THE PSYCHOLOGICAL IMPACT OF SKIN DISORDERS ON CHILDREN AND THEIR FAMILIES

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Thesis submitted for the degree of Doctor of Philosophy

University College London

2001

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ABSTRACT

This thesis examined the impact of two different skin disorders on children and their families.

The first skin disorder was the rare skin condition, Epidermolysis Bullosa (EB). This was studied using both quantitative and qualitative methodology. In the quantitative study, the relationship between the family environment, the severity of the condition and the psychological adjustment of the child and his or her parents was evaluated. The study showed that there was no correlation between the severity of the EB and the child's psychological adjustment. Although the results showed some evidence of a relationship between the severity of the illness and the psychological impact on the fathers, no relationship was found for the mothers. Families that were cohesive and had lower levels of conflict had children with significantly better psychological adjustment. In the qualitative study, the experiences of mothers of children with EB were described. Four central themes were identified as important features of the mothers' experiences and these are discussed in relation to existing research.

The second disorder studied was the common condition of childhood eczema. The relationship between the severity of eczema, the mother's resolution of the child's condition, and the psychological adjustment of both the mother and the child was examined. A standardised interview was used to assess the mother's resolution of the child's eczema. Mothers who were resolved about the child's illness reported lower levels of parenting stress and psychological distress than mothers who were unresolved.

The study showed that mothers caring for a child with eczema reported higher levels of parenting stress than a comparison group of mothers of children with other chronic illnesses. No differences in levels of psychological distress in the child or his or her mother were found between the children with eczema and the comparison group.

THE PSYCHOLOGICAL IMPACT OF SKIN DISORDERS ON CHILDREN AND THEIR FAMILIES

CONTENTS

LIST OF TABLES

LIST OF TABLES (continued)

LIST OF FIGURES

ACKNOWLEDGEMENTS

The research on Epidermolysis Bullosa (EB) was supported by the Dystrophic Epidermolysis Bullosa Research Association (DEBRA) and I would like to thank DEBRA for both the financial support and the help in contacting families with EB. I would also like to thank Dr David Atherton and all the members of the EB clinics at Great Ormond Street for their help with the EB study. I am grateful to Dr Sian Critchley-Robins for help with coding the interviews with the children with EB. Professor Christine Eiser and Dr Richard Lansdown provided very useful advice and I am grateful for their help with the design of the study.

I am grateful to Dr Catherine Smith for her support in setting up the eczema study and would like to thank Sarah Davies and all the staff in the paediatric out-patient department at University Hospital Lewisham for their help recruiting the eczema and comparison groups. I would also like to thank Dr David Atherton and the staff at St John's Institute of Dermatology who helped to recruit participants at St Thomas' Hospital. I am particularly grateful to Lesley Fairplay who completed the severity measures for the children with eczema and to Dr Caroline Campbell who rated all the interviews with the mothers of children with eczema.

I would like to thank my husband Dr Stephen Scott for his encouragement and support, and for tolerating my demands for child-free time to enable me to complete this thesis.

Dr Chris Barker has been an exemplary supervisor, who has encouraged me to persist with this research and provided a lot of constructive criticism along the way. I

would particularly like to thank him for all the time he has put into supervision, and for all that I have learned from him about research over the years.

Finally, I am grateful to all the families who participated in the research and I would like to thank all the parents and children who were willing to talk to me about their experience of skin disorder. I hope that my respect for the way families adapt to having a child with a skin disorder is conveyed in this thesis.

CHAPTER ONE

INTRODUCTION

Treatments for physical illness in childhood have improved considerably in the past fifty years (Garrison & McQuiston, 1989). Far fewer children die from acute infectious diseases and the outcomes for many chronic illnesses have also improved (Gortmaker & Sappenfield, 1984). However, there are many forms of chronic illness that affect children for which there is no cure as yet. Families with a child with a chronic illness have to manage the symptoms of the child's condition and long term treatment regimes, in addition to the usual demands of bringing up a child. This involves increased exposure to physical and psychological stress over the long term, which is likely to have some psychological impact on both the affected child and his or her family (Hobbs and Perrin, 1985).

