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Parent, Family, and Disease Variables
as Predictors of
Psychological Functioning in Children with Cystic Fibrosis

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A thesis presented to the Faculty of Graduate Studies
in partial fulfillment of the requirements for the
degree of Master of Arts.

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Abstract

The purpose of the present study was twofold: to determine the relative importance of disease, family, and parental variables in the psychological functioning of children with cystic fibrosis; and, to study which parental, family, and disease variables are associated with the parents' ability to cope with the disease and provide appropriate care to the child. The sample consisted of 36 children with cystic fibrosis, 24 mothers and 21 fathers of these children.

The children's behavioral problems and self-esteem based upon physical appearance were found to be associated with maternal trait anxiety, overprotection, and with the impact of disease on the family. Social competency as measured by quantity and quality of social activities and athletic competence were however associated with the disease status, particularly the nutritional status of the child.

The impact of the disease on the family appeared to be the most important factor in the parent's coping with the disease. The disease status and general family functioning were not significant as was expected.

Differences were also found between maternal and paternal overprotection. Maternal overprotection was noted to be associated with the disease status whereas paternal overprotection was related to family functioning and depression.

Subsequent research should include sociodemographic variables such as sex, age of the child, family income, family constellation, parental education as factors which might be associated with the child's psychological functioning and with parent's coping.

The results of the present study indicate that behaviorally oriented therapies for the child must also take into consideration parental factors such as anxiety and overprotection. Whereas, the disease status must be considered when providing treatments geared towards increasing social competence.

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Introduction

It had been over two nerve racking hours - anxiously pacing the floor and wiping the sweat from his palms. What must have been an eternity, the nurse walked into the waiting room:

"Mr. Smith ... congratulations

Is it a boy? A girl? Is the child healthy? The questions sped through the mind of the new father.

.... Mr. Smith, you're the father of a healthy 6 pound baby girl!"

Although times have changed and today the scenario may be different as more and more fathers participate and are present in the delivery of the child, the parent's questions remain the same: what is the sex of the child and is the child healthy? Undoubtedly all will be either male or female, but not all will be born healthy. Others will be born apparently healthy, only to be later, either in infancy or early childhood, diagnosed to have some type of chronic illness. In fact, the prevalence of chronic illness has been estimated at approximately 10 percent of children ages birth to 18 years (Pless & Roghmann, 1971; Gortmaker & Sappenfield, 1984).

Long-term childhood disorders can be categorized as: 1. diseases due to chromosomal aberrations; 2. diseases as a result of abnormal hereditary traits; 3. diseases due to harmful intrauterine factors; 4. disorders resulting from perinatal traumatic and infectious events including permanent damage to the central nervous system and motor apparatus; and, 5. diseases due to serious postnatal and childhood infections, injuries,

neoplasms, and other factors (Mattson, 1972). Some of the more frequent disorders include asthma (38/1000), mental retardation (25/1000), visual impairments (30/1000), hearing impairments (16/1000), to some less frequent but more severe disorders such as leukemia (.1/1000), seizure disorders (3.5/1000), congenital heart disease (7/1000), and cystic fibrosis (.5/1000) (Gortmaker & Sappenfield, 1984).

Not only does chronic illness tax the body of its resources, there is also a psychological price. A chronic disease, whether or not life threatening, places the child and family under considerable stress. Some of the more severe diseases (ie. leukemia, cystic fibrosis) cause the parent and child to live "in the shadow of death". In all cases, parents and children must take time from their daily activities for therapeutic management, and, in some cases, time away from friends and family due to hospitalization and disease exacerbation. Some children and their families cope adequately with the stress of the disease and its impact on the individual's psychological functioning. For those who cannot cope, the daily trauma of living with a disease results in psychological maladjustment. Yet, which factors contribute to the psychological maladjustment of the child and family remains unknown. The purpose of the study was twofold: to determine what factors affect the child with cystic fibrosis psychological functioning; and secondly, to determine how the parents cope with the disease and provide the child with care and love.

Psychological Functioning of Children with a Chronic Disease

The way in which a chronic illness affects the child and/or family has been described using two paradigms. The first, which is slowly becoming less popular, argues that psychological responses are dependent upon the specific disease; that is, a child with diabetes reacts differently from one with leukemia (Mattson, 1972; Wolfish & McLean, 1974; Koocher & Sallan, 1978; Garner & Thompson, 1978; Magrab & Papadopoulou, 1978). The second paradigm used to assess the impact of chronic illness on psychological functioning maintains that the impact of the illness goes beyond the boundaries of specific diseases; in other words, psychological problems developed by the child and/or family are similar despite various diagnoses (Pless & Pinkerton, 1975; Stein & Jessop, 1982; Pless, 1983, 1984). Whereas, the first approach was based more upon clinical impression, the second paradigm, referred to as a non-categorical approach, has found empirical support with children (Tavormina, Kastner, Slater, & Watt, 1976) and adults (Cassileth, Lusk, Strouse, Miller, Brown, Cross, & Tenaglia, 1984). However, the non-categorical approach has its limitations in that, it does not deal with children who have behavioral or emotional problems directly related to their disease. For instance, children with epilepsy often have attention deficient disorders and hyperactivity which are due to the central nervous system anomaly or to medication side effects and not to any psychological factor. Nevertheless, despite which of two paradigms used, the

results from studies on the actual impact of chronic illness on the child and family have been confusing.

The psychological sequelae of a chronic disease, regardless of the type of diagnosis, have been reported to be maladaptive for both the child and family (Douglas, 1964; Rutter, Tizard & Whitmore, 1970; Pless & Rughmann, 1971; Pless, Rughmann, & Haggerty, 1972; Steinhauer, Mushin, & Rae-Grant, 1974; McKey, 1976; Steinhausen & Schindler, 1981; Dorenbaum, Cappelli, Keene, McGrath, 1985). Children with a chronic disease were often described as anxious, depressed, socially maladjusted, and having low self-esteem (Swift, Seidman, & Stein, 1967; O'Malley, Koocher, Foster & Slavin, 1979; 1980) and families as being burdened financially, had strains in their relations, and were socially restricted (Turk, 1964; Mattson & Gross, 1966; Stein & Riessman, 1980).

However, others have argued that children with a chronic disease are not psychologically maladjusted. Rather, they maintain that the psychological strengths of the child and family outweigh any deficiencies (Tavormina et al., 1976; Drotar, Doershuk, Stern, Boat, Boyer, & Matthews, 1981; Sabbeth & Leventhal, 1984). Tavormina et al. (1976) reported:

The 'typical' child [with a chronic illness] in the study performed much like a 'normal' child and the scoring distribution also followed normative patterns with respect to variance. Even though there were some clear-cut exceptions, the normalcy rather than the deviance of this sample is demonstrated across measures. In short, the children's

functional strengths noticeably outweighed their weaknesses (p. 108).

Why have these studies produced such conflicting conclusions?

Some have argued that the conflicting results may be partially due to methodological faults such as lack of control groups and objective measures, and use of small sample sizes (Gayton & Friedman, 1973; Tavormina et al., 1976). In addition, many studies used inappropriate outcome measures since either established tools to measure proper outcome did not exist or were not used (Pless, 1983; Lamping, 1985). For instance, only recently has an objective measurement tool of the impact of chronic disease on family been established (Stein & Riessman, 1980). Previous research had resorted to simple interviews or open-ended questions.

Despite these criticisms, three large epidemiological studies have provided convincing evidence that children with a chronic illness have a 1.5 to 2.6 times greater risk of some degree of maladjustment as measured by objective questionnaires of behavioral problems and personality assessment (Douglas, 1964; Rutter et al., 1970; Pless & Roghmann, 1971). These findings perhaps indicated that a chronic illness is much like any other life stressor which only in interaction with other factors may increase the risk of maladjustment but is not by itself the cause (Pless, Roghmann, Haggerty, 1972; Pless & Pinkerton, 1975; Bendell, Giordani, Amour, Tavormina, & Boll, 1977; Drotar, 1981; Lewis & Khaw 1982; Orr, Weller, Satterwhite, Pless, 1984). Two factors which have been frequently associated with the

child's psychological functioning are parental and family factors, and, disease related factors.

Family and Parental Factors

Allen & Pearson (1928) in a psychological assessment of twelve children with a chronic disease reported a "tremendous drive on the part of the child to hold infantile, dependent relationships with the parents, especially the mother" (p. 232) which was seen as a combination of the child's attempts to gain attention from the mother and the mother's attitude of overprotection. This similar theme has been echoed by a number of present day studies. Mattson (1972), based in part, from some of his earlier work (Mattson & Gross, 1966a, 1966b; Mattson, Gross, & Hall 1971) classified children with a chronic illness who were maladjusted into three groups: the first group was described as fearful, inactive, lacked outside interests, and had marked dependency on the family; the second group engaged in prohibited and risk taking activities, denied realistic dangers, and were rebellious; and, the third group was described as shy, lonely, harboring resentment and having hostile attitudes towards healthy individuals. However, the common feature in all three groups was the mothers' overprotection. If the parents, and particularly, the mothers were overprotective, the child would resent the pressure and become more anxious which would further reduce chances of successful independence and parent's adaptation to the disease (Weitzman, 1984; Sabbeth, 1984). However, it must be noted that overprotection was only an impression and not until recently has a validated questionnaire to measure overprotection

been designed (Parker, Tupling, & Brown, 1979; Parker, 1983).

Maladjustment has also been associated with family functioning. Pless, Roghmann, & Haggerty (1972) in a comparison of 356 children with a chronic illness and 110 healthy children found a positive relationship between the child's adjustment as measured by behavioral problems, self-esteem, overall adjustment, and family functioning. Pless et al. (1972) reported the lower the marital satisfaction, family happiness, communication, leisure time, and the greater the frequency of disagreements, the greater the child's likelihood of maladjustment.

Similarly, Lewis & Khaw (1982) found children with more behavioral problems came from families with extremely poor family functioning. The members belonging to these malfunctioning families had poor emotional bonds with each other and could not change in times of stress.

Family factors do appear to play a significant role in the adjustment of the child with a chronic illness. However, this notion has not been firmly validated. For instance, the overprotective issue has not been objectively assessed; Pless et al. (1972) relied on a questionnaire of family functioning developed during the study, which although it was a reasonably good scale, did not take into consideration other factors such as the impact of the disease on family functioning; and, Lewis & Khaw (1982) used the Family Adaptability and Cohesiveness Evaluation Scale which had not been extensively validated and never used with an illness sample.

Disease Related Variables

Although several disease related variables such as age of onset, duration of illness, nature of therapy, visibility of the disease have been addressed to a limited degree, the most frequent disease factor found to be associated with maladjustment has been the severity of the disease (O'Malley et al., 1980; Pless, 1983, 1984). Two somewhat opposing hypotheses have been generated regarding severity and maladjustment: 1. the greater the severity, the greater the maladjustment; and, 2. a marginality hypothesis that states children with moderate severity will be more maladjusted than those with mild or severe disease. Several studies have reported supporting findings on each of the hypotheses.

Pless & Roghmann (1971) based on a survey of 209 chronically ill children, found the greater the severity of the disease as measured by its debilitating effect, the greater the degree of maladjustment. This finding has been replicated by several studies (Mattson, 1966, 1972; Orr et al., 1984; Stein & Jessop, 1984).

However, Bruhn, Hampton, & Chandler (1971) and McAnarney, Pless, Satterwhite, & Friedman (1974) found that moderate or marginal severity has a more detrimental effect on adjustment than either mild or extreme severity. Both studies argue that children in the moderate group have additional cultural and familial pressures. Children with mild severity can basically function as well as healthy children given proper medical

compliance. On the opposite extreme, those with extreme disease have significant limited function and are not expected by either the family or society to function as healthy individuals. However, the marginal group possesses severity that is neither totally incapacitating nor completely absent. Whereas in both the mild and extreme severity groups, the children know what is expected from them, the marginal group must rely on external sources to determine their behaviors (Bruhn et al., 1971). This creates considerable internal conflict resulting in a higher degree of maladjustment.

The association between disease related factors, particularly severity of the disease, and the child's psychological functioning remains ambiguous. Many of the problems of these studies reflect earlier mentioned criticisms such as inappropriate measures of outcome and subjective measures of severity. For example, McAnarney et al., (1974) based many of their conclusions on projective tests and clinical interviews, and Bruhn et al., (1971) employed subjective criteria in development of the measure of severity. These criticisms have been dealt with, to a limited degree, in studies of cystic fibrosis. Likewise, the impact of family and child with cystic fibrosis and the association between family factors, disease factors, and maladjustment have been examined in several studies. However, no attempts, in studies examining specifically cystic fibrosis or any other disease, have been made to systematically study how these factors together affect the child's psychological functioning.

Cystic Fibrosis: The Disease

Cystic fibrosis is the most common, lethal, genetic, disease and the most frequent cause of chronic obstructive pulmonary disease and pancreatic insufficiency in the first three decades of life (Sant'Agnese & Davis, 1979; Matthews & Drotar, 1985). The complexities of the disease and its manifestations defy simple description.

The distinct clinical pathological features were first described by Dorothy Andersen (1938) and at that time, the disease was solely associated with fibrosis of the pancreas. However, Schwachman, Dooley, Guilmette, Patterson, Weil, & Leubner (1956) later noted that 15 percent of children with cystic fibrosis did not have pancreatic problems. At present, it is known that the disease results in multi-organ abnormalities including pancreatic, respiratory, gastrointestinal, hepatic, and reproductive tract anomalous functioning which are manifested in various degrees for different patients and ages (Matthews & Drotar, 1984). Complications of the disease are known to include pneumothorax, hemoptysis, diabetes mellitus, and intussusception (Taussig, Kattwinkel, Friedewald, & di Sant'Agnese, 1973). Anderson & Goodchild (1976) note that the cardinal clinical features of the disease include a progressive obstructive suppurative chest disorder in addition to malabsorption which is predominantly due to pancreatic digestive enzyme insufficiency.

The diagnosis of cystic fibrosis can be made at any stage of development and is based on the following criteria: presence of an elevated sweat chloride greater than 50 to 60 mol/liter (3 to 4 times greater than the normal rate), and either chronic pulmonary disease or pancreatic insufficiency (di Sant'Agnese & Davis, 1979).

The pathogenesis of cystic fibrosis has been described by numerous authors (ie. di Sant'Agnese, 1959; Lloyd-Still, 1983), however, Anderson & Goodchild (1976) provide a "lay-man's" description of the general pattern of the disease. A dysfunction of the exocrine mucous producing glands causes the mucous to become abnormally sticky and serous secretions to become abnormally concentrated which cause ducts and ductules of various organs to become blocked. The sticky concentrated mucous secretion affects the intestine and lungs causing meconium ileus or meconium ileus equivalent and chronic respiratory disease. The concentrated serous secretion affects particularly two organs: 1. the pancreas resulting in malabsorption; 2. and the liver causing liver failure. In addition, the elevated sweat chloride levels which result from altered electrolyte transport, can cause heat prostration.

Genetics

Cystic fibrosis is a genetically transmitted disease and is almost exclusive to the Caucasian race (Steinberg & Brown, 1960). The incidence of cystic fibrosis is approximately 1 per 1800 in live white births; 1 in 17,000 black births; 1 per 90,000

Oriental births; and rare for American Indians, African blacks, Japanese, Malaysians, and Asian Indian populations (Crozier, 1974; Wright & Morton, 1968; Matthews & Drotar, 1984). The disease is transmitted via the autosomal recessive mode of inheritance (Danks, Allan, & Anderson 1965) and as such, both parents must carry the abnormal gene in order for the child to inherit the disease. When both parents are carriers of the abnormal gene, the recessive quality of the gene produces a 25 percent risk of acquiring cystic fibrosis, 50 percent risk of being a carrier, and 25 percent free of risk in new borns (Anderson & Goodchild, 1976). Approximately 5 percent of the Caucasian population carry the abnormal gene, but neither a chromosomal marker for the gene has been indentified (Goodchild, Edwards, Glenn, Grindley, Harris, Mackintosh, & Wentzel, 1976) nor has a reliable method of distinguishing a cystic fibrosis carrier in utero been established (Anderson & Goodchild, 1976).

