no reason I will in the future. | <input type="radio"/> T <input type="radio"/> F |
| 10. My past experiences have prepared me well for the future. | <input type="radio"/> T <input type="radio"/> F |
| 11. All I can see ahead of me is unpleasantness rather than pleasantness. | <input type="radio"/> T <input type="radio"/> F |
| 12. I don't expect to get what I really want. | <input type="radio"/> T <input type="radio"/> F |
| 13. When I look ahead to the future, I expect that I will be happier than I am now. | <input type="radio"/> T <input type="radio"/> F |
| 14. Things just don't work out the way I want them to. | <input type="radio"/> T <input type="radio"/> F |
| 15. I have great faith in the future. | <input type="radio"/> T <input type="radio"/> F |
| 16. I never get what I want, so it's foolish to want anything. | <input type="radio"/> T <input type="radio"/> F |
| 17. It's very unlikely that I will get any real satisfaction in the future. | <input type="radio"/> T <input type="radio"/> F |
| 18. The future seems vague and uncertain to me. | <input type="radio"/> T <input type="radio"/> F |
| 19. I can look forward to more good times than bad times. | <input type="radio"/> T <input type="radio"/> F |
| 20. There's no use in really trying to get anything I want because I probably won't get it. | <input type="radio"/> T <input type="radio"/> F |

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15 19 20 A B C D E

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