Chronic illness affects approximately 10 % of all children, although the figure varies from 5 – 20% depending on the definition of chronic illness that is used (Bradford, 1997). Eiser (1990) defined chronic diseases as:

> "conditions that affect children for extended periods of time, often for life. These diseases can be "managed" to the extent that a degree of pain control or reduction in attacks, bleeding episodes or seizures can generally be achieved. However, they cannot be cured." (p 3)

However, other definitions vary with respect to how severe a condition has to be to count as a disease or illness, or exactly how long the symptoms have to be present to count as chronic. These differences between definitions give rise to slightly different prevalence figures.

Skin disorders, particularly eczema, are very common during childhood (McHenry, Williams & Bingham, 1995). However, despite a large number of studies of children with chronic illnesses, skin disorders have not been extensively studied. For example, in the meta-analysis carried out by Lavigne and Faier-Routman (1992) of all existing studies on the impact of chronic illness on children and families, no studies on skin conditions were identified. Even since then, few studies have been published on the impact of skin disorders on children and families.

This chapter will begin with a review of the literature on the impact of chronic illness on children and families. Then, the main theoretical models describing the impact of chronic illness will be reviewed. Finally, the literature on the impact of skin disorders on children and families will be summarised.

Review of Studies on the Impact of Illness on Children and Families

There have been a very large number of empirical studies on the psychological impact of chronic illness on children and their families over the past twenty-five years. Prior to this, most research in this area was based on individual clinical reports or theories that illustrated some of the difficulties faced by children with a chronic illness (Freud, 1952; Solnit & Stark, 1961). However, since these were based on selected clinical cases it was not possible to know whether these descriptions applied to all children with a chronic illness or just clinically referred cases. The initial focus of the recent empirical studies was therefore to determine whether or not children with chronic illness were more likely to develop psychological difficulties than healthy children. Since each study focused on a small local sample of children with one particular illness, the findings were often contradictory or inconclusive and it was difficult to compare across studies because of the different measures used and differences in methodology. However, there are now good epidemiological studies and meta-analyses that have answered this initial question and the findings from these will be reviewed below.

Epidemiological Studies on the Impact of Chronic Illness on the Child

Rutter, Tizard and Whitmore's (1970) large epidemiological study based on the Isle of Wight demonstrated higher rates of psychological disorder among children aged 10 – 12 years with chronic physical handicap, and found that children with conditions that affect the brain and central nervous system were at even higher risk.

Cadman, Boyle, Szatmari and Offord's (1987) report on the Ontario Health Study concluded that the rate of psychiatric disorders among children with chronic illness and associated disability was three times the rate amongst healthy children. The rate among children with chronic illness but no associated disability was somewhat lower - twice the rate for healthy children. This study was based on a large community sample and involved interviews with 1869 families including 3294 children aged 4-16, and diagnoses based on DSM-III (APA, 1990) were used.

A recent large epidemiological study carried out by Meltzer, Gatward, Goodman and Ford (2000) in the U.K. that used ICD-10 (WHO, 1993) criteria for diagnosis, demonstrated that having any physical illness (compared to no physical health condition) increased the odds of having a "mental disorder" by 82%. However, this study also demonstrated very different rates of psychological disorders for different types of physical disorder. For example, 37% of children with epilepsy and 35% of children with co-ordination problems had mental disorders, compared to only 12% of children with heart problems and 12% of children with diabetes. This compares with 10 % of healthy children. This study supports the earlier studies demonstrating higher risks for children with neurological problems (Howe, Feinstein, Reiss, Molock & Berger, 1993; Rutter, Tizard & Whitmore, 1970).

Clinic Studies on the Impact of Chronic Illness on the Child

These epidemiological studies are the exception rather than the rule. Most of the studies reported in the literature are based on clinic samples. Some of these studies found no evidence of increased psychological difficulties among children with a chronic illness (Spaulding & Morgan, 1986; Noll et al., 1994), whereas others found substantial differences between chronically ill and healthy children (Ungerer, Horgan, Chaitow & Champion, 1988). The conflicting findings from clinic studies on different illnesses can partly be explained by the characteristics of the samples studied and the different outcome measures used. In addition, many sample sizes have been too small to have the power to detect anything other than large effect sizes. However, when the evidence from methodologically sound studies are evaluated as a whole, the clinic studies also show that children with a chronic illness are at increased risk of developing psychological difficulties.