Clinical Features of the Cystic Fibrosis Patient

In infancy, respiratory symptoms including increased respiratory rate and lack of movement of the upper chest, failure to thrive although having a voracious appetite, and abnormally offensive, bulky, soft and greasy stools are often noted (Anderson & Goodchild, 1976). Childhood is marked by slow weight gain, recurrent or persistent bronchitis, wheezy breathing, harsh cough, rectal prolapse, and continuing abnormal stools (Anderson & Goodchild, 1976; Matthews & Drotar, 1984). In adolescence and early adulthood, sterility in males, delayed puberty, signs of malnutrition, cyanosis, finger clubbing, dyspnea, chronic

sinusitis, and recurrent abdominal pain are not uncommon (Anderson & Goodchild, 1976).

In addition, clinical differences between sexes both in features observed and prognosis have been noted. The Cystic Fibrosis Foundation (1978) reported that the median survival age for males in 1976 was 21 years but for females it was only 17 years. Further, girls have much worse pulmonary functioning and significantly more are below the 50th percentile in weight compared to males. Although there is, as of yet, no scientific explanation for the differences, Gurwitz et al. (1979) suggests that the differences may be due to either the onset of puberty rendering females more susceptible to lung disease, or a number of social cultural factors - girls tend to eat less in hopes of a better appearance and tend to participate in less activities. However, these remain unsubstantiated.

Prognosis and Management of Cystic Fibrosis

There is no cure for cystic fibrosis - prognosis is poor. Gurwitz et al. (1979) have noted several factors which differentiate the severity of prognosis. Good prognosis depends upon being a male, maintaining appropriate weight, single system involvement at presentation of the disease, and normal chest x-ray film within the first year of presentation. Poor prognosis is related to being a female, poor weight gain or loss of weight, multiple system involvement (pulmonary and gastrointestinal), and abnormal chest x-ray films in the first year of presentation (Gurwitz et al., 1976).

The wide variation of the disease manifestations between individuals requires highly comprehensive and individualized treatments involving a number of physicians and ancillary caregivers (Matthews & Drotar, 1984). Patients must attend regular clinic visits in order for physicians to closely monitor their disease process and initiate appropriate interventions. Management of cystic fibrosis is time consuming for both parents and patient who must follow a daily regimen of various interventions to control the disease. Therapeutic management is directed at respiratory disease, nutritional problems, and complicating features (ie. diabetes).

In order to remove the mucus caused by bronchial secretion and to control lung infections, management of the respiratory problems includes administration of various oral antibiotics; chest physiotherapy with coughing and breathing exercises and postural drainage; and intermittent inhalations of moisture, antibiotics, and mucolytic agents (Anderson & Goodchild, 1976). Malnutrition is caused by the malabsorption of fats, proteins, and carbohydrates resulting from pancreatic insufficiency. In addition, respiratory infections reduces the child's appetite and further aggravate nutritional problems. Management of the nutritional problems include administration of pancreatic enzyme replacements which increase caloric utilization; dietary management; and vitamin supplements (Anderson & Goodchild, 1976). A chronic enteral feeding program which provides continuous high energy nutrition to severely malnourished patients has been also

noted to improve nutrition (Boland, Patrick, Stoski, MacDonald, & Beaudry 1984).

Finally, management of complicating factors such as meconium ileus, intestinal obstruction, rectal prolapse, liver disease, diabetes mellitus, and cor pulmonale are treated according to established medical protocols. No cure exists, but as Anderson & Goodchild (1976) note "optimal treatment, although it still has its disappointments, can be extremely rewarding; suboptimal treatment may do little more than prolong the patient's life to some extent, as a respiratory cripple" (p. 2).

However, prognosis is not as bleak as it once was. With the improvements in basic research on the pathogenesis of the disease, better nutrition, earlier diagnosis, improved antimicrobial therapy, more aggressive utilization of more efficient pharmacologic agents and technical therapeutic equipment, the result has been an increase in longevity for the patient with cystic fibrosis (Gurwitz, Corey, Francis, Crozier, & Levison, 1979; Matthews & Drotar, 1984). In fact, the median survival age of cystic fibrosis patients has risen from 4 years in 1950 to 19 year by 1976 to a reported 23 years in 1984 (Cystic Fibrosis Foundation, 1978; Cotter, 1984). In addition to research on the physical aspects of the disease, a number of investigators have sought to identify psychological and psychosocial problems of these children and their families.

Psychosocial Aspects of Cystic Fibrosis: The Family

The first study to appear in the literature on the impact of cystic fibrosis on family functioning was conducted by Turk (1964). Twenty eight parents from 25 families of children with cystic fibrosis were interviewed using both open and closed ended questions relating to social and emotional deprivation and problems in communication between family members. Turk found that most parents felt deprived of time and energy to engage in leisure-time and adult activities, to be alone with the other spouse or themselves. Parents also reported being deprived of vacations and family activities with other friends and relatives. In terms of communication problems, parents were unable to discuss their child's disease with other children or relatives and neighbours. Almost 60 percent of parents refused to discuss cystic fibrosis with the afflicted child. The problems in communication resulted in what Turk referred to as the "web of silence", not to discuss the demands and issues that relate to the illness and holding these frustrations within.

Lawler, Nakielny, & Wright (1966) followed with a clinical study of families with children with cystic fibrosis. The authors interviewed 11 parents about their marital relationship, attitude towards the illness, the influence of the illness on the family situation, and their overall adjustment. Lawler et al. (1966) found that 8 of the 11 mothers were clinically depressed and reported "living in the shadow of death" (p. 1044). Fathers showed evidence of psychopathology with 1 being grossly

psychotic, one paranoid, another paranoid and alcoholic, three others had peptic ulcers, and another had a gastrectomy. Marital relationships were severely strained with 6 couples considering separation and 3 couples simply living in the same house without sexual contact. The authors concluded that need for psychological help was immediately required.

Tropauer, Franz, & Dilgard (1970) interviewed 23 mothers examining their attitudes, feelings, and reactions to the adolescent with cystic fibrosis. The authors also noted that the mothers were often preoccupied with death and the disability of the adolescents. Three of the mothers were clinically depressed and another 8 had frequent periods of dejection and discouragement. All the mothers were noted to be guilt ridden and either overprotective or rejecting behavior were common methods used to cope with guilt, anxiety, and frustration. Tropauer et al. (1970) noted that the few families that appeared well adjusted had more communication and openness within the family. However, the authors concluded that the majority of families with cystic fibrosis were in need of psychological interventions.

One of the widely recognized studies on family adaptation to cystic fibrosis was conducted by McCollum & Gibson (1970). The paper which was published in the Journal of Pediatrics received an editorial comment suggesting to all physicians to carefully read the paper and incorporate the findings. McCollum & Gibson (1970) obtained information from 56 families with children with

cystic fibrosis from a multiple choice questionnaire, interviews, and group discussions. The authors noted that the majority of family reactions tended to focus around three stages of the disease: prediagnostic stage, confrontational stage, and long-term to termination stage. Feelings of self-doubt and self-reproach were noted from the mothers during the prediagnostic stage from their incapacity to successfully nurture the child. These feelings often led to despair and hostility towards the child. In the confrontational stage, which marked the confirmation of the diagnosis, feelings of disbelief, denial, anxiety, and helplessness were reported. Finally, in the long-term to termination stage, intermittent mourning and denial of prognosis were often noted. In addition, parents would often report profound guilt when disciplining their child.

These findings received support from a number of other studies that also noted the high levels of stress, anxiety, depression, overprotection, and other psychological problems in families with children with cystic fibrosis (Sibinga, Friedman, & Huang, 1973; Belmonte & St. Germain, 1973; Kulczycki, Lucas, Regal, & Tantisunthron, 1973; Goldberg, Kutscher, Lorin, & Denning, 1973; Leiken & Hassakis, 1973; Allen, Townley, & Phelan, 1974). However, Gayton & Friedman (1973) in a review of the literature on psychosocial aspects of cystic fibrosis criticized many of these studies and questioned the validity of their findings because they used small sample sizes, no controls, and relied on subjective interviewing rather than reliable assessments. For instance, McKey (1973) based on the observations

of the cystic fibrosis clinic at the University at Maimi School of Medicine, reported that over 75 percent of this population had family breakdowns. However, Begleiter, Burry, & Harris (1976) in a better controlled study of parents of 34 children with cystic fibrosis found only 5 divorces in 29 families giving a prevalence rate of divorce of 17.2 percent. This rate was not significantly different from the population divorce rate of the United States.

Other studies have appeared in the literature which attempted to correct some of the noted methodological faults. Gayton, Friedman, Tavormina, & Tucker (1977) administered two objective measures, the Family-Concept Q Sort (Q sort) and the Minnesota Multiphasic Personality Inventory (MMPI), to 29 fathers and 43 mothers from 43 families having children aged 5 to 18 years with cystic fibrosis. The authors first compared the mothers' and fathers' scores and then compared these scores with pre-existing normative data. On the MMPI, mothers scored significantly higher on the depression and social scales and lower on the mania scores compared to fathers. In comparison to a group of normal parents, fathers with children with cystic fibrosis scored higher on the lie, hypochondriasis, hysteria, and psychopathic deviate scale and mothers scored higher on the depression and masculinity-femininity scales of the MMPI. If the cysticfibrosis child was a male, 32 percent of fathers and 22 percent of mothers were found to have one or more scale scores in the pathological range. On the Q Sort, no differences between mothers and fathers were found on the perceived family adjustment or family satisfaction. However, when the parents were asked to

respond on the Q sort items as if the child did not have cystic fibrosis, they reported increased family satisfaction and adjustment. The authors concluded that use of objective measures provide further evidence that cystic fibrosis does have a negative impact on the family.

Similarly, Stienhausen & Schindler (1981) interviewed 36 mothers of children with cystic fibrosis and a matched control group on family functioning and mental health, and found parents of the cystic fibrosis group had higher amounts of disagreement, fewer mutual talks, and were more overprotective. Mothers displayed neurotic and depression symptoms and fathers had an increased likelihood of physical disease.

Phillips, Bohannon, Gayton & Friedman (1985) using a semistructured interview, asked 43 mothers and 29 fathers what they found to be major problems in their families. The impact of hospitalization was reported to be the most prevalent major problem. However, 28 percent of mothers compared to only 3 percent of fathers reported that communication within the family was a major problem.

Although it would appear that families with cystic fibrosis are at much greater risk of having family problems, two recent studies have provided evidence demonstrating the parents and family cope with stress of the disease and that appropriate family functioning is maintained.

Lewis & Khaw (1982) studied a group of chronically ill children of whom 31 were cystic fibrosis patients and their

mothers and matched the chronically ill to a control healthy group on age, gender, and race. As a measure of family functioning and coping, mothers completed the Family Adaptability and Cohesiveness Evaluation Scales. The authors analysed the results using a MANOVA and found no statistical differences between the two groups on measures of adaptability and cohesion.

Cowen, Mok, Corey, MacMillan, Simmons & Levison (1986) administered the Family Adaptability and Cohesiveness Evaluation Scale, the Family Assessment Measure to 294 mothers and fathers and also obtained measures of the disease status and the child psychological functioning. Cowen et al (1986) found the parents scores to be well with the norms. Small but significant correlations ($-.13$ & $-.17$) were reported between parents scores on family assessment and pulmonary function (FEV 1) but no correlation was found between family assessment and the child's weight for height. Significant correlations between family functioning and family problems were found only for mothers. Cowen et al. (1986) concluded that there is no evidence to suggest psychopathology in families with children with cystic fibrosis.

Whether the presence of cystic fibrosis produces family dysfunction remains unanswered. Studies conducted in the 1960's and early 1970's revealed that parents and families had a number of problems. More recent research has provided data not supporting increased pathology. These differences might well be due to the increased psychosocial care that families have been

receiving within the CF community and from CF care-givers.

Psychosocial Adjustment of Children with Cystic Fibrosis

The study conducted by Lawler et al. (1966) was the first to appear on the psychological implications on children having cystic fibrosis. Along with studying the effects of cystic fibrosis on the family, the authors measured intelligence using the Stanford Binet and Weschler Intelligence Scale for children, achievement with the Schonell Reading test, and personality using the Rorschach, Draw-A-Person, and Bender Gestalt tests on 11 children with cystic fibrosis. The children were all noted to have normal or above normal intelligence although underachieving at school. In terms of the personality measures, the authors found the children's defence mechanisms to include denial, excessive use of fantasy, hostility toward self and others, regression, repressed aggression and depression.

Lawler et al. (1966) findings were replicated by Spock & Stedman (1966) who had administered the Peabody Picture Vocabulary Test, Goodenough Draw-A-Person Test, and the Bender Visual Motor Test to 34 children aged 3 to 16 years. In addition, Spock & Stedman (1966) also noted the presence of a high level of anxiety, a need for strength, and feelings of inadequacy.

Kulczycki, Robinson, & Berg (1969) later replicated the findings of normal intelligence but underachievement for 26 children with cystic fibrosis aged 3 to 10 years.

Cytryn, Moore, & Robinson (1973) conducted an in depth analysis of 29 children with cystic fibrosis aged 8 months to 9 years on appearance, self-view, defense and coping styles, and emotional health using the Rorschach, human figure drawings, and clinical assessment. Cytryn et al. (1973) noted from the human figure drawings and the responses on the Rorschach that girls had an adequate view of themselves; however, boys were not as aggressive and self-assured as boys of their own age and most viewed themselves as younger, dependent, and inadequate. The major defense and coping styles observed included reaction formation, fantasy, regression, withdrawal, and avoidance. Emotional health was judged from ratings by a psychiatrist and a psychologist who reviewed each case and rated the child as having mild, moderate, severe, or no emotional disturbance. The authors reported that although 41 percent of their sample did not have any disturbances, 17 percent had mild disturbance, 28 percent moderate, and 14 percent severe disturbance. Moderate or severe disturbances in the 42 percent of cystic fibrosis children was reflected in the child's limited drive, perseverance, creativity, passive-dependency, high stress, withdrawal, to the more severe social adjustment problems including inappropriate and regressive behavior, severe disorganized anxiety, hostility, and lack of cooperation.

Similar results, but with an older sample of cystic fibrosis patients, were found by Boyle, di Sant'Agnese, Sack, Millican, & Kulczycki (1976). The authors obtained measures, on a group of adolescents and young adults aged 13 to 30 years, from the

Weschler Adult Intelligence Scale and Wisconsin Intelligence Scale for children, Thematic Apperception Test, the Rorschach and the Draw-A-Person Test. Boyle et al. (1976) noted much anxiety and depression amongst their sample and reported no relationship between intelligence and the patient's ability to cope emotionally. Boyle et al. (1976) suggested that the stress causing emotional difficulties resulted from: altered physical appearance; strained interpersonal relationships; conflicts in upbringing; and the increased negative awareness of the future. The authors also noted distorted human picture drawing reflecting lack of close interpersonal relationship. The authors concluded that 60 percent of their sample had significant adjustment problems.

Drotar (1978) compared a group of 32 cystic fibrosis patients referred for psychological assessment to a group of unreferred cystic fibrosis patients matched for age, sex, and social class. Based on clinical assessment of these children, Drotar (1978) found five major psychological areas of problems: learning problems; behavioral problems; depression; anxiety; and somatic symptom reporting problems. The younger children had more learning disturbances and behavioral problems. Adolescents also had behavioral problems, but in addition were depressed, had anxiety reaction and anticipatory grief, and, somatic symptom reporting problems. Drotar suggested that maladjustment may result from the family's inadequate handling of the disease and life stress.

Steinhausen & Schindler (1981) compared 36 cystic fibrosis patients aged 5 to 18 years to a matched healthy control group on age, sex, and socioeconomic status. The authors independently interviewed mothers to determine whether the child had psychopathology using the Graham & Rutter interview schedule. Interrater reliability between the two authors was .81. Steinhausen & Schindler found that 42 percent of children with cystic fibrosis had no or slight psychiatric disorders compared to 70 percent of the control group. Further, only 11 percent of the control group had marked to definite psychiatric disorders compared to 36 percent of children with cystic fibrosis. Thirty three percent of these cystic fibrosis children would have been diagnosed as having an emotional disorder specific to childhood and adolescence.