Lavigne and Faier-Routman (1992) conducted a meta-analysis of 87 studies evaluating psychological adjustment to chronic illness in childhood. Their overall conclusion was that there is strong evidence that children with chronic physical illness are at increased risk of psychological adjustment problems compared to healthy children (mean effect size 0.62). They also showed that children are at greater risk of internalising disorders than externalising disorder (mean effect sizes 0.55 and 0.26 respectively) although the difference between these two rates was only significant when teachers, as opposed to parents, were the informants. In addition, children with physical illnesses had lower self-esteem or self-concept scores than healthy children (mean effect size 0.31) although this effect size reduced with careful matching of controls on SES, age and ethnic background. The methodology used by each individual study affected the reported size of effect. Studies using comparisons to norms and careful control group matching reported larger effect sizes than studies with a control groups who were matched on only one or two variables. Lavigne and Faier-Routman (1992) found that there was considerable variation in effect sizes even within one disease category, as well as across disease categories. These differences can partly be explained by methodological differences between studies, but they conclude that there may also be important differences between the different types of illnesses in terms of psychological impact.

Bennett (1994) carried out a meta-analysis of 60 studies of depression among children with chronic medical problems. He concluded that children with chronic illness are at increased risk of developing depressive symptoms (mean effect size 0.27) and at increased risk of major depressive disorder compared to healthy children. He also showed considerable variation in effect sizes across disease groups. For example, the mean effect size for children with diabetes was 0.22, whereas the mean effect size for asthma was 0.54, and there was actually no difference between children with cancer and controls. Given the small numbers of studies on any particular disease group, these findings have to be interpreted cautiously, but do add to the evidence that there are important differences between the psychological impact of different illnesses.

Impact of Chronic Illness In a Child on the Family

The studies of the impact of having a child with a chronic illness on parents have followed the same pattern as those on the children themselves. Conflicting findings emerged from various different clinic studies, with some studies reporting similar rates of distress between mothers of children with a chronic physical condition and mothers of healthy children (Spaulding & Morgan, 1986), and others reporting significant differences (Kupst, 1992). Few studies have included both fathers and mothers but those that have done so have documented differences in the impact of the illness on the two parents (Mastroyannopoulou, Stallard, Lewis & Lenton, 1997; Sloper, 2000; Timko, Stovel & Moos, 1992). Different risk factors seem to be relevant for mothers and fathers, e.g. Sloper (2000) found that employment status was an important predictor of distress in fathers, but this was not a predictor for levels of distress in mothers. Whilst it is still true that the major burden of care of a chronically ill child falls on the mother, the supportive contribution of the father is an important aspect of family functioning (Sloper & Turner, 1993). Overall, the studies show that there is considerable variation in parents' psychological response to having a child with a chronic illness, and that although there is an overall higher rate of distress compared to families of healthy children, only a minority of parents develop psychological disorders.

Drotar (1997) reviewed studies on the relationship between family functioning and psychological adjustment of children with chronic health conditions. He showed that measures of family or parental functioning that reflected supportive family relationships (e.g. cohesion) predicted fewer behavioural symptoms and more competent psychological functioning (e.g. self esteem) whereas measures of problematic family qualities (e.g. conflict) generally predicted less competent psychological adjustment and/or higher levels of behavioural symptoms. The amount of variance in child adjustment predicted by family measures was between 10% and 15%. Drotar warns that since only a very small proportion of the studies reviewed were prospective in design, it is possible that these findings reflect the influence of child behavioural difficulties on family functioning, rather than the other way round, as they are usually interpreted. In addition, many studies relied on the mother as the sole informant on both the family measures and the child adjustment measures, so there is a possibility that at least at some of the association found is due to shared method variance.

Drotar (1997) concludes that it is important that future studies test explicit models of how family processes influence the psychological development of children with chronic health conditions with particular reference to the course of the illness itself and the developmental stage of the child. He also highlights the lack of intervention studies that test the possibility of modifying family influences in order to prevent or reduce the child's adjustment difficulties.

Several authors have pointed out that research about the impact of chronic illness has focused heavily on the child and mother, with few studies on the impact on siblings (Eiser, 1993). This gives a very limited view of the impact of chronic illness on family life as a whole. There is a general consensus that the consequences of having a brother or sister with a chronic illness can lead to some additional difficulties for siblings. For example, the well sibling may feel they receive less attention than the ill child and may experience higher levels of distress in their parents. Cadman et al.'s (1987) study showed that siblings of a chronically ill child were at higher risk of developing emotional problems than siblings of healthy children. However there was no difference between the two groups in the rates of behavioural problems or other indices of functioning, such as attending leisure activities or school. Some authors suggest there may also be some positive influences on siblings. For example, a study by Horowitz and Kazak (1990) showed no significant differences between a group of siblings of children with a chronic illness and another group of siblings in terms of psychological adjustment. However, the children with a sibling with a chronic illness were rated higher on measures of helping, praising, showing affection, giving gifts and sharing. As with the studies on children or parents, the focus of studies now needs to shift to understanding why some siblings are more at risk of developing difficulties than others.

Methodological Issues

Taken together, the results of the epidemiological studies and individual clinic studies do demonstrate that children with a chronic illness and their parents are more likely to develop psychological problems than are healthy children and their parents. However, there are some important methodological difficulties with these studies, and some of the main areas of controversy will be discussed below.

Measures of Adjustment

Most of the published studies on the psychological impact of chronic illness have used either psychiatric diagnoses or symptom based measures as their dependent variable e.g. DSM-IV (APA, 1994) or ICD 10 (WHO, 1993) or the Child Behaviour Checklist (CBCL: Achenbach & Edelbrock, 1983). However, the presence of a psychiatric disorder or high levels of psychological distress are very crude indicators of psychological functioning. Many authors have argued that a more sophisticated measure of psychological functioning should be used because the impact of an illness on a child or family may not be detected by symptom based measures (Eiser, 1993).

One of the reasons why deficit measures are used is due to the difficulty in defining or measuring psychological adaptation or well being. There have been attempts to define adjustment. For example, Wallander and Thompson (1995) define psychosocial adjustment in this way:

> "Good adjustment is reflected as behaviour that is age appropriate, normative and healthy and that follows a trajectory toward positive adult functioning." (p 125)

Stein and Jessop (1990) developed the Personal Adjustment and Role Skills Scale (PARS III) as a more positive measure of childhood functioning. It covers six domains: peer relations, dependency, hostility, productivity, anxiety/depression and withdrawal. Despite the criticism of the symptoms based measures these are still widely used and the broader measure of psychological adjustment have not been reported widely in the literature.

In addition, many studies do not take into account the variation in both the age of the child and the length of time since the diagnosis. For example, children across a wide age range are given the same measure of adjustment and the study does not take into account the differences due to how long the child has had the condition.

Alternatively, some studies have used quality of life measures, which tend to be specific to the particular illness studied (Titman, Smith & Graham, 1997). This makes it more difficult to compare findings across studies, but does provide a measure that is based on the impact of the condition on the individual concerned. Although there are now general quality of life measures for children (Graham, Stevenson & Flynn, 1997), these have not been widely used. Whilst there may be difficulties in using quality of life measures with young children, children from about the age of six can use these measures to give their own view of the impact of the illness on themselves.

The emphasis in research on psychological difficulties has been seen by many parents of children with chronic illnesses as misleading. It emphasises the difficulties families encounter rather than the resilience that many families show in difficult situations. As a result, there have been attempts to shift the focus and this is reflected in the terminology sometimes used. For example, Antonovsky (1993) argues that this new orientation could be described as "salutogenesis" - the study of the development of health as opposed to pathology. This has given rise to some new terminology e.g., "challenged by diabetes" as opposed to “diabetic” or “suffering from diabetes”. Part of the perceived need for this change is due to the focus on pathology on which many scientific reports of chronic illness are based.

There is considerable evidence that professionals predict more negative effects than families themselves do and the focus on deficit based measures may reflect some of this bias. For example, Hobart Davies, Noll, de Stefano, Bukowski and Kulkarni (1991) showed that professionals working with families of children with cancer predicted much greater differences between families of children with cancer and families of healthy children on the Child Rearing Practices Report (Block, 1980) than families of a chronically ill child did themselves. Walker, Garber and van Slyker (1995) found that parents of healthy children were more likely to describe misbehaviour of children with a chronic illness as less intentional, more excusable and due to more external reasons than misbehaviour of healthy children. This highlights the need to ensure that the measures used reflect the concerns of the affected families rather than the professionals carrying out the study or families with healthy children.