On a more positive note, Tavormina, Kastner, Slater, & Watt (1976) compared 144 children, ages 5 to 19 years, with a chronic illness of which 30 had cystic fibrosis to the norms on the Piers-Harris Self-Concept Scale, Nowicki-Strickland Locus of Control, Junior Eysenck Personality Inventory, Missouri Children's Picture Series, and the Psychological Screening Inventory. The author found that many of scores on the scales were well within the normal range of established norms. On the Missouri Picture Series, children with cystic fibrosis were found to appear more dependent, less mature, voiced more problems with intellectual and school status and physical appearance. Tavormina et al. (1976) argued that although children with a chronic disease do have some problems, they are not severely

psychologically maladjusted. Similar findings have been reported by Gayton et al. (1977), Kellerman, Zeltzer, Ellenberg, Dash, & Rigler (1980), Drotar, Doershuk, Stern, Boat, Boyer, & Matthews (1981), Cowen, Corey, Simmona, Keenan, Robertson, & Levison (1984), and, Simmons, Corey, Cowen, Keenan, Robertson, & Levison (1985).

Gayton et al. (1977) compared 33 children with cystic fibrosis with 31 of their healthy siblings on the Piers-Harris Self-Concept Scale, Missouri Children's Picture Series, the Holtzman Inkblot test for the 5 to 13 year olds, and, the Tennessee Self-Concept Scale, Rotter's Locus of Control Scale, the Holtzman Inkblot test, and the MMPI for the 14 to 18 year olds. No significant differences were found on any of the measures between the cystic fibrosis and sibling group.

Kellerman et al. (1980) compared 349 healthy adolescents to 168 chronically ill children of which 23 had cystic fibrosis on measures of anxiety, esteem, and perception of control. Again, no differences were found between the healthy and cystic fibrosis group on any of the measures.

Drotar et al. (1981) compared four groups of children: 91 children with cystic fibrosis; 47 children with various chronic illnesses; 71 healthy siblings of the cystic fibrosis patients; and, 61 healthy children. Mothers of these children were administered the Louisville Behavior Checklist and their teachers completed the School Behavior Checklist. All mean scores were

within the normal range of established norms. When the cystic fibrosis and chronic illness group were pooled together and compared to the two control groups, differences in aggression, infantile aggression, hyperactivity, inhibition, social withdrawal, sensitivity, fear, normal irritability, and total disability were found. However, Drotar et al. (1981) suggested that these differences were not due to maladjustment but rather, to the excellent adaptation of the healthy group. They concluded that the average school age child with cystic fibrosis had age adequate adjustment to home and to school and that cystic fibrosis does not necessarily pose a major disruption to the child's life adjustment.

Cowen et al. (1984) administered the Cornell Medical Index, and the Tennessee Self-Concept Scale to 226 cystic fibrosis patients aged 16 years and older. Differences were noted between younger (16-19 year olds) and older (20 years and over) patients. Younger females and males had lower self-criticism and higher positive scores and younger males also had lower conflict scores. Overall gender differences were also noted with 50 percent of women aged 20 years and over and 30 percent aged 16 to 19 years reporting higher emotional disturbances compared to 22 percent of older males and 12 percent of the males aged 16 to 19 years. However, all scores were within the normal range according to established norms.

Simmons et al. (1985) administered the Tennessee Self-Concept Scale and Children Health Locus of Control to 62 children with cystic fibrosis aged 12 to 15 years and the Children

Behavior Checklist to their mothers. Normative comparisons revealed differences only on the Children's Behavior Checklist. Boys did not function socially as well as girls and displayed higher scores on all the behavioral scores. However, only 6.5 percent of the total group scored significantly above the clinical cut-off. This compared to the 6.8 percent of the normal population scoring above the cutoff. The authors also noted that scores on the self-concept scale and health locus of control were well within the general population norms.

In keeping with the other studies, the inconsistent and conflicting findings on the psychological adjustment of children with cystic fibrosis are plentiful. However, several studies had suggested that family functioning and disease factors are mediating variables in the child's adjustment (ie. Boyle et al. 1976; Drotar, 1978).

Association of Family Functioning and Children's Maladjustment

The association between family functioning and the child's adjustment has been for the most part inferred rather than systematically assessed. For instance, Spock & Stedman (1966), Tropauer et al., (1970), and Leiken & Hassakis (1973) noted maternal overprotection leading to increased maladjusted in the child. However, overprotection was measured by clinical impression.

Family functioning as a mediator of maladjustment was noted by Lewis & Khaw (1982). Lewis & Khaw conducted two multiple

regressions using deviation adaptability and cohesion scores of the Family Adaptability and Cohesiveness Scales as predictor variables and the child's self-concept in the first regression and number of behavioral problems in the second regression as outcome variables. The authors found that deviations from normal adaptability and cohesion could significantly predict the child's psychological adjustment. Lewis & Khaw (1982) concluded that family functioning does appear to be a mediating variable in the child's adjustment to cystic fibrosis. However, the authors failed to report the squared multiple correlation and also neglected to consider the interaction of other significant variables, particularly disease factors.

The Effects of Disease Variables on Psychological Functioning Disease Variables

The physical manifestations of cystic fibrosis can be broken down into how they effect the performance of various organs and the child's overall well-being. Several researchers have developed clinical scores which assess the patient's current physical status and predict to a lesser degree, the prognosis (Schwachman & Kulczycki, 1958; Taussig, Kattwinkel, Friedewald, di Sant'Agnese, 1973). The National Institute of Health (NIH) clinical score developed by Taussig et al. (1973) is based upon objective measures of pulmonary functioning and weight, and clinical impression of activity, and attitude. Children with cystic fibrosis who have poor pulmonary functioning, are below normal weight, tire easily and participate in few activities, and are non-compliant would have the worst prognosis and clinical

scores. In addition, these factors are weighted so that pulmonary functioning is the most important predictor of the physical status and attitude which is the least significant.

Effects of Disease Variables on Family Functioning

A number of studies, many of which have already been mentioned (ie. McCollum & Gibson, 1970; Cytryn et al., 1973; Boyle et al., 1976; Drotar et al., 1981; Cowen et al., 1984; Simmons et al., 1985) have in addition, examined how the overall clinical scores and/or their components, such as pulmonary functioning, affect psychological functioning in the family and the child.

McCollum & Gibson (1970) noted from the responses of 56 parents that the severity of the disease and negative changes in the disease process would often trigger a mourning episode. Parents also expressed anxiety when discussing the child's poor body development and prognosis.

Leiken & Hassakis (1973) presented a case study of four families with children with cystic fibrosis. Again, they noted in one family panic with every change in the disease status or whenever the child had an infection.

Bywater (1981) interviewed 23 mothers of 27 patients (12 to 16 years) and obtained a clinical score of the disease using the Crispin-Norman Scoring (1974) scoring system (The Crispin-Norman scoring system is based upon the degree of damage to the lungs caused by cystic fibrosis using chest x-rays). Bywater found 12

of the mothers were depressed but noted that 8 of these 12 had children who were seriously ill.

Finally, McCubbin, McCubbin, Patterson, Cauble, Wilson, & Warwick (1983) administered the Coping Health Inventory For Parents (CHIP) to 100 mothers and fathers of children with cystic fibrosis. Factor analysis of the scores revealed three independent coping patterns: 1. maintaining family integration, cooperation, and an optimistic definition of the situation; 2. maintaining social support, self-esteem, and psychological stability; and, 3. understanding the medical situation through communication with others parents and consultation with the medical staff. In addition, the authors collected two indices of physical health three months before the parents completed the CHIP and immediately after completion. The first index was the the percentage of predicted weight and height. The second index, which reflected overall pulmonary functioning, was the average of the percentage of predicted capacity of the child's small airway, mixed airway, vital capacity, the peak flow. The three coping patterns were then compared to the change from the first and second health indices. McCubbin et al. (1983) found that mother's efforts to maintain family integration, cooperation and an optimistic definition of the situation were correlated with positive gains in the child's weight/height index ($r=.23$, $p<.05$). Both mothers' and fathers' efforts to maintain social support, self-esteem, and psychological stability were correlated with pulmonary function (mother $r=.23$ $p=.05$; father, $r=.31$ $p<.05$). The weight/height index was found to correlate only with the

father's score on this factor ($r=.22$ $p .05$).

Disease variables appear to be related to several measures of parent's psychological well-being. The worse the prognosis the higher the parent's anxiety and depression. Changes in the disease process also affect anxiety but seem to play a more vital role in the parent's self-esteem, psychological stability, family integration, social support, and an optimistic view of the present situation. Likewise, several studies have demonstrated to some degree that the child's psychological adjustment may be influenced by the disease.

Relationship Between the Child's Psychological Adjustment and Disease Variables

Cytryn et al. (1973), Boyle et al. (1976), Drotar (1978), and Bywater (1981) found that the severity of the illness as determined by clinical ratings was not related to day-to-day functioning, but directly affected emotional disturbance. Cytryn et al. (1973) found 42 percent of their sample to have significant emotional disturbances despite well daily functioning.

Boyle et al. (1976) noted that despite the children's poor clinical rating, they were able to fulfill a number of required daily functions. However, there appeared to be a direct relationship between anxiety and depression and the patient's altered physical appearance and poor future prognosis.

Drotar (1978) found that a group of 32 psychology referred

patients had significantly lower clinical scores. Depression was found to be the most prevalent emotional problem among children with severe disease. This finding was later replicated by Bywater (1981) on a sample of 27 patients.

Bywater (1981) found that his study sample which were comprised of teenagers, were not socially isolated and had a number of close friends. Despite this, 63 percent were found to be depressed which was directly related to clinical scores. Those with a more severe degree of disease were more depressed, 75 percent, compared to those who were more healthy, 44 percent.

In contrast, Zeltzer, Kellerman, Ellenberg, Dash & Rigler (1980) and Drotar et al. (1981) found a relationship between the the severity of the disease and interference with daily functioning. Zeltzer et al. (1980) found on a group of 23 children with cystic fibrosis, those with the most severe disease also reported the greatest disruption of social functioning. However, the authors did not mention how severity was rated.

Drotar et al. (1981) in a more controlled study, in addition to parents' and teachers' ratings of the children's psychological adjustment, obtained clinical ratings of each child's disease. The clinical rating did not correlate with any of the adjustment scales but a relationship was found between greater physical impairment (low clinical rating) and greater social withdrawal ($r = -.29$; $p < .01$).

The child's level of anxiety has often been found to be effected by the severity of the disease (Drotar, 1978) or by

prognosis (Kellerman et al., 1980). Drotar (1978) noted that children with more severe disease often suffered from anxiety reactions and anticipatory grief. Kellerman et al. (1980) found adolescents with cystic fibrosis were more anxious than other groups of chronic illness with stable prognosis.

On the other hand, Simmons et al. (1985) correlated scores on psychological measures from 62 patients to disease variables which included measures of pulmonary function (forced vital capacity (FVC), first second forced expiratory volume (FVC1), forced expiratory flow in the middle of the forced vital capacity, partial oxygen tension) and, a measure of weight and height (percentage of ideal weight for height). Simmons et al. (1985) found significant correlations only with males and only on positive physical self concept. This correlated with the pulmonary function ($r=.55$ FVC, and $r=.57$ FEV1, $p=.01$) and ideal weight for height ($r=.48$, $p=.01$). The authors concluded that there appears to be only a very slight relationship between measures of psychological well-being and physical status. This conclusion was supported by one of the same authors in a previous study (Cowen et al. 1984) that found no correlations between psychological and clinical measures on a group of cystic fibrosis patients aged 16 and older.

Again contradictory and inconclusive results have been found. Some studies demonstrated a significant association between disease related variables whereas others found no or only a negligible relationship. It is not certain why the results

have been inconsistent but it may be due to examining the relationship of disease and psychological functioning as a linear association rather than a curvilinear one as described by the marginality hypothesis (ie. Bruhn et al., 1973). However, an alternative explanation may be that the disease variables do not play an important role in the child's psychological functioning and that the significant correlations obtained in the various studies were due to chance findings.

Rationale for the Present Study

For the child with a chronic disease, growing up in a world that is designed for healthy people can be emotionally stressful. These children are often stigmatized by their peers and society and given the demands placed by the disease such as daily injections, physiotherapy, special diets, use of wheelchairs, hearing aids, and special feeding devices, the child may find it difficult to lead a "normal" life.

Not surprisingly, study of the psychological sequelae of chronic illness has received considerable attention in both the medical and behavioral literature. The findings of most of these studies have demonstrated that children with a chronic disease are at a higher risk of psychological maladjustment than normal. Behavioral and social problems, and inadequate self-concept lead the list of secondary problems these children have in addition to abnormal anxiety and depression levels. Although it is quite clear that children with a chronic disease are at a greater risk of maladjustment, why some of these children are maladjusted and

others are not, remains unknown. Pless (1985b) writes:

As has been indicated in many of the articles in this volume (Pediatric Clinics of North America, 31), there is still a great deal of uncertainty about many of psychosocial aspects associated with the care of these children. Although there is no longer any doubt that such children are at increased risk for emotional and social problems, it still not decided what factors, apart from the presence of disease, render one child at greater risk than another. (pp. 268-269)

Tables 1 to 3 summarizes the studies that have examined the effects of cystic fibrosis and other chronic diseases on psychological functioning.

insert Tables 1, 2, 3 about here

The primary problem of these studies which has prevented any explanation to why some children are at greater risk of maladjustment than others has been that none have examined the association between the child's psychological functioning and, the family and the disease within the same sample. The exclusion of any of these crucial factors has resulted in the propagation of inconclusive and conflicting results. Based upon incomplete analysis of the whole system (family - child - disease), clinical impression, subjective interviews, and unvalidated measure, the authors have concluded:

Table 1

Presentation of studies on psychological impact of cystic fibrosis on the family functioning and psychological adjustment of the child

Impact of Cystic Fibrosis On:

<u>Family Functioning</u>	<u>Adjustment of the Child</u>	<u>Family Functioning & Adjustment of the Child</u>
Turk (1964) 0*	Spock & Stedman (1966) 2	Lawler et al. (1966) 0,2**
Tropauer et al. (1966) 0	Kulczycki et al. (1969) 0	Gayton et al. (1977) 2
McCollum & Gibson (1970) 0	Cytryn et al. (1973) 2	Steinhausen & Schindler (1984) 1,3**
Allen et al. (1974) 0	Tavormina et al. (1976) 2	Lewis & Khaw (1982) 2
Phillips et al. (1985) 1	Boyle et al. (1976) 2	
Cowen et al. (1986) 2	Drotar (1978) 0	
	Kellerman et al. (1980) 3	
	Drotar et al. (1981) 3	
	Cowen et al. (1984) 2	
	Simmons et al. (1985) 2	

* - Each study is rated on a 0 to 3 point scale where:

- 0 - descriptive study that used no control group or standardized questionnaires
- 1 - the study used a control group but no standardized questionnaires
- 2 - the study used no control group but did use standardized questionnaires
- 3 - the study used both a control group and standardized questionnaires

** - Lawler et al. (1966) and Steinhausen & Schindler (1981) used standardized questionnaires only on evaluation of the child but used interview data for family functioning. Steinhausen & Schindler (1981) did have a control family group.

Table 2

Presentation of studies on the impact of disease variables of cystic fibrosis on family functioning and the psychological adjustment of the child

Impact of Disease Variables On:

<u>Family Functioning</u>	<u>Adjustment of the Child</u>	<u>Family Functioning & Adjustment of the Child</u>
McCollum & Gibson (1970) 0*	Cytryn et al. (1973) 2	Bywater (1983) 0,2**
Leiken & Hassakis (1973) 0	Boyle et al. (1976) 2	
McCubbin et al. (1983) 2	Drotar (1978) 0	
	Drotar et al. (1981) 3	
	Zeltzer et al. (1981) 3	
	Kellerman et al. (1980) 3	
	Cowen et al. (1984) 2	
	Simmons et al. (1985) 2	

* - Each study is rated on a 0 to 3 point scale where:

- 0 - descriptive study that used no control group or standardized questionnaires
- 1 - the study used a control group but no standardized questionnaires
- 2 - the study used no control group but did use standardized questionnaires
- 3 - the study used both a control group and standardized questionnaires

** - Bywater (1983) used a standardized questionnaire only for the evaluation of the child and an interview for the family.