Self Report Measures

Most studies rely on self-report measures but there are limitations to using such measures. Self-report measures rely entirely on the respondent's own framework or viewpoint, and it has been suggested that mothers of healthy children use a different framework than mothers of ill children (Eiser, 1993). For example, the anchor points used by mothers of chronically ill children may be very different to those used by mothers of healthy children because the mother may already take into consideration the child's restrictions caused by the illness. For example, she may expect different levels of independence for her child with spina bifida and her healthy child, and hence fill in any self-report question about her child's independence accordingly. This may obscure differences if the only measure is based on the mother's report. Studies that have used multiple informants e.g. teachers and fathers as well as mothers, do provide a broader assessment and may identify some difficulties that are not apparent to one informant only.

Cramer (2000) criticises studies that rely on self report measures because of what we know about how unconscious defence processes and social factors influence responses to self report questionnaires. Although interview methods are more time consuming to carry out and more difficult to analyse they do allow for more exploration of the respondents concerns and for more sophisticated measurement of psychological processes.

Noncategorical vs Disease Specific Approaches

Stein and Jessop (1982, 1989) argued that the commonalities between the psychological impact of the different types of chronic illnesses outweigh any effects due to the specific illness itself and that it was therefore justifiable to combine groups of children with different chronic illnesses for research purposes. This has become known as the noncategorical approach and many studies have been based on this approach (Meijer, Sinnema, Bijstra, Mellenbergh & Wolters, 2000; Wallander, Varni, Babani, Banis & Wilcox, 1988). The original drive to combine illness groups was largely a pragmatic one, because there are very few children with any one type of illness in any particular centre, and by combining different illnesses it is possible to obtain larger sample sizes. In addition, at the time Stein and Jessop (1982) originally proposed the noncategorical approach, the focus was still on demonstrating whether there were differences between healthy and ill children at a very global level, whereas the focus has now changed.

Pless and Perrin (1985) proposed a modified noncategorical approach. They suggested that physical illnesses should be categorised according to features that are thought to have psychological importance rather than along the medical categories based on diagnosis. The five categories they proposed included the child's level of mobility, whether the illness was static or dynamic, the child's age when the disease process started, whether the child's cognitive and sensory functioning was affected and whether the condition was visible or not. Some of these features have been shown to be important in subsequent studies e.g. the effect of the condition on mobility and if the illness affects cognitive or sensory functioning. However, the other features have not been consistently supported across studies. This categorisation system is quite difficult to use in practice because some diseases have different combinations of these different factors.

A few authors have criticised the noncategorical approach and argued for a more contextual and in depth approach to understanding the specific difficulties associated with a particular illness (Quittner, DiGirolamo, Michel & Eigen, 1992; Thompson, Gustafon, Gil, Godfrey & Murphy, 1998). There is now considerable evidence from good epidemiological studies and from clinic studies that there is variation in psychological impact between different chronic illnesses (Frank et al., 1998; Meltzer, Gatward, Goodman & Ford, 2000). In addition, the evidence for the noncategorical approach was largely based on very global differences, such as differences in child psychological symptoms measured by the CBCL. There may well be no difference between diseases at this level, however, in terms of the impact at the level of psychological processes, there is a difference. Despite the popularity of the noncategorical approach, the recent evidence suggests that important differences in psychological impact may be obscured by combining different groups of illnesses.

Qualitative and Quantitative Methodology

The vast majority of studies on the impact of chronic illness on children and their families have used quantitative methods. However, within health psychology there has been increasing interest in the use of qualitative methodology (Murray & Chamberlain, 1999; Yardley, 1997). This originally developed out of the dissatisfaction with the assumptions of quantitative studies, particularly the positivist scientific basis of psychology, and a desire to take context and systems into account when studying psychological phenomena. However, there is now a general acceptance that qualitative and quantitative approaches are not competing methodologies but can be seen as complementary (Woolgar, 1996). Different methodologies are appropriate for different research questions and hence methodological pluralism has become more widely accepted (Barker, Pistrang & Elliott, 1994).

There are very few qualitative studies of the impact of chronic illness on children and families. Fiese and Bickham (1998) claim that this is partly because many paediatric psychologists feel ill-equipped to evaluate qualitative research in this field. Fiese and Bickham (1998) provide a checklist of important features of good quality qualitative research e.g. clear rationale for the study, adequate description of sample, clear description of procedures and understanding of the study's contribution to existing literature. These guidelines are very similar to those produced by other authors, for example Stiles (1993) and Elliott, Fischer and Rennie (1999).

Eiser and Twamley (1999) outline the ways in which qualitative methods can take the field forward by increasing our understanding of the experience of children with chronic illnesses. They argue that qualitative methods have often been avoided because of the inherent difficulties of interviewing a young child in a way that enables them to accurately represent his or her own viewpoint. This is a difficult task because of the power imbalance and the child's lack of familiarity with such methods, as well as the interviewer's ethical dilemma about how to manage potentially upsetting areas.

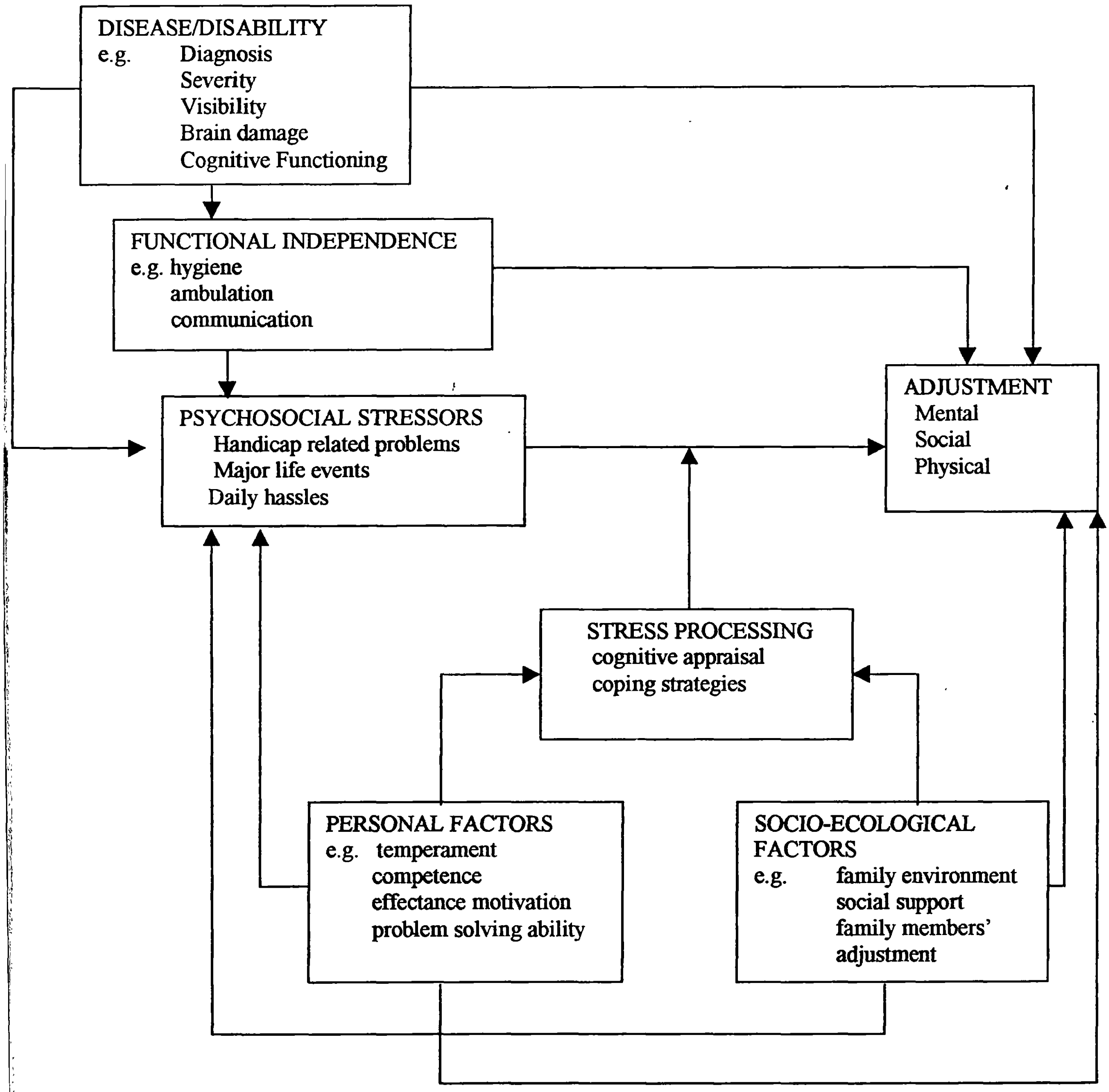
There are some examples of good qualitative studies in this field. Weiss, Marvin and Pianta (1997) used ethnographic methods to describe family strategies for child care of children with cerebral palsy. Abrams and Goodman (1998) studied how clinicians broke "bad news" to families. Sloper and Turner (1993) and Sloper (2000) used a combination of qualitative and quantitative methodology in their studies on the impact of disability on children and families. However, this methodology has not been widely adopted within paediatric psychology despite the need for the field to move towards greater understanding of psychological processes. Fiese and Bickham (1998) argue that "qualitative methods are best suited for asking questions that pertain to understanding the meaning that individuals or groups make out of experiences" (p 80). Qualitative methods are then appropriate both for hypothesis generation and for moving the field of research on beyond quantitative predictive studies to more detailed and in depth understanding of the psychological problems faced by children and families.