Table 3

Presentation of studies done on the impact of parents' coping styles on family functioning and psychological adjustment of the child

Impact of Parents' Coping Styles On:

<u>Family Functioning</u>	<u>Adjustment of the Child</u>	<u>Family Functioning & Adjustment of the Child</u>
None	Spock & Stedman (1966) 2* Tropauer et al. (1970) 0 Leiken & Hassakis (1973) 0 Lewis & Khaw (1982) 2	None

* - Each study is rated on a 0 to 3 point scale where:

- 0 - descriptive study that used no control group or standardized questionnaires
- 1 - the study used a control group but no standardized questionnaires
- 2 - the study used no control group but did use standardized questionnaires
- 3 - the study used both a control group and standardized questionnaires

Family factors and child's psychological adjustment

1. Maternal overprotection increases child maladjustment (Allen & Pearson, 1928; Spock & Stedman, 1966; Tropauer et al., 1966; Leiken & Hassakis, 1973; Mattson, 1972; Weitzman, 1984; Sabbeth, 1984)
2. Poor family functioning is a mediating variable which increases child maladjustment (Pless et al., 1972; Lewis & Khaw, 1982)

Disease factors and the child's adjustment

1. As severity of disease increases and prognosis worsens, there is an increased risk of child maladjustment (Pless & Roghmann, 1971; Drotar, 1978; Zelter et al., 1980; Kellerman et al., 1980; Drotar et al., 1981; Steinhausen & Schindler, 1981; Bywater, 1981; Orr et al., 1984).
2. Children with moderate severity will be more maladjusted than those with mild or extreme disease severity (Bruhn et al., 1971; McAnarney et al., 1974).
3. Maladjustment occurs despite good day to day functioning and severity of disease (Cytryn et al., 1973; Boyle et al., 1976; Drotar, 1978).
4. There is no or little relationship between maladjustment and severity of disease (Cowen et al., 1984; Simmons et al., 1985).

The primary concern of this study was to determine within the same sample which and what degree of disease, family and

parental factors are associated with the child's psychological functioning. For instance, is the child's clinical status more important in determining his or her psychological well-being or does it remain independent of physical parameters and associated with perhaps maternal overprotection? Only by providing an analysis of the child's psychological functioning in relation to both disease and family factors can an association be identified. Whereas, previous studies have ignored the interaction of disease and family factors as mediating variables, the interaction of these variable was paramount to the rationale of this study.

A secondary concern of the study was to examine the relationship between disease and family functioning, and parental variables. There is no doubt that raising a child with a chronic disease is stressful for the parents. The literature review on the effects of chronic disease on family functioning and parental well-being has amply demonstrated the problems and consequences that can arise. How the parents react to the stresses of the disease can have a significant effect on the child. For instance, overprotection is a much discussed problem which enhances the child's risk of secondary maladjustment (ie. Mattson, 1972). Only recently, has any systematic approach been attempted to analyse what factors can affect parental functioning.

The two most discussed parental variables include overprotection and coping behaviors. The relationship between coping, disease variables, and family functioning has received limited analysis (ie. McCubbin et al., 1983) and for

overprotection, whatever analysis have been conducted are clinical and subjective in nature (ie. Allen & Pearson, 1928; Mattson et al., 1966a, 1966b, 1971). Again, there has been no attempt to consider parental coping variables, family variables, other parental variables, and disease variables in the same study. As has been discussed, the only studies conducted thus far have examined solely disease and family functioning. Their main findings have included:

1. The worse the prognosis, the higher parent's anxiety (McCollum & Gibson, 1970).
2. Changes in disease will heighten anxiety (Leiken & Hassakis, 1973).
3. The greater the severity, the more depressed parents will become (McCollum & Gibson, 1970; Bywater, 1981).
4. Social support, self-esteem, psychological stability, family integration, and an optimistic definition of the situation are directly related to pulmonary function, weight, and height of the cystic fibrosis child (McCubbin et al., 1983).

An analysis of what variables affect parental coping might later help in the understanding of family functioning under the stress of chronic disease.

Hypotheses

The primary concern of this study was to determine what factors increase the risk of psychological maladjustment in the child. To date, the predictive model between disease and family factors and the child's psychological functioning is illustrated in Figure 1.

insert Figure 1 about here

The child's psychological functioning has been stated to be associated with the disease factors (arrow 1.) and family factors (arrow 2.). In addition, the family factors are influenced by the disease factors (arrow 3.) which place further burden on the child. These relationships have been noted in a number of previously mentioned studies. However, the relative influence of disease and family factors in relation to psychological maladjustment in the child is not known. That is, do family factors contribute any additional understanding of the child's psychological functioning after having taken the disease variable into consideration?

Figure 1.

From a purely medical or organism orientation, one would expect that the disease variable would account for the child's psychological functioning. That is, the worse the disease, the worse the child's psychological functioning. Yet, this view has not received conclusive validation. Rather, as was seen in the review of the literature section on the relationship between the child's psychological adjustment and disease variables, a linear relationship was found some of the time (ie. Pless & Roghmann, 1971; Orr et al., 1984) but not in other studies (ie. Pless et al., 1974). In addition, others have found only small correlations between disease variables and psychological functioning (ie. McCubbin et al., 1983; Cowen et al., 1984). On the other hand, the effects of parental and family factors has been found to be consistently and strongly associated with the child's psychological functioning (ie. Lewis & Khaw, 1982; Bywater, 1983).

Thus, the first hypothesis is that, although the psychological functioning of the child will be in part explained by the disease variables, parental and family factors would significantly explain more aspects of the child's psychological functioning than will the disease factors even after having taken the disease variables into consideration.

However, a note of caution must be taken in the interpretation of the model and hypothesis being studied. Both are predictive such that the additional variance understood from studying the familial and parental variables does not indicate

the direction or cause of the relationship. In other words, the assumption of unidirectionality is predictive and the cause of the relationship may well be bidirectional. In fact, the bidirectional view, ie. that children are as much responsible for way that parents behave as are parents responsible for the way children behave, has received considerable support in the developmental literature (Bell, 1968; Thomas, Chess, & Birch, 1969; Sameroff & Chandler, 1975; Thomas & Chess, 1979; Belsky, 1981). However, neither the model or hypothesis in this study underscore the importance of this bidirectional or transactional model; rather, the purpose of this study was to determine and understand the relationship of the child and family while taking into account another intervening variable, the disease. In other words, is the child's psychological state associated with how the family is functioning or whether the parent is anxious and/or depressed, or, can it is associated with disease severity?

The second concern of this study was to determine the effects of disease variables and family factors on the parents' coping style and ability. What has been generally considered to be the relationship between parents' coping and disease and family factors is illustrated as a predictive model in Figure 2.

insert Figure 2 about here

As was discussed earlier (rationale and literature review) coping with a chronic disease has been noted to most widely affected by

the disease factors (arrow 1). Disease factors have also been associated with parental and family variables (arrow 2). However, it is unknown, in a family with a child with a chronic disease, how the parents' coping is associated with family, parental, and disease variables. For instance, is coping or overprotection associated more with a disease variable such as clinical status or are they independent of disease factors and more related to a parental variable such as anxiety. There is only scant information about how these variables are interrelated. At this time, formulation of a hypothesis was unsubstantiated. Rather, the purpose of the analysis was to explore any relationship and suggest hypothesis testing.

Figure 2.

Method

Subjects

All patients aged 8 years and older at the Children's Hospital of Eastern Ontario's Cystic Fibrosis Clinic and their parents were asked to participate in the study. Of the 41 children in the clinic, 36 agreed to participate. Of the five children who refused, 1 child became upset after reading one of the questionnaires (Children's Depression Inventory (Kovacs, 1978)) and refused to continue in the study; 1 mother found the Child Behavior Checklist (Achenbach & Edelbrock, 1983) offensive and refused to have her child, her husband, and herself, return any of the questionnaires; 1 child simply refused to be involved; and 2 children and their mothers could not see any value in the study and refused to participate. Of the parents who were asked to participate, 24 mothers and 20 fathers completed all their questionnaires; an additional 5 mothers completed some of the questionnaires; 1 set of parents had 2 children; and 6 adolescents asked that their parents not be included in the study.

The distribution of the sample of children's sex and age is presented in Table 4.

insert Table 4 about here

Three breakdowns of sex and age are provided: 1. for the total 36 children; 2. for the 25 children whose mothers have completed all the study; and 3. the 20 children whose fathers have also completed the study.

Table 4

Age and sex of all the children participating in the study, and of the children whose mothers and fathers completed the study

Group	Sex	n	Age (Years)		
			Range	<u>M</u>	<u>SD</u>
Total Sample	Males	22	8-19	12.9	3.5
	Females	14	7-22	11.7	4.1
	Total	36	7-22	12.4	3.7
Mothers who Participated	Males	15	9-17	11.4	2.7
	Females	10	7-11	9.8	1.1
	Total	25	7-17	10.7	2.4
Fathers who Participated	Males	12	8-15	11.0	2.4
	Females	8	7-11	9.6	1.2
	Total	20	7-15	10.4	2.1

Dependent Measures

Measures were obtained to assess the child's and parent's psychological functioning, the effect of the disease on family functioning, and the disease status. The choice of which constructs would be measured was based upon the literature review. For the child's psychological functioning, the important factors have included: depression, anxiety, self-concept, social functioning, and behavioral problems. Family and parental factors which needed to be assessed included: depression, anxiety, impact of disease on family functioning, family functioning, overprotection, coping. The physical measures included those which have been used in previous studies as well as ones which have been demonstrated to be valid measures of disease status. All the psychological assessment tools were translated into French by the Translation Department at CHEO. These questionnaires were administered to the French speaking subjects.

Measures of Psychological Functioning of Children

"The State-Trait Anxiety Inventory for Children Ages 6 to 16 Years" (Appendix A)

The "State-Trait Anxiety Inventory for Children" (STAIC) (Spielberger, 1973) were used to measure anxiety. Both anxiety scales consist of 20 items. The state anxiety inventory measures transitory or specific situation anxiety; whereas, the trait anxiety inventory measures general anxiety. The children are asked to respond on a 3 point scale how they feel right now or how they generally feel, respectively. The two scales are

based on the adult version (see STAI) but modified so that the items are understandable for young children.

Norms on the STAIC have been developed based on the data of 737 males and 814 females from grades 4 to 6. The child's raw score is transformed into a normalized t-score.

Reliability coefficients on the trait scale of .65 for males and .71 for female; and, .31 for males and .47 for females on the state scale were found on 8 week test-retest. Internal consistency coefficients on the trait scale were .82 for males and .87 for females and coefficients of .78 for males and .81 for females were found for the state scale. Low reliability on the state scale is expected since the measure is transitory. The trait scale appears to have reasonable reliability and both scales have good consistency.

Both scales have been widely used and received extensive content, criterion, and construct validity (Spielberger, 1973). These scales were selected for use in this study because of their extensive validation, sound reliability, and popular use in psychological research.

"Child Behavior Checklist (Parent Form)" (Appendix B)

The parent form of the Child Behavior Checklist (CBCL) with the Revised Child Behavior Profile is a diagnostic rating scale used to assess behavioral disorders and social competence of children aged 4 to 16 years (Achenbach & Edelbrock, 1983). The CBCL is completed by the child's parents and can be used for both

clinical and research purposes. The CBCL contains two major scales: a behavioral problem scale and a social competence scale.

The social scale screens the areas of activities, social tasks, and school performance. The behavioral scale includes two broad band categories which classifies disorders as either internal (ie. depressed withdrawn) or external (ie. hyperactive, delinquent). These broad band categories include scales for specific behavioral problems labelled but not keyed according to the established nomenclature categories included in the DSM-III (ie. hyperactivity, obsessive-compulsive, conduct disorder). Additional categories are also provided for other behavior patterns not listed in the DSM-III (ie. hostile-withdrawal, cruel, uncommunicative).

The CBCL was developed based on the results of 2300 clinically referred children broken down by sex and age group (4-5 year olds, 6-11 years, and 12-16 years). However, norms for the checklist were standardized using 1300 normal children again broken down by sex and age. The CBCL therefore takes into account developmental issues by providing a separate profile for each gender at ages 4-5, 6-11, and 12-16 years (Achenbach, 1978; Achenbach & Edelbrock, 1979; Achenbach & Edelbrock, 1983). Normal T-scores were calculated for all scales are provided. For the behavioral problem scales, the range of normal T-scores is 50-70 and scores above the 70 th value are suggestive of clinical maladjustment. For the social scales, the normal T-score range is between 30 and 50 and values below the 30th level suggest clinical maladjustment. The CBCL also provides a global

T-score for behavioral (deviancy T-score above the 70th value) and social scales (deviancy below 30th T-score).

The CBCL has been found to have excellent reliability (Achenbach & Edelbrock, 1983), validity (Achenbach & Edelbrock, 1983), and sensitivity (Kazdin & Heidish, 1984). Test-retest reliability measured by intraclass correlation coefficients (proportion of total variance of item scores after variance due to a specific source of unreliability has been subtracted ie. rank ordering and magnitude effects) were found to be .95 and .99 at one week; .84 and .97 at 3 months followup on the behavioral and social scales respectively. A median Pearson correlation coefficient of .89 was found at one week test-retest (Achenbach & Edelbrock, 1983).

Clinically referred children were found to score significantly higher on behavioral scales and lower on the social scales than normal children suggesting good content validity. The CBCL also has good construct validity with high correlation with other behavioral questionnaires (Achenbach & Edelbrock, 1983). Kazdin & Heidish (1984) also found that high sensitivity but not as good specificity with conduct disorders and depression.

The CBCL was selected to measure the behavioral and social functioning of children with a chronic disease because it is a reliable and valid diagnostic tool and an especially powerful diagnostic research tool. The checklist has many qualities that make it appealing ie. provides for gender and age and norms using

a normal population. In addition, the scale has had repeated use in research with chronic disease and found to be an effective evaluative tool (ie. Cowen et al., 1984; Dorenbaum, Cappelli, Keene, McGrath, 1985). However, in order to limit the number of dependent variables, due to the small sample size, only the four social competence scales and the four overall behavioral scales were used.

"Children's Depression Inventory (CDI)" (Appendix C)

The CDI is a downward extension of the Beck Depression Inventory (BDI) (Kovacs & Beck, 1977) and considered to be the best researched self-report depression inventory for children (Saylor, Spirito, Finch, & Bennett, 1984). Although some have argued that child and adult depression are not similar, the majority of studies in childhood depression have not supported this view (Kovacs & Beck, 1977). The CDI was adapted in fact from BDI because the characteristics of adult depression have been sighted in childhood depression and the similarities of symptoms outweigh differences (Kovacs, 1980/1981).

The CDI consists of 27 items and the child chooses 1 of 3 descriptions that best applies to him or her in the last 2 weeks (Kovacs, 1978). Although the scale has had considerable use, only preliminary normative data is available (Kovacs, 1980/1981), and the factor structure, reliability, and validity are still being investigated (Hodges, Siegal, Mullins, & Griffin, 1983; Saylor, Spirito, et al., 1984).

Reliability has been examined for stability and internal consistency with normal and emotionally disturbed samples. One-week test-retest reliability for 28 emotionally disturbed children was .87 but only .38 for 69 normal children (Saylor, Spirito, et al., 1984). However, Kovacs (1980/1981) for normal children reported a .72 one-month test-retest coefficient. Saylor, Spirito, et al. (1984) also found a .59 six week stability coefficient for the 28 emotionally disturbed children which is very similar to a .50 coefficient found earlier by Kazdin, French, Unis, & Esveltd-Dawson (1983) for 37 emotionally disturbed children. Good internal consistency of the CDI has been reported with coefficients ranging from .86 to .94 (Kovacs, 1980/1981; Saylor, Spirito, 1984).

The CDI has also received some validation. The content and criterion validity has been established (Saylor, Spirito, et al., 1984) as has convergent and discriminative validity (Kazdin, Esveltd-Dawson, Unis, & Rancurello, 1983; Saylor, Baskin, Finch, Furey, & Kelly, 1984). However, the CDI was not found to correlate with rating of depressions provided by parents or caregivers with children not rating their depression as severe as others (Kazdin et al., 1983; Saylor, Baskin et al., 1984). As such, the CDI does need additional research before it can be totally acceptable. However, it remains the best available self-report measure of children's depression at this time.

"The Self-Perception Profile for Children (SPC)" (Appendix D)

The Self-Perception Profile for Children (1985) was developed on the model that individual children have different competency in various skill domains and that general self-worth is not a summation of different competencies but an independent construct (Harter, 1982, 1985). The SPC is a revision of "Perceived Competence Scale for Children" (Harter, 1982) expanding the number of subscales from 3 to 5. The scale provides a profile of the child's perception of esteem in the areas of scholastic competence, social acceptance, athletic competence, physical appearance, behavioral conduct, and global self-worth. competence in the cognitive, social, and physical domains, as well as a general self-worth. The advantage of this scale include: the assumption of individual areas of esteem; minimization of social desirability response tendencies; and a sound factor structure.

The SPC was initially developed on two samples (N=433) and normed based on 4 samples (N=2271). The factor structure on all samples remained the same suggesting sound factorial validity. The internal consistency of the scale was found to range from .71 to .86 based on a sample of 341 students. Three month test-retest reliability based on 208 subjects ranged from .70 to .80 and 9 month test-retest for 810 subjects ranged from .69 to .80 (Harter, 1982, 1985).

Convergent validity was demonstrated based on significant correlations between teachers ratings and the child's rating of

perceived competence. Significant correlations between motivation and competence suggested construct validity. Discriminant validity was also demonstrated (Harter, 1982). The scale appears to have good validity and excellent reliability.

Measures of Psychological Functioning in Parents and Family

"The State-Trait Anxiety Inventory for ages 16 years and older" (Appendix E)

The "State-Trait Anxiety Inventory" (STAI) (Spielberger, Gorsuch, & Lushene, 1970) also measures state and trait anxiety. The scales each consist of 20 items and respondents answer items relative to how they feel now and how they generally feel.

Normative data was collected 1366 undergraduate college students, 587 males and 879 females. The individual's raw score STAI is transformed into a normalized t-score.

Test-retest reliability was determined on 197 students at 1 hour, 20 days, and 104 days. The reliability coefficients of the trait scale were .84, .86, .73 for males and .76, .76, .77 for females, respectively. Coefficients of the state scale were as expected much smaller. Coefficients of .33, .54, .33 for males, and .16, .27, .31 for females were obtained, respectively. Internal consistency coefficients of both scales were found to range from .86 to .92 for the trait scale and .83 to .92 on the state scale.

The STAI has also been extensively validated on content, criterion, and construct validity (Spielberger, Gorsuch, and Lushene, 1970).

"Coping Health Inventory For Parents (CHIP)" (Appendix F)

The CHIP is a 45 item self-report checklist that asks parentsto evaluate coping with illness behaviors on a 3 point helpfulness scale (McCubbin & Patterson, 1985). The inventory was chosen because it was specifically developed to assess how parents cope with a chronically ill child. The underlying hypothesis of the scale states that the larger the number of coping behaviors, the more likely the family will cope effectively with a chronically ill child. The scale has not been fully validated and test-retest reliability is not known. However, the scale provides an objective measure of coping as well as having been used on several studies involving cystic fibrosis patients (Patterson & McCubbin, 1983; McCubbin, Patterson, McCubbin, & Wilson, 1983; McCubbin & Patterson, 1983).

Based on the results of the checklist to 95 mothers and 90 fathers of children with cystic fibrosis, the factor structure was determined. Three factors were identified: 1. maintaining family integration, cooperation, and an optimistic definition of the situation; 2. maintaining social support, self-esteem, and psychological stability; 3. understanding the medical situation through communication with other parents and consultation with medical staff (McCubbin & Patterson, 1983; 1985).

Normative data was obtained from a sample of 308 mothers and fathers of children with chronic illness. Individual scores on the checklist are compared to the means and standard deviations based on the normative data on each of the factor structures

(McCubbin & Patterson, 1985).

Test-retest reliability has not been determined but the internal consistency of the scale ranges from .71 to .79. The questionnaire has received some validation. Comparison of high and low conflict in families with children with cerebral palsy, high conflict families scored significantly higher on each of the dimensions (McCubbin & Patterson, 1985). Changes in the child's health status has also been reflected in changes in coping (McCubbin & Patterson, 1983).

"Beck Depression Inventory" (BDI) (Appendix G)

The Beck Depression Inventory (Beck, 1967) consists of 21 items and adults choose the best description of how they felt in the past two weeks on each item. The BDI is based on the cognitive theory of depression that depression is collection of symptoms and diagnosis is dependent upon psychobiological changes in the individual (Beck, 1967; Kovacs & Beck, 1977). The inventory is one of the most widely used for depression in adulthood.

The BDI has been normed on a sample of 529 normal adults and cutoffs of none, mild, moderate, and severe depression are established (Beck, 1967). Factor analytic studies have extracted four factors: 1. vital depression comprised of fatigability, loss of appetite, somatic preoccupation, weight loss, difficulty sleeping, and loss of libido; 2. self-debasement consisting of self-dislike, sense of failure, expectation of punishment, and guilty feeling; 3. pessimism-suicide consisting of pessimism and

suicidal wishes; and, 4. indecision-inhibition comprised of indecision and work inhibition (Beck, 1967). The BDI has been extensively shown to have excellent reliability and validity (Beck, 1967; Beck & Beamesderfer, 1974).

"Impact-on-Family Scale" (IFS) (Appendix H)

How the chronic disease affects family functioning was assessed using the "Impact-on-Family Scale" (Stein & Riessman, 1980). The scale consists of 24 items and parents answers each item on a 4 point Likert scale from strongly agree to strongly disagree. A factor analysis of results obtained from a 100 mothers with children having a chronic disease revealed four factors: 1. financial functioning; 2. social and familial; 3. personal strain; and, 4. mastery.

The scale is still in its preliminary psychometric evaluation and only internal consistency is known (alpha coefficient = .88). Validation of the the scale has not yet been completed (R. E. Stein, personal communication, May 25, 1983). However, there is no other scale available that objectively addresses the impact of the disease on family functioning.

"The Family Functioning Index" (FFI) (Appendix I)

The Family Functioning Index was developed during a study of families with a child with a chronic disease (Pless et al., 1972). The scale does not provide an indepth analysis of family functioning, but it does deal with a multi-dimensional concept of functioning that reflects clinical impressions and is readily administered (Pless & Satterwhite, 1973). The FFI consists of 16

items and from a factor analysis of 356 responses 6 principal components emerged: communication, togetherness, closeness, decision making, child orientation, and marital satisfaction.

The FFI has not been analysed for test-retest reliability or internal consistency. However, a correlation of .72 between husbands and wives scores was noted as a form of reliability (Pless & Satterwhite, 1982).

Validation of questionnaire is also limited but the evidence found from three studies does suggest reasonable validity. On the first study, the ratings of 8 social care workers of 43 families were found to correlate significantly with FFI scores of the mothers ($r=.48$) and fathers ($r=.35$). Similarly, a second study found significant correlation between 6 care workers and FFI scores from 65 families ($r=.39$). Finally, a study comparing families receiving counselling ($N=43$) scored significantly lower on family functioning than a random sample of normal families ($N=356$) (Pless & Satterwhite, 1982).

The FFI is probably not as psychometrically sound a measure of family functioning as some other scales. However, Pless & Satterwhite (1972) emphasize that the FFI is only an index not a scale and should be viewed as a screening tool rather than as an explanation tool. The advantage of the FFI over other scales, is that it has been used in studies of chronically ill children and comparison can be made to these findings.

"The Parental Bonding Instrument" (PBI) (Appendix J)

The Parental Bonding Instrument (Parker, Tupling & Brown, 1979) was developed to measure parental care and overprotection in the child's first 16 years. The PDI was designed for an adult in retrospect to score his or her parents during the first 16 years. After a number of factor analytic studies to reduce items, the final analysis yielded two factors which accounted for 52 percent of the variance: 1. care/involvement versus indifference/rejection; and, 2. control/overprotection and intrusion versus encouragement of independence. The final draft of the PBI consists of 25 items, 12 measuring care, and 13 overprotection answered on a four point Likert scale ranging from very like to very unlike.

The scale was standardized based on a non-clinical Australian sample of 410 adult respondents (Parker et al., 1979), a non-clinical sample British sample of 132 adults (Parker, 1983), and a clinical sample of 212 respondents (Parker, 1983). There were no differences on either the care or overprotection scales but in all cases mothers were scored as more caring and overprotective than fathers. In addition, intercorrelations ranging from $-.20$ to $-.46$ were found between the two dimensions suggesting that care and overprotection are in fact related.

The reliability and validity of the scale was also checked on clinical and non-clinical samples. On a sample of 17 non-clinical respondents three week test-retest correlation coefficients of $.76$ (care) and $.63$ (overprotection) were found

compared to a nine-week test-retest correlation for 36 depressed patients .87 (care) and .92 (overprotection). Split-half reliability of the scale based on the 17 respondents were found to range from .85 (care) to .74 (overprotection). The scale appears to have reasonable reliability.

Concurrent, content, and construct validity of the scale was determined for non-clinical and clinical samples based on interview data, administration of the scale to different family members, and comparison of clinical and non-clinical scores (Parker et al., 1979; Parker, 1983). In all cases, the results demonstrated the validity of the care and overprotection scales.

Although the scale appears to have reasonable psychometric qualities, there is one consideration. The scale was designed for adults or adolescents to determine the extent of their parents' care and overprotection. However, the sample in this study included for the most part children under 12 which was the youngest age that Parker (1983) included in his validation study. However, Parker (1983) as part of his validation asked mothers to rate how their children would rate them as well as asking children to rate their mothers. Significant correlations were found although mothers tended to score themselves as more caring and less overprotective. Ideally the child should complete the scale, but since the children in this study were too young to understand the scale, the second method of having the parent complete the questionnaire was used. This will inevitably affect the scores of caring and overprotection but to a lesser extent,

the validity. However, this approach must be utilized since there exists no other objective questionnaire which has received similar psychometric analysis.

Physical Measures of the Child and Disease

"Nutritional Assessment"

The percent weight for height and the nutritional classification will be used to determine the nutritional status of each child.

Weight and height represent the two most basic and widely used anthropometric measures (Jensen, Englert, & Dudrick, 1983; Guthrie, 1983). Both provide valuable nutritional information but since each measure is individualistic, they cannot be used to compare the nutritional status between children (Jelliffe, 1966). A standardized measure, percent weight for height, is available that incorporates the two measures on growth curves.

There are various standardized growth curves for percent weight for height (ie. Tanner, 1978; Martin & Beal, 1978). However, the most widely used standard (including at CHEO) is the Growth National Center for Health Statistics Percentiles (NCHS; Hamill, Drizd, Johnson, Reed, Roche, & Moore, 1979). These are the most recent standards for males and females aged 2 to 18 years. The percentiles were normed on the weights and heights of 20,000 subjects and generalizable for 70 million American children. Each child's weight for height is plotted on a growth curve and the percent weight for height is determined by the child's position on the curve. The 50th percentile is the average

percent weight for height and scores between the 25th and 75th percentile are considered normal (Hamill et al., 1979).

Although the Cystic Fibrosis Clinic at CHEO uses the NCHS, the clinic has arbitrarily changed its scoring. Children with cystic fibrosis are often below average in percent weight for height. In view of this, the CF clinic has defined the normal 50th percentile as the 100 percentile for their CF patients. Therefore a child with CF at CHEO with a 100 percent weight for height is actually only on the 50th percentile for normal children. Although the score has changed, the value in comparison to other children with cystic fibrosis remains the same.

The final anthropometric measure to be included in the study is the nutritional classification of the child. The nutritional classification is based on the expected percent weight for height and percent height for age and has four categories; 1. normal (between the 90th and 100th percentile); 2. stunted (less than 82.5% in height for age but normal weight); 3. wasted (less than 80% expected weight for height but normal height for weight); and 4. wasted and stunted (less than 80% expected weight for height and 82.5 percent height for age) (Waterlow, 1972; Waterlow, 1973). This system was developed on the results of a number of world wide studies on malnutrition (Waterlow, 1972). The classification system is widely used, can distinguish between acute (usually wasted) and chronic (usually stunted or wasted and stunted) malnutrition, and sensitive to improvements in nutrition (Trowbridge, 1979).

"Pulmonary Assessment"

Three measures of pulmonary function were used in this study: 1. forced vital capacity (FVC); 2. forced expiratory volume (FEV 1); and, 3. forced expiratory volume 25% to 75% (FEV 25-75%). The FVC is the volume of gas (breath) that a subject can expire as forcefully and rapidly as possible after maximal inspiration (Ruppel, 1975). FEV 1 is the forced expiratory volume of gas expired over one second during the FVC (Ruppel, 1975). The FEV 25-75% is the average rate of gas flow during the middle half of a forced expiratory volume (Ruppel, 1975). All three measures are recorded in litres and measured using a spirometer (Ruppel, 1975).

All three measures have been standardized for sex and age by Polgar & Promadhat (1971). These standards were normed based on the results of 34 studies involving over 45,000 subjects between the ages of 3 and 19 years between 1921 and 1970 (Polgar & Promadhat, 1971). The validity of pulmonary measures to reflect disease severity has been demonstrated in studies showing a correlation between the pulmonary measures and the child's clinical status (Doershuk, Matthews, Tucker, & Spector, 1965; Taussig et al., 1973; Brasfield, Hicks, Soong, & Tiller, 1979) and response to therapy (Corey, Levison, & Crozier, 1976; Beaudry, Marks, McDougall, Desmond, & Rangel, 1980; Hyatt, Chipps, Mellits, Lietman, & Rosenstein, 1981; Nolan, McIvor, Levison, Fleming, Corey, & Gold, 1982).

"Clinical Status"

The National Institute of Health (NIH) score (Taussig et. al., 1973) was used to evaluate the child's clinical status. The NIH score has already been briefly discussed (see section Effect of Disease Variables on Psychological Functioning). The NIH score is used by the CF clinic at CHEO because it is the only objective measure of clinical status available for CF. The score is computed by the CF medical staff at CHEO based on the set criteria.

The NIH score was developed over a twelve year period by following 73 CF patients ages 5 to 30 years on 3 year follow-ups. The score is calculated based on chest roentgenograms, pulmonary function tests, weight, activity, attitude, and disease complications (hemoptysis, pneumothorax, and cor pulmonale). Scores range from 0 to 100 reflecting poor to good prognosis. A correlation coefficient of .99 for interscorer reliability was found (Taussig et al., 1973). The validity of the NIH scale was supported by mortality rates with patients with high scores surviving significantly longer than patients with low scores.

Procedure

Permission from the Director of Cystic Fibrosis Clinic and the Ethics and Research Committee of the Children's Hospital of Eastern Ontario to conduct the study was obtained.

At regular CF clinic visits held usually every Tuesday morning, the patients were asked if they would participate in the

study. In almost all cases (exception of the 6 adolescents who came to clinic alone and 1 case whose father came to the clinic), the mother was also present. They were told that the purpose of the study was to determine: 1. in general, how well the clinic CF patients, the parents and family as a group cope with the disease; 2. what factors, ie. physical, parental, or family are associated with how the child feels. All subjects were assured confidentiality and written informed consent was obtained from the participating subjects.

During normal clinics, the patient's physical status is monitored and changes in therapy are initiated if necessary. There are usually 8 to 12 children in each clinic visit. The comprehensive nature of these visits requires the child to undergo various tests from different care-givers ie. nutrition, physiotherapy, respiratory, and x-rays. The children are individually sent from station to station so that waiting time is kept to a minimum. The Director of the Clinic has provided permission to include this study as part of the overall assessment of the child during the clinic visit.

In order not to disturb the flow of physical testing and to limit the amount of time the parent and child has to remain after the clinic visit to complete questionnaires, only the questionnaires that the child was to complete were administered. The children completed first the Self-Perception profile for Children, followed by the STAIC scales, and finally the Children's Depression Scale.

The mothers were given instructions on how to complete their questionnaires and were also given a set of questionnaires for the fathers to complete. Both parents completed the same sets of questionnaires at home except for the mother who was also asked to complete the Child Behavior Checklist. A separate pre-stamped pre-addressed envelope for the mother and father was provided for the return of the questionnaires. Parents who did not return their questionnaires were followed-up by telephone and asked to mail their responses as soon as possible.