Models of Psychological Adaptation to Chronic Illness

The studies summarised above clearly show that only a minority of children with a chronic illness do develop psychological or mental health difficulties and the majority do not. The focus of research studies has moved on to trying to identify what processes account for the individual variation in response to illness and why some children and families are more at risk than others (Wallander and Varni, 1998). In response to the rather ad hoc development of this field Wallander (1992) argued that research should become more theory-driven and this statement has become very influential in directing further research. The main theoretical models that have been developed to try and explain variation in adjustment to chronic illness are described in the next section.

Wallander and Varni's (1998) model of adjustment has been the most widely researched within this field and was originally described in a slightly different format in Wallander, Varni, Babani, Banis and Wilcox (1989). The model is based on stress and coping theory (Lazarus & Folkman, 1984) and is shown in Figure 1.1.

Figure 1.1 Wallander and Varni's (1998) Model of Child and Maternal Adaptation to Chronic Illness



Disease or disability parameters, functional independence and psychosocial stressors are all seen as risk factors for psychological adaptation. Wallander and Varni (1998) hypothesise that these risk factors have both a direct effect on adaptation as well as an indirect effect. Intrapersonal factors, socio-ecological factors and stress processing are all seen as resistance factors. They too have both a direct and indirect effect on adaptation.

Various studies carried out by this group have tested aspects of the model. The model is very complex and because of this Wallander and Varni (1998) argue that the first step is to test specific predictions from one part of the model rather than attempting evaluation of the model as a whole. These studies have produced a mixture of findings (see Wallander and Varni, 1998 for review). For example, one study examined the effect of severity of limb damage (hypothesised as a risk factor) on psychological outcome in children with congenital or acquired limb damage. Whilst the severity of degree of limb loss was not associated with general self esteem in young children, it was for adolescents (Varni & Setogutchi, 1996). In another study using a prospective design, evaluating the link between psychosocial stressors and adjustment, Varni, Katz, Colegrove and Dolgin (1996) showed that mother's level of stress 6 months after receiving a diagnosis of cancer in her child did predict negative affect at 9 months. However, it was also true that negative affect at 6 months post diagnosis predicted perceived stress at 9 months, indicating that psychosocial stress cannot be seen solely as a cause of adjustment difficulties, but also as a consequence.

Whilst the model has been helpful in driving theoretical research and a more systematic evaluation of the impact of social and disease factors on adjustment to chronic illness, it has not been validated as yet by many independent studies (Bradford, 1997). No attempt has been made to evaluate the model as a whole. There has been some criticism of the way in which the various aspects of the model are theoretically related to each other and a lack of clarity in the formulation of the model. For example, why are some factors seen as "risk" factors and other as "resistance" and is there a theoretical difference between these two?