Gathering of Physical Measures

All physical status measures were obtained from the medical charts of the patients. The nutritional and pulmonary scores for each child are collected minimumly every three months and clinical scores yearly.

Ideally, these measures should be obtained at the same time as the psychological assessment. However, this was not feasible since neither this researcher has the expertise to conduct these tests nor can the staff at the Children's Hospital provide the time and expense to conduct the tests. However, the physical scores do reflect the child's present condition. If the child is doing physically well, the three month testing is sufficient to reflect present state. If however, the child's physical condition had been deteriorating since the previous clinic visit, the child would have been re-tested and these measures would reflect the present physical status.

Data Analysis

Although obtaining data from more subjects would be ideal, the limited number of children with cystic fibrosis at CHEO precluded a larger sample size. The present study should be viewed as a preliminary attempt to determine and describe the association between the child's psychological functioning and parental, family, and disease variables.

Sample Comparisons

Since all parents did not complete the questionnaires, an analysis on the children's measures comparing the scores of those children whose parents participated in the study versus those whose parents did not participate was conducted. The results of the t-tests on the children's scores for group whose mothers completed their questionnaires (n=25) versus whose mothers did not participate or complete all the questionnaires (N=11) is presented in Table 5, and the results of the comparison of the

insert Table 5 about here

children's scores whose fathers participated in the study (N=20) and those who did not (N=16) are presented in Table 6.

insert Table 6 about here

Table 5

Comparison of childrens' psychological and physical measures grouped by mothers who completed all the questionnaires (Group 1: n=25) versus mothers who did not participate in the study or completed only parts of the questionnaires (Group 2: n=11)

	Group				
	Mothers Group 1		Mothers Group 2		
	<u>M</u>	<u>SD</u>	<u>M</u>	<u>SD</u>	<u>t</u> (34)
<u>Children's Measures</u>					
Depression	3.6	4.1	7.2	5.0	2.26 *
State Anxiety	41.7	9.5	50.8	14.4	2.19 *
Trait Anxiety	36.6	9.0	46.7	11.6	2.74 **
Scholastic Competence	3.2	.6	2.8	.8	1.29
Social Acceptance	3.4	.4	3.1	.4	2.22 *
Athletic Competence	3.1	.7	2.8	.7	1.22
Physical Appearance	3.0	.6	2.6	.6	1.56
Behavioral Conduct	3.4	.6	3.1	.8	1.01
Global Self-Worth	3.4	.5	3.0	.7	1.70
Age	10.7	2.3	16.4	3.4	5.72 **
<u>Physical Measures</u>					
Weight/Height	95.7	9.2	92.0	16.7	< 1
Nutritional Status	1.4	.8	1.9	1.1	1.65
NIH Score	79.6	19.1	74.6	13.6	< 1
FVC	85.5	22.7	88.9	30.8	< 1
FEV 1	68.8	25.6	72.1	31.6	< 1
FEF 25-75%	52.6	32.1	54.0	40.6	< 1

* $p < .05$

** $p < .01$

Table 6

Comparison of childrens' psychological and physical measures grouped by fathers who completed all the questionnaires (Group 1: n=20) versus fathers who did not participate in the study or completed only parts of the questionnaires (Group 2: n=16)

	Group				
	Fathers Group 1		Fathers Group 2		
<u>Children's Measures</u>	<u>M</u>	<u>SD</u>	<u>M</u>	<u>SD</u>	<u>t</u> (34)
Depression	4.5	4.6	4.9	4.7	< 1
State Anxiety	42.9	11.5	46.1	12.1	< 1
Trait Anxiety	37.7	10.4	41.8	11.0	1.13
Scholastic Competence	3.2	.6	2.9	.7	1.18
Social Acceptance	3.4	.5	3.2	.5	1.37
Athletic Competence	3.1	.7	2.8	.7	1.18
Physical Appearance	3.1	.6	2.6	.6	1.91
Behavioral Conduct	3.3	.7	3.2	.7	< 1
Global Self-Worth	3.4	.6	3.2	.6	< 1
Age	10.4	2.0	15.0	3.8	4.53 **
<u>Physical Measures</u>					
Weight/Height	95.4	8.9	93.5	15.1	< 1
Nutritional Status	1.4	.8	1.7	1.1	1.28
NIH Score	79.4	19.9	76.4	14.4	< 1
FVC	86.4	22.3	86.7	28.9	< 1
FEV 1	69.6	24.7	70.0	30.7	< 1
FEF 25-75%	53.4	33.4	52.5	36.7	< 1

** $p < .01$

Children whose parents completed all the questionnaires were significantly older than the group of children whose parents did not complete the study. No differences were noted in any of the physical status measures for either group. Whereas, the children's scores on the two father groups did not differ, children whose mothers' did not complete the questionnaires scored higher on the depression scale, on both the state and trait anxiety measures, and scored lower on self worth based upon social acceptance. Non-significant findings were probably not obtained in the two father group comparisons due to younger children's scores who came from broken families entered into the group of fathers who did not complete the study.

A comparison of mothers and fathers (N=20) scores on the various measures is presented in Table 7.

insert Table 7 about here

Only the communication subscale of the Family Functioning scale was found to be significant with mothers reporting less communication in the family than fathers. In addition, the parents scores were not highly correlated suggesting much individual variability within the family. These low correlations will be later seen to produce different factor structures for each parent.

Table 7

Comparison of mothers' and fathers' scores

	Mothers		Fathers			
	<u>M</u>	<u>SD</u>	<u>M</u>	<u>SD</u>	<u>t</u> (19)	<u>r</u>
<u>Impact on Family</u>						
Financial	10.4	3.0	11.5	3.5	1.48	.44 *
Familial and Social	25.6	4.6	27.7	5.6	2.05	.60 **
Personal Strain	15.6	2.7	18.7	6.2	2.06	.06
Mastery	8.6	2.5	8.7	1.9	< 1	.40
Total	60.1	9.7	65.5	12.8	2.02	.47 *
Depression Inventory	5.2	4.9	5.6	7.7	< 1	-.06
State Anxiety	48.4	11.0	45.4	11.9	< 1	.37
Trait Anxiety	42.3	11.6	44.9	13.0	< 1	.20
<u>Parental Bonding Instrument</u>						
Parental Care	29.8	3.6	27.6	3.8	< 1	.10
Parental Overprotection	12.7	5.0	12.0	5.4	1.96	.45 *
<u>Coping Health Inventory</u>						
Family Integration	41.2	10.9	38.3	8.8	1.07	.26
Support	31.1	12.1	29.2	12.0	< 1	.39
Understanding	16.7	5.9	13.8	6.3	1.78	.28
<u>Family Functioning Index</u>						
Communication	5.5	1.8	6.5	1.1	2.26 *	.19
Togetherness	2.7	.7	2.9	.2	1.29	.24
Closeness	3.4	1.4	3.6	.9	1.01	.54 **
Decision Making	2.7	1.2	2.8	1.0	< 1	.60 **
Child Orientation	4.9	1.5	4.9	1.4	< 1	.71 **
Marital Satisfaction	9.6	.7	10.0	.7	1.44	-.09
Total Index	28.7	4.6	30.7	2.6	2.05	.41

* $p < .05$ ** $p < .01$

Reducing The Number of Variables

A problem that plagues many of the medical behavioral research studies is the use of multiple predictor and outcome measures which increase the experimentwise error rate. In order to control type I error, elimination of redundant variables was conducted using the system discussed by Shaffer (1979).

Principal component analysis with orthogonal varimax rotation were individually conducted on the sets of children's, mothers', fathers', and physical measures. Three criteria were used to retain variables: 1. only factors with an eigenvalue greater than 1 were retained; 2. only the variable with the highest loading on the factor was retained; and, 3. since multiple regression would be subsequently employed in the data analysis, the ratio of predictor variables to sample size was selected to be no less than 1:5 (therefore, with 36 subjects, only 7 predictor variables would be retained).

The results of the principal component analysis of the children's set of data is presented in Table 8.

insert Table 8 about here

Five factors were extracted in the component analysis explaining approximately 80.5 percent of the total variance. The first factor clearly loaded on the behavioral subscales of the Child Behavior Checklist and accounted for 29.2 percent of the overall variance. The second factor loaded on the social competence

Table 8

Factor loadings obtained in the principal component analysis with orthogonal varimax rotation of the childrens' measures

<u>Childrens' Measures</u>	<u>Factors</u>				
	1	2	3	4	5
Depression	.24	-.07	.69*	-.25	-.27
State Anxiety	.17	-.19	.85**	-.03	-.08
Trait Anxiety	.09	.21	.72*	.11	-.02
<u>Self-Perception Profile</u>					
Scholastic Competence	.01	-.12	-.50	.29	-.37
Social Acceptance	.15	.37	-.25	.64*	.01
Athletic Competence	.06	-.01	-.22	.64**	.26
Physical Appearance	-.12	.09	-.17	.22	.65**
Behavioral Conduct	-.34	-.07	-.26	.37	.07
Global Self-Worth	-.08	.25	-.61*	.57*	.19
<u>C.B.C.L.</u>					
Activities Competence	.12	.56*	.14	.58*	-.29
Social Competence	.15	.84*	.04	-.15	.21
School Performance	-.28	.68*	-.04	.13	.05
Total Social Competence	.02	.95**	-.03	.27	-.01
Number of Behavioral Problems	.97**	.03	.11	-.08	-.04
T score Behavioral Problems	.96*	-.04	.14	.04	-.09
T score Internalizing Problems	.90*	.01	.12	.12	-.05
T score Externalizing Problems	.94*	-.06	.07	.04	.01
<u>Component Summary</u>					
Eigenvalue	4.96	3.69	2.63	1.25	1.12
Percent of Variance	29.2%	21.8%	15.5%	7.4%	6.6%
Cumulative Percent	29.2%	51.0%	66.5%	73.9%	80.5%

* Variables with loadings on the factor

** Variables retained from the factor

subscales of the Child Behavior Checklist and accounted for an additional 21.8 percent of the variance. The third factor appeared to measuring general psychological functioning with high loadings of depression, state and trait anxiety, and overall general self-worth. This factor accounted for another 15.5 percent of the variance. The last two factors loaded for the most part on the subscales of the Self-Perception Profile for Children. An additional 7.4 and 6.6 percent of the variance was accounted for, respectively. The following variables were retained from each of the factors obtained in principal component analysis of the children's data: 1. number of behavioral problems; 2. T-score on the social competence scale; 3. state anxiety; 4. athletic competence; and 5. self-worth based upon physical appearance.

Five factors accounting for 74.7 percent of the total variance were extracted on the mothers' set of data. The results of the component analysis are listed in Table 9.

insert Table 9 about here

The first factors had high loadings on the family functioning variables accounting for 22.9 percent of the variance. The family functioning index was retained on this first factor. The second factor accounted for 21.6 percent of the variance and loaded on the Impact on Family scales with the highest loading on the total impact score which was subsequently retained. With the exception of the togetherness subscale of the Family Functioning

Table 9

Factor loadings obtained in the principal component analysis with orthogonal varimax rotation of the mothers' measures

	Factors				
	1	2	3	4	5
<u>Impact on Family</u>					
Financial	.01	.74*	-.01	-.16	-.29
Familial and Social	-.06	.92*	-.08	.07	-.02
Personal Strain	.06	.83*	-.24	-.09	.21
Mastery	-.53*	.32	-.13	.08	.45
Total	-.08	.97**	-.08	-.01	.11
Depression	.07	-.02	-.02	.46*	.04
State Anxiety	-.03	.11	.21	.74*	.03
Trait Anxiety	-.09	-.21	.11	.79**	.18
<u>Parental Bonding Instrument</u>					
Parental Care	-.01	-.30	.14	-.12	-.59*
Parental Overprotection	-.05	-.37	.22	.05	.79**
<u>Coping Health Inventory</u>					
Family Integration	.21	-.12	.79*	.23	-.40
Support	-.05	-.28	.76*	.31	.02
Understanding	-.19	-.18	.86**	.01	-.04
<u>Family Functioning Index</u>					
Communication	.87*	-.01	.14	-.18	.11
Togetherness	-.21	-.16	-.56*	.47	-.32
Closeness	.87*	-.09	-.23	.13	.01
Decision Making	.78*	.08	.43	.01	.03
Child Orientation	.70*	.09	-.08	-.07	-.24
Marital Satisfaction	.49*	-.04	-.17	-.19	-.01
Total Index	.95**	-.01	-.05	-.04	-.03
<u>Component Summary</u>					
Eigenvalue	4.57	4.32	2.47	2.01	1.55
Percent of Variance	22.9%	21.6%	12.4%	10.1%	7.8%
Cumulative Percent	22.9%	44.5%	56.9%	66.9%	74.7%

* Variables with loadings on the factor

** Variables retained from the factor

Scale, the third factor loaded highly on the 3 scales obtained in the CHIP. The factor accounted for an additional 12.4 percent of the variance and the third subscale score of the CHIP, the coping by understanding medical situation, was retained. The fourth factor accounted for 10.1 percent of the variance and loaded on the anxiety and depression scales with the highest loading and retained variable being the trait anxiety scale score. The final factor loaded on the 2 scales of the Parental Bonding Instrument accounting for an additional 7.8 percent of the variance. The overprotection subscale score was retained since it had the highest factor loading.

The factor analysis of the fathers' set of measures did not produce factors as defineable as the childrens' or mothers' scores. The results of the component analysis of the fathers' data is presented in Table 10.

insert Table 10 about here

Five factors were extracted accounting for 75.1 percent of the total variance. The first factor which accounted for 36.1 percent of the variance loaded on the scales of the Impact of Disease scale, state and trait anxiety scores, and the depression score. The total score on the Impact of Disease scale was retained since it had the highest loading. The second factor which accounted for 15.3 percent of the variance had high factor loadings on the Family Functioning scales as well as the overprotection scale. The family functioning index was retained.

Table 10

Factor loadings obtained in the principal component analysis with orthogonal varimax rotation of the fathers' measures

	Factors				
	1	2	3	4	5
<u>Impact on Family</u>					
Financial	-.81*	.15	-.16	-.10	-.28
Familial and Social	-.91*	.14	-.20	-.01	.01
Personal Strain	.54*	.27	.23	.17	-.41
Mastery	.04	-.09	.12	.01	.65*
Total	-.93**	.16	-.17	-.04	.02
Depression	.76*	.02	.28	-.06	-.29
State Anxiety	.65*	.30	.30	.26	.22
Trait Anxiety	.69*	-.17	.43	-.03	.29
<u>Parental Bonding Instrument</u>					
Parental Care	-.07	.05	.08	.03	-.10
Parental Overprotection	.37	-.56*	.45	.18	-.01
<u>Coping Health Inventory</u>					
Family Integration	.38	.05	.85**	-.19	.12
Support	.44	-.21	.74*	.15	.06
Understanding	.47	-.44	.38	.05	.42
<u>Family Functioning Index</u>					
Communication	-.03	.21	.02	-.82**	.04
Togetherness	.40	.01	-.21	.38	.11
Closeness	-.13	.77*	-.21	-.18	-.06
Decision Making	.49	.57*	.23	.20	-.28
Child Orientation	-.28	.63*	.17	.27	-.06
Marital Satisfaction	-.04	.07	.07	.11	-.18
Total Index	-.04	.93**	-.02	-.23	-.06
<u>Component Summary</u>					
Eigenvalue	7.22	3.06	1.76	1.54	1.43
Percent of Variance	36.1%	15.3%	8.8%	7.7%	7.2%
Cumulative Percent	36.1%	51.4%	60.2%	67.9%	75.1%

* Variables with loadings on the factor

** Variables retained from the factor

The final three factors accounted for approximately the same variance. The third factor accounted for 8.8 percent of the variance, the fourth factor, 7.7 percent, and the fifth factor, 7.2 percent of the variance. The coping by maintaining family integration subscale score of CHIP was retained in the third factor; the communication scale of the Family Functioning Scale was retained in the fourth factor; and the final measure retained was the mastery subscale score of the Impact on Family Scale.

The final principal component analysis was conducted on the physical measures. As can be seen in Table 11, two factors accounting for 89.4 percent of the total variance were extracted.

insert Table 11 about here

The first factor had high loadings on the pulmonary measures and the NIH score and accounted for 63.4 percent of the variance. The FEV 1 had the highest factor loading and was retained. The second factor which accounted for an additional 18.8 percent of the variance had high loadings on the two anthropometric scores. The nutritional status score was retained since its factor loading was higher than that of percent weight for height.

The variables retained from the four principal component analysis were then employed in the hypothesis testing.

Table 11

Factor loadings obtained in the principal component analysis with orthogonal varimax rotation of the physical measures

	Factors	
	1	2
Weight/Height	.20	.67*
Nutritional Status	-.21	-.72**
NIH Score	.70*	.34
FVC	.82*	.36
FEV 1	.96**	.28
FEV 25-75%	.89*	.14

Component Summary

Eigenvalue	3.81	1.13
Percent of Variance	63.4%	18.8%
Cumulative Percent	63.4%	89.4%

* Variables with loadings on the factor

** Variables retained from the factor

Analysis of the First Hypothesis

Multiple regressions were employed to determine the association between the child's psychological functioning, parental and family variables, and physical status. Each of the 5 children's measures obtained in the component analysis were used as outcome variables. The predictor variables included the mothers, fathers, and physical scores retained in previous component analysis. Regressions were conducted independently for each parent and blockwise entry was employed to determine the importance of the predictors. Two blocks consisting of the physical measures, and the second, parental and family measures were entered using forward selection and within each block, stepwise selection was used to determine the best predictors. F-tests on the increment of variance on the predictors were conducted in order to determine only those variables that accounted for significant variance. The physical measures block was entered first into the regression followed by the entry of the parental and family measures block. The sequence of entry is based on the hypothesis that parental and family variables contribute significantly additional variance above that of the physical measures.

Analysis of the Second Hypothesis

The Coping Health Inventory for Parents and the Parental Bonding Instrument were used to obtain the criterion measures of coping, care, and overprotection. Multiple regressions were again employed to determine the effect of disease, parental and family

variables on coping, care, and overprotection. The format of the regressions was the same as in the first hypothesis analyses. The predicted measures were the three scales on the CHIP, and the care and overprotection scales of the Parental Bonding Instrument. The two physical scores, nutritional status and FEV 1, were entered as the first block of variables followed by the second block of variables which included the depression, anxiety, Impact on Family total score, and the Family Functioning Index score. The regressions were conducted separately for each parent.

Results

Analysis of Mothers' Data: First Hypothesis

The first set of regressions analysed the predictability of mothers score (trait anxiety, overprotection score, coping by understanding the medical situation, the total score on the Impact on Family scale, and the Family Functioning Index score) and physical scores (nutritional status and FEV 1) on the childrens' number of behavioral problems, social competence, state anxiety, atheletic competence, and physical appearance self-esteem. The regressions were conducted with list-wise deletion and were therefore based on the 25 subjects with complete sets of data. Since 7 predictor variables were used, the F values that are reported in each step of the regressions had the following degrees of freedom: step 1 (1,25); step 2 (2,24); step 3 (3,23); step 4 (4,22); step 5 (5,21); step 6 (6,20); step 7 (7,19).

The number of behavior problems as measures by the Child Behavior Checklist was found to be highly predicted by the mothers' trait anxiety score and overprotection score. The results of the regression are presented in Table 12.

insert Table 12 about here

Nutritional status and FEV 1 accounted for only 1.7 percent of the variance. When the mother's trait anxiety score was entered into regression, it accounted for an additional 43.1 percent of the variance which is significant ($F(1,21)=156.64, p<.01$). Although overprotection accounts for an additional 8.2 percent of the variance, this was not significant ($F(1,20)=3.56, p>.05$). Positive simple correlations were found between trait anxiety and overprotection and the number of behavior problems. Therefore, the higher the mothers' scores on trait anxiety and overprotection, the greater the number of behavioral problems evidenced by the children. Although the final three variables are also significant in the regression, they do not account for significantly more variance. Since the F-value obtained in the increment of variance due to overprotection was not significant, by default, the increment in variance due to the remaining variables were also not significant.

Table 12

Number of behavioral problems obtained from the CBCL predicted by physical and mother scores

Variable	r	Cumulative R square	R square chg.	F
Nutritional Status	.11	.011	.011	< 1
FEV 1	.02	.016	.005	< 1
Trait Anxiety	.63	.447	.431	5.66 **
Overprotection	.47	.529	.082	5.62 **
Total F.F.I	.09	.565	.036	4.93 **
Total Impact	-.22	.574	.009	4.05 **
Coping by Understanding	.29	.578	.004	3.33 *

* $p < .05$

** $p < .01$

Whereas behavioral problems appears to be predicted by the mothers' score on trait anxiety and overprotection, social competence was found to associated with the disease status of the child. The results of the regression, as listed in Table 13, shows a strong association between nutritional status and social competence.

insert Table 13 about here

Nutritional status accounted for 18.5 percent of the variance. Although FEV 1 accounted for an additional 7.1 percent of variance, this was not significant ($F(1,22)=2.08$, n.s.). The simple correlation between nutritional status and social competence was found to be negative which demonstrates the more normal the child's nutritional status, the more competent he or she is socially. Likewise, the better the pulmonary function as measured by FEV 1, the higher the child's score on social competence.

No association was found for either physical or mothers' score on the child's state anxiety score. The physical disease variables accounted for only 3.7 percent of the variance and the mother variables contributed an additional 27.5 percent of the variance.

Table 13

T score of the CBCL social competence scale predicted by physical and mother scores

Variable	r	Cumulative R square	R square chg.	<u>F</u>	
Nutritional Status	-.43	.185	.185	5.25	*
FEV 1	.41	.256	.071	3.79	*
Coping by Understanding	-.11	.280	.024	2.76	
Total Impact	-.03	.331	.051	2.47	
Overprotection	.09	.335	.004	1.92	
Trait Anxiety	.09	.337	.002	1.52	
Total F.F.I.	.08	----	----	----	**

* $p < .05$

** The family functioning index was not entered because F was too small

Much like the results obtained in the social competence regression, athletic competence as determined by the child's ratings on the Self-Perception Profile, can be seen in Table 14, to be predicted for the most part by the disease variables.

insert Table 14 about here

The nutritional status of the child accounted for 39.5 percent of the variance. Fev 1 did not contribute additional significant variance though the mothers coping by understanding the medical situations accounted for an additional 14.4 percent of the variance ($F(1,21)=6.80, p < .01$). The remaining variables were not found to increment significantly the proportion of variance. The correlations between the nutritional status and coping variables with athletic competence indicated that children with better nutritional status scored higher on the scale and those children with higher scores on the athletic competence had mother's who sought more support from the medical situation.

The child's physical appearance self-esteem was however predicted not by the disease variables, but by the mothers' scores, particularly, the Impact on Family scale. As seen in Table 15, only the Impact on Family score was significant accounting for 19.3 percent of the variance after the physical scores had been entered into the regression.

Table 14

Athletic competence scale predicted by physical and mother scores

Variable	r	Cumulative R square	R square chg.	<u>F</u>	
Nutritional Status	-.62	.395	.395	16.38	**
FEV 1	.28	.398	.003	7.96	**
Coping by Understanding	.38	.543	.144	9.10	**
Trait Anxiety	.06	.548	.005	6.68	**
Total F.F.I.	-.01	.549	.001	5.12	**
Total Impact	.12	.550	.001	4.07	**
Overprotection	-.14	.5501	.0001	3.32	*

* $p < .05$
 ** $p < .01$

insert Table 15 about here

The Pearson correlation between these two variables suggests a strong association between high scores on the child's self worth based upon physical appearance and the ability of the families to cope with impact of the disease.

Analysis of Fathers' Data: First Hypothesis

The second set of regressions analysed the predictability of fathers' scores (coping by family integration, Family Functioning Index score, Impact on Family mastery score, family communication, and, the total score on the Impact on Family scale) and physical scores (nutritional status and FEV 1) on the childrens' number of behavioral problems, social competence, state anxiety, atheletic competence, and physical appearance self-esteem. The regressions were based on the 18 of 20 subjects who completed the study since no scores were available for 2 fathers on the Family Functioning Index. Since 7 predictor variables were used, the F values that are reported in each step of the regressions have the following degrees of freedom: step 1 (1,16); step 2 (2,15); step 3 (3,14); step 4 (4,13); step 5 (5,12); step 6 (6,11); step 7 (7,10).

Table 15

Children's self-esteem based upon physical appearance predicted by physical and mother scores

Variable	r	Cumulative R square	R square chg.	F
Nutritional Status	-.34	.096	.096	2.65
FEV 1	.20	.120	.024	1.64
Total Impact	.51	.333	.213	3.83 *
Trait Anxiety	-.17	.341	.008	2.85
Overprotection	-.12	.343	.002	2.20
Coping by Understanding	-.12	.345	.002	1.75
Total F.F.I.	-.04	----	----	---- **

* $p < .05$

** Total F.F.I. was not entered because F was too small

Only the child's self-esteem based upon athletic competence was found to have a significant association with either the physical or father scores. Similar to the mothers' findings, the child's athletic competence was found associated mostly with the disease variables. The results of the regression presented in Table 16 show that nutritional status accounted for 29.3 percent of the variance. Neither FEV 1 ($F(1,15) = 1.91$, n.s.), nor coping by integration ($F(1,14) = 2.42$, n.s.) contributed significant variance.

insert Table 16 about here

The regressions conducted on the the number of behavioral problems, social competence, state anxiety, and self-esteem based upon physical appearance were not significant. This was probably due to use of a small sample size.

Table 16

Children's self-esteem based upon athletic competence predicted by physical and father scores

Variable	r	Cumulative R square	R square chg.	<u>F</u>
Nutritional Status	-.54	.293	.293	6.65 *
FEV 1	.31	.373	.080	4.47 *
Coping by Integration	.21	.466	.093	4.07 *
Impact on Mastery	.18	.492	.026	3.15
Total Impact	.05	.512	.020	2.52
Total F.F.I.	-.19	.553	.041	2.27
Family Communication	.12	.561	.008	1.83

* $p < .05$

Analysis Second Hypothesis

1. Coping By Maintaining Family Integration, Cooperation, and an Optimistic Definition of the Situation

The mother's score on this coping strategy was not found associated with either of the physical measures or any of the mothers' remaining scores. However, the father's score, as seen in Table 17, could be predicted by his score on trait anxiety.

insert Table 17 about here

It is interesting to note that both trait anxiety and the total impact of disease have the same correlation value but that only trait anxiety is significant. This illustrates a problem with regression known as multicollinearity that when two variables such as trait anxiety and impact of disease are highly intercorrelated ($r = -.81$) only the variable which is slightly more correlated with the predicted variable will be retained in the regression. Thus, there is a sort of masking in the data in that although both impact of disease and trait anxiety are important, since trait anxiety is slightly more correlated, it comes out significant and impact of disease does not. In any case, trait anxiety account for 36.5 percent of the variance and the correlation indicates that fathers with high trait anxiety scores tending to use this coping strategy more than fathers with low trait anxiety scores.

Table 17

Father's coping by maintaining family integration, cooperation, and an optimistic definition of the situation predicted by physical and other father scores

Variable	r	Cumulative R square	R square chg.	<u>F</u>
Nutritional Status	.22	.049	.049	< 1
FEV 1	.12	.069	.019	< 1
Trait Anxiety	.57	.434	.365	3.58 *
Total F.F.I.	.04	.478	.044	2.98
Total Impact	-.57	.524	.046	2.64
State Anxiety	.50	.526	.002	2.03
Depression	.47	.527	.001	1.59

* $p < .05$

2. Coping by Maintain Social Support, Self-esteem, and Psychological Stability

The mothers' score on this coping strategy was found to be most associated with the physical score on FEV 1, and the mothers scores on the impact of disease on family scale and state anxiety. The results of the regression are summarized in Table 18 which shows that the FEV 1 accounts for 12.2 percent of the variance, impact on family scale an additional 20.1 percent of variance, and the state anxiety score another 7.6 percent of the variance ($F(1,20)=2.74, p<.05$).

insert Table 18 about here

The depression scale did not account for a significant additional proportion of variance ($F(1,19)=1.11, n.s.$). Mothers' scores on this coping strategy were found to correlate with higher scores on FEV 1 and state anxiety, and low scores on the impact of disease on family (which is interpreted as indicating the disease has a detrimental impact on the family).

The fathers' regression results are presented in Table 19.

insert Table 19 about here

Although both the total score on the impact of disease on family scale and state anxiety were predictive of this coping strategy, the state anxiety score did not account for significant variance

Table 18

Mother's coping by maintaining social support, self-esteem, and psychological stability predicted by physical and other mother scores

Variable	r	Cumulative R square	R square chg.	F
FEV 1	.34	.122	.122	3.48 *
Nutritional Status	-.25	.139	.017	1.94
Total Impact	-.36	.340	.201	3.95 *
State Anxiety	.35	.416	.075	3.92 *
Depression	-.06	.469	.053	3.72 *
Trait Anxiety	.40	.493	.024	3.25 *
Total F.F.I.	-.15	.502	.009	2.75 *

* $p < .05$

** $p < .01$

Table 19

Father's coping by maintaining social support, self-esteem, and psychological stability predicted by physical and other father scores

Variable	r	Cumulative R square	R square chg.	F
Nutritional Status	.18	.031	.031	< 1
FEV 1	.14	.033	.002	< 1
Total Impact	-.63	.555	.522	5.84 **
State Anxiety	.48	.579	.024	4.48 *
Total F.F.I.	-.25	.591	.012	3.46 *
Trait Anxiety	.58	.595	.004	2.69
Depression	.46	----	----	----

* $p < .05$

** $p < .01$

($F(1,13) < 1$). The impact on family score accounted for an additional 52.2 percent of the variance after the two disease variable had been entered into the regression. A negative correlation was also found for the fathers' score which indicates higher support scores associated with high impact of disease on the family.

3. Coping by understanding the medical situation through communication with others, parents and consultation with medical staff

The regression on the mothers' scores on this coping strategy was not found to be predicted by either the physical scores or the remaining mothers' scores. However, the fathers' scores for this coping strategy was again most predicted by the score on the total impact of disease on family score. As seen in Table 20, the impact of disease on family score accounted for 49.9 percent of the variance after the accounting for the disease variables.

insert Table 20 about here

Although the remaining scores are also significant predictors with the impact of family score, they do not account for further significant variance. Since the F-value obtained on the increment of variance due to Family Functioning Index was not significant ($F(1,13)=2.64$, $p>.05$), the remaining three variable, trait and state anxiety, and depression, are by default, also not significant.

Table 20

Father's coping by understanding situation through communication with other parents and consultation with medical staff predicted by physical and fathers' other scores

Variable	r	Cumulative R square	R square chg.	<u>F</u>
Nutritional Status	-.24	.058	.058	< 1
FEV 1	.16	.090	.032	< 1
Total Impact	-.66	.589	.499	6.71 **
Total F.F.I.	-.42	.648	.058	5.98 **
Trait Anxiety	.65	.682	.344	5.17 **
State Anxiety	.28	.714	.032	4.58 *
Depression	.36	.718	.004	3.64 *

* $p < .05$

** $p < .01$

4. Parental Care

The results of the regression of maternal care is presented in Table 21. The disease variables accounted for only 8.6

insert Table 21 about here

percent of the variance. When the total impact of disease on family score was added into the regression, an additional 17.6 percent of the variance was accounted. But, the regression was not significant until trait anxiety was also entered incrementing the variance by an additional 10.7 percent of the variance. It would appear that perhaps the reason for the significant finding was not due to accounted variance, but rather, due to increase of the number of predictor variables. This result should be cautiously interpreted. For the paternal care score, the disease variables accounted for 5.1 percent of the variance, and the father and family variables accounted for an additional 10.3 percent of the variance and neither were significant.