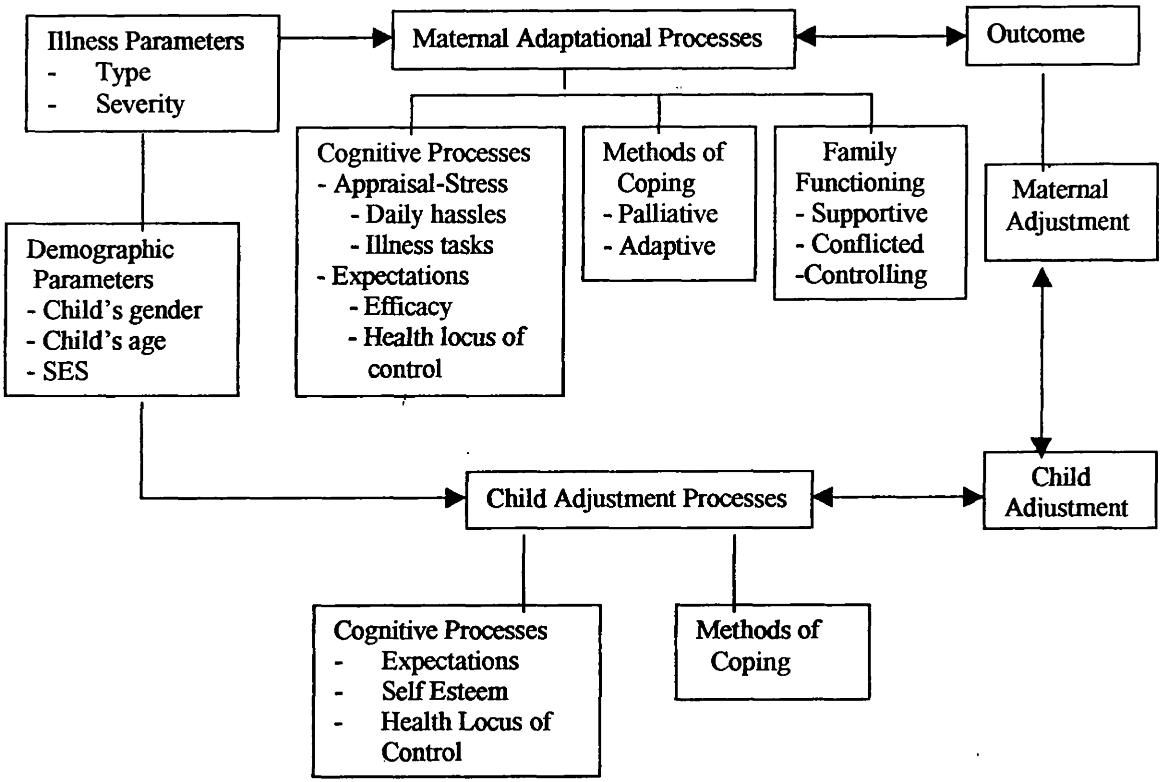
Wallander and Varni (1998) suggest that resistance variables "moderate" the relationship between risk factors and outcome. However, as Holmbeck (1997) points out, their use of the term "moderate" is somewhat confusing because this terms is usually used to describe an interaction (Baron & Kenny, 1986). In the usual use of "moderate", outcome would be worse for those who have high risk and a resistance attribute, compared to those with high risk and no resistance attribute. However, it would be no different for those at low risk whether or not they possess the resistance attribute. In fact, Wallander and Varni (1998) test whether or not these risk and resistance factors are significant predictors of the outcome variable, using hierarchical multiple regression, so the use of the term moderate is somewhat misleading.

Thompson, Gustafson, George and Spock (1994) describe a similar model, also based on the stress and coping model of Lazarus and Folkman (1984). They hypothesise that psychosocial processes influence the relationship between illness or demographic factors and psychological adjustment.

> "The focus of the model is on the contribution of adaptational processes hypothesised to influence the psychological adjustment of children and their mothers over and above the contributions of illness parameters of type and severity and demographic parameters of socio-economic status (SES) and patient age and sex." (p 190).

The model is shown in Figure 1.2.

Figure 1.2 Thompson et al. (1994) Transactional Model of Adjustment to Chronic Illness



Whilst the central features of this model include the same two basic cognitive processes of appraisal and methods of coping, Thompson et al. (1994) place more emphasis on appraisal than Wallander and Varni (1998). Thompson et al. (1994) have developed a measure of "illness appraisal", which is a brief interview to assess the mothers appraisal of stress and expectations of efficacy in managing stress in terms of four specific tasks related to illness. These are a) dealing with the child's medical problems and symptoms; b) maintaining the child's emotional well-being; c) maintaining their own emotional well-being and d) preparing for an uncertain future.

Thompson et al. (1994) adopt a disease specific approach and have confined their studies to specific illnesses and, in support of the disease specific model, have shown some differences between