5. Parental Overprotection

Opposite findings were obtained for the mothers' and fathers' scores on the overprotection. As seen in Table 22, although all the variables are predictive in the regression, only the physical measures account for significant proportion of variance.

insert Table 22 about here

Table 21

Maternal care predicted by physical and other mother scores

Variable	r	Cumulative R square	R square chg.	<u>F</u>
FEV 1	-.20	.041	.041	1.07
Nutritional Status	-.12	.086	.045	1.13
Total Impact	-.34	.262	.176	2.72
Trait Anxiety	-.17	.396	.107	3.22 *
Depression	.09	.428	.059	3.15 *
Total F.F.I.	.06	.447	.019	2.69 *
State Anxiety	-.13	.448	.001	2.20

* $p < .05$

Table 22

Maternal overprotection predicted by physical and other mother scores

Variable	r	Cumulative R square	R square chg.	<u>F</u>
FEV 1	.49	.246	.246	8.16 *
Nutritional Status	.31	.538	.292	14.02 **
Trait Anxiety	.35	.651	.113	14.33 **
Depression	.38	.662	.011	10.78 **
State Anxiety	.05	.666	.004	8.40 **
Total F.F.I.	-.03	.668	.002	6.72 **
Total Impact	-.27	.669	.001	5.49 **

* $p < .05$ ** $p < .01$

FEV1 accounted for 24.6 percent of the variance and nutritional status another 29.2 percent of the variance. Maternal trait anxiety contributed an addition 11.3 percent of the variance which was also significant ($F(1,21)=6.79$, $p<.05$). The Pearson correlation for the two physical scores are somewhat opposing with better scores on the FEV 1 correlated with higher scores on overprotection but worse nutritional status correlated with higher overprotection. The simple correlation for trait anxiety indicated higher overprotection scores associated with higher trait anxiety scores.

The results of the regression in Table 23 clearly shows that paternal overprotection is not associated with the disease variables but with measures of family functioning and the psychological well being of the father.

insert Table 23 about here

The Family Functioning Index accounted for 39.2 percent of the variance and an additional 33.8 percent of the variance was accounted for by the father's score on the depression scale. State anxiety does not significantly increment the proportion of variance ($F(1,13) < 1$ n.s.). Worse family functioning as indicated by low scores on the family functioning index was correlated with higher scores on paternal overprotection and higher depression scores were also correlated with higher scores on paternal overprotection.

Table 23

Paternal overprotection predicted by physical and other father scores

Variable	r	Cumulative R square	R square chg.	F
Nutritional Status	.16	.024	.024	< 1
FEV 1	-.04	.024	.0006	< 1
Total F.F.I.	-.61	.416	.392	3.34 *
Depression	.56	.755	.338	10.02 **
State Anxiety	.23	.765	.010	7.83 **
Trait Anxiety	.62	.775	.010	6.33 **
Total Impact	-.55	.779	.004	5.05 **

* $p < .05$ ** $p < .01$

Synopsis of Results

In the first section of the analyses, multiple regression were employed separately for the mother and the father to determine the relative importance of disease, parental, and family variables on the child's psychological functioning. The children's number of behavioral problems were found to be associated with maternal trait anxiety and overprotection. Self-worth based upon athletic competence was associated with nutritional status and maternal coping by understanding the medical situation; whereas, self-worth based upon physical appearance was associated with maternal ratings of the impact of disease of family. The child's social competence was found to be predicted by nutritional status.

The second section of the results analysed parental use of coping patterns, care, and overprotection, predicted by disease variables, family functioning, depression, anxiety, and impact of disease on family functioning. Father's coping by maintaining family integration was predicted by his trait anxiety; coping by maintaining social support and coping by understanding the medical situation were both associated with the impact of disease on family functioning. The mother's use of coping by maintaining social support was found to be associated with both FEV 1 and the impact of disease on the family. Although maternal care was found to be predicted by trait anxiety, this was probably a chance finding. Differences were noted between maternal and paternal overprotection as maternal overprotection was associated

with the disease variables (FEV 1 and nutritional status) and maternal trait anxiety; whereas, the father's score on overprotection was not predicted by the disease variables but associated with only his ratings on family functioning and depression.

Some statistical problems were noted in the second section of the results. There was high multicollinearity in the predicting variables which influenced the choice of predictor variables. Some of R-squares were noted to be unusually high due to the combination of multicollinearity and suppressor effects. It is suggested that these results be cautiously interpreted.

Discussion

Approximately 10 percent of children have a chronic disease. Epidemiological studies have demonstrated that 17 to 27 percent of children with a chronic disease compared to 7 percent of healthy children display significant psychosocial and psychological problems (Rutter et al., 1970; Pless & Roghmann, 1971). However, the factors that are associated with the child's psychological functioning have not been established. Family, parental, and disease related factors appear to be related to the child's psychological functioning, but the relative importance of these variables is unknown.

The purpose of the present study was two fold. The primary interest of this study was to examine the relative importance of parental, family, and disease variables in the psychological functioning of children with cystic fibrosis. The data analysis was conducted in a hierarchical order by accounting for the effects of the disease variables before considering the psychological variables. It was expected that family and parental variables would account for significant additional variance in the child's psychological functioning after taking into consideration the variance due to the disease variables.

The secondary purpose was to study which parental, family, and disease variables were associated with the parents' ability to cope with disease and provide appropriate care to the child. This has been a neglected field and no hypothesis was generated. The analysis were expected to provide some understanding of the relationship and suggest further study.

Thirty six children with cystic fibrosis, 29 mothers and 20 fathers participated in the study. The children were asked to complete questionnaires of depression, anxiety, and self-worth. Parents completed depression, anxiety, overprotection, coping, family functioning, and impact of disease on family questionnaires. In addition, mothers also completed a child behavior checklist. None of the scores obtained in this study were pathological. However, it was interesting to note that children whose parents did not participate in the study scored higher on the depression and anxiety scales and lower on the social acceptance scale. A number of disease variables were extracted from the patient's chart. Each set of variables were factor analysed as a method to reduce the number of variables and multiple regressions were subsequently employed to examine the relationships between the outcome variables, child's psychological functioning, parents' coping styles, parents' care and overprotection, and the predictor variables which include the parental and family variables and disease status variables. Each analysis was conducted separately for the mother and the father. In view of the small number of fathers who participated in the study, their results should be cautiously interpreted.

The Association Between Child, Family, Parents, and Disease

Deciding which psychological or psychosocial measures should be used as the most appropriate outcome measures has often limited the usefulness of many studies and delayed the implications of findings in clinical practices (Pless, 1983).

The results of this study clearly indicate that at least three measures of the child's psychological and psychosocial functioning are required: 1. a measure of behavioral problems; 2. a measure of social competence; and 3. a measure of self-esteem based upon athletic competence and physical appearance or body image. The factors that are associated with these measures were found to vary.

Although it was hypothesized that the familial and parental variables significantly increment accounted variance, this was true for only the number of behavioral problems and physical appearance. Maternal anxiety and overprotection can significantly predict the number of behavioral problems displayed by the child as perceived by the mother. Overprotection has been repeatedly demonstrated to be a contributing factor in the maladjustment of children with chronic disease (ie. Allen & Pearson, 1928; to, Sabbeth, 1984). Maternal anxiety has also been often noted. However, the direction of the association is unclear; that is, children who have behavioral problems may well cause their mothers to be more anxious, or, mothers who are abnormally anxious may cause children to behave abnormally. What is of importance, is however, children's behavior is associated with maternal variables and is independent of physical disease. The opposite is true for social competence.

The measure of social competency analysed in this study was an aggregate of the child's quantity and quality of social activities in three areas: school (ie. school performance); general activities (ie. participation in sports); and, general

social (ie. number of friends) (Achenbach & Edelbrock, 1983). Based upon these measures, the child's social competency was not influenced by parental or family factors although one might expect that overprotected children would be socially isolated. Rather, the disease factors appear to be the most responsible in the ability of the child to perform in social activities. The debilitating force of the disease strongly influences social competency. As severity increases both in terms of pulmonary and anthropometric effects, so does the child's participation in social tasks. Whether the disease influences the child with a chronic disease social skills was not addressed in this study. Cadman, Boyle, Szalmari, & Offord (1985) reported that children with a chronic disease are more isolated than healthy children but they are not socially inept.

Similar to the previous findings, the results on the two self-esteem measures were predicted by either the physical or parental/family variables. Athletic competence was found associated with disease. Those children with worse disease were less athletically competent. However, children whose mothers relied on understanding of the medical situation as a coping pattern, tended to have children who scored higher on athletic competency. One might conjecture that these children were allowed to be more active in sports since the mothers better understood the disease and its limitation on the child's body and knew that children with CF can be physically active.

Finally, self-esteem based upon physical appearance was

not correlated with any of the physical measures but rather with maternal ratings of the total impact of disease. The measures obtained in the Impact on Family Scale provide information on how the child and the disease affect the family (Stein & Riessman, 1980). Children, whose mothers report that the disease does not have a detrimental effect on the family, have a higher self-image of their physical appearance.

Previous studies have reported various associations between disease variables, particularly the severity of the disease, and the child's psychological functioning. Some have stated that as severity of disease increases and prognosis worsens, there is a greater association with maladjustment (ie. Pless & Roghmann, 1971; Bywater, 1981); whereas, others have reported that maladjustment is most associated with moderate severity (ie. Bruhn et al., 1971). The results of this study indicate that a linear relationship does exist between the severity of the disease but only with some aspects of psychological functioning - social competence and athletic competence. No linear association was noted on the number of behavioral problems and disease severity. However, the nature of the analysis conducted in this study (ie. linear regressions) does not totally refute the marginality hypothesis. It is clear that the marginality hypothesis is not an appropriate alternative in the analysis of social and athletic competence but further research would be required to determine whether the hypothesis holds true for behavioral problems and self-esteem in children with a chronic disease.

No evidence was found to support the importance of family functioning as a factor associated with the child's psychological functioning. This is somewhat disturbing since the studies conducted by Pless et al., (1972), Lewis & Khaw (1982), and Cowen et al. (1986), all well designed studies using a large sample size have found significant correlations between family functioning and the child's psychological functioning. Two possibilities might explain why family functioning was not found to be associated with the child's psychological functioning. The Family Functioning Index has not been extensively validated and perhaps the information obtained could not accurately reflect family functioning. In future studies of families with children having a chronic disease, it is recommended that better validated measures of family functioning be employed such as the "Family Adaptability and Cohesion Evaluation Scales" (Olson, Bell, & Portner 1978). Alternatively, the Family Functioning Index is a tool which assesses various aspects of the family (ie. communication, child orientation) independent of the effects of the disease. However, when the disease variable is also taken into consideration such as in the "Impact on Family Scale", a more sensitive measure is obtained. Therefore, the effect of the disease itself on certain aspects of family functioning and the effect of parental characteristics such as depression or anxiety simply play a greater role in the interaction of family and child than does general family functioning alone.

Association between Parents Coping, Care, Overprotection, Disease Variables, Family Functioning, and Other Parental Variables

I: Parental Coping

Parental coping is one of the most neglected areas of research in chronic disease. How parents with children with chronic disease cope with the stress of child, social, and disease management has only recently been an area of investigation (McCubbin & Patterson, 1985).

McCubbin et al. (1983) reported significant correlations between the three coping patterns (maintaining family integration, maintaining social support, and, understanding the medical situation) and family functioning for both mothers and fathers. Specifically, mothers' scores on these coping patterns correlated with family cohesion and expressiveness and fathers' scores correlated with family organization, cohesion, and conflict. However, much like the findings in the primary analyses, the results of this study indicate that the association between the coping patterns and general family functioning is not significant. Rather, when family functioning is assessed by taking into consideration the disease such as the measure obtained in the Impact on Disease on family scale, then an association is found between the coping patterns and family functioning.

For two of the coping patterns (maintaining social support and understanding the medical situation) a strong association was

found between the impact of the disease on family and the coping patterns. Fathers higher use of the two coping strategies was associated with less impact of the disease on family functioning. Lower scores on the mother's report of the impact of disease was related only to the second coping pattern (coping by maintaining social support).

The disease factors were not as strongly associated with the coping patterns as would be expected (McCubbin et al., 1983). The association between pulmonary function and the second coping pattern was noted in the mother's results but nutritional status had little effect on coping. No other associations between coping patterns and disease variables were found. McCubbin et al. (1983) had reported that the mother's score on the second coping pattern (maintaining social support) was correlated with pulmonary functioning and the father's score correlated with pulmonary functioning and nutritional status. Mother's score on the first coping pattern (maintaining family integration) was correlated with nutritional status (McCubbin et al., 1983). What appears to have happened in this study, is when variables other than disease related variables are also taken into consideration, the effects of the disease variables on the coping strategies is limited.

A major pattern can be seen in the results of parental coping. Mothers' coping strategies with the exception of the second coping pattern were found not to be associated with any of the disease, family, or remaining maternal factors. This strongly suggests that there are factors which are important in

maternal coping with chronic disease which have not been assessed in this study. Subsequent study should include sociodemographic variables such as family income, sex of child, age of child, parental education, number of siblings with and without cystic fibrosis as possible factors that might be associated with coping.

II: Care and Overprotection

Although maternal care was found to be associated with maternal anxiety, this finding was probably due to statistical manipulation (ie. entry of another predictor variable). Any interpretation of this finding may be limited. This is however, not the case with the measure of parental overprotection.

Differences were noted between maternal and paternal overprotection. Mother's overprotection was found to be associated with the disease variables whereas, father's overprotection was correlated with family functioning and father's depression. This finding does make some clinical intuitive sense. Mothers as primary care-givers have the responsibility of bringing the child to hospital for the regular clinic visit. They are, therefore, much more aware of the child's disease status and any changes that have occurred. With cystic fibrosis, changes in disease almost always reflect deterioration. It is therefore not surprising that the natural tendency of the mother is to hover over the child or adolescent knowing that the child's future is uncertain. The mothers'

overprotective behaviors may reflect her attempt to ensure that the child is compliant with medication and nutritional needs. Fathers however, do not play such a significant role in physical care of the child. Therefore, it is less likely that the child's physical well-being will influence their overprotective attitudes and behaviors.

III: General Discussion of the Secondary Analysis

The purpose of the analysis of the coping and overprotection measures was to suggest further research. The individual variation in coping strategies for mothers indicates the need to further evaluate what methods mothers find effective in coping with the stress of raising a child with chronic disease and what factors influence the mother's use of the coping strategies.

It would also be of considerable interest to examine the effects that the child's age and sex have on the parents' coping strategies and overprotection. Although not statistically significant, in all analyses, the father's scores on coping and overprotection were associated with the nutritional status of the child rather than the pulmonary functions. It would be appear that the child's physical appearance is somehow more important with the father's psychological functioning than the mothers. It would be of interest to examine whether the sex of child (ie. boys) is associated with how the fathers cope.

Implications of the Present Study

Two factors preclude making unequivocal implications. A study utilizing a larger sample size must be conducted to replicate the findings of this study. Use of a small sample size would have: 1. increased the likelihood of obtaining a significant regression and accounting for a large proportion of variance; and 2. would limit the generalizability of the results to other cystic fibrosis patients and families in other clinics. However, since the sample composition of this study was the population of cystic fibrosis patients in the Eastern Ontario area, the findings are representative for this whole group. Secondly, the measures obtained in this study were self ratings or ratings of parental perception. A recurrent comment made in the chronic disease literature is that parents and children often deny many of their psychological and psychosocial problems. It is not possible to assess the validity of the responses parents or child provided since other methods for collecting data were not employed (ie. collecting observational data, interview data, and/or, data from a third person such as the child's teacher).

There are, however, results that do clarify a number of previous problems and warrant further investigation. Whereas, a number of previous studies in the assessment of children with chronic diseases tended to address the child's psychological functioning as a global term, this study has clearly indicated that the child's psychological functioning should be perceived and studied in specific areas: behavioral, social, and self-esteem.

The factors that are associated with the child's behavioral, social and self-esteem scores vary. Familial and parent factors tend to be associated with the behavioral and self-esteem based upon physical appearance. Whereas, the disease factors are most associated with social functioning self-esteem based upon athletic competence. These findings provide some information on what factors should be taken into consideration during a design of a future clinical study. If the study is to be directed towards behavioral problems, the clinical researcher must take into consideration both the child and the parents, particularly the mother. However, if the study is to address the social problems that children with chronic disease often face, then the clinician must consider the role of disease severity as a factor that might affect outcome.

Since this study included only patients with cystic fibrosis, these implications might be limited to only this disease group. Whether they are true for other disease groups must be further investigated. It is the opinion of this author (a non-categorical supporter), regardless of disease, similar results will be obtained.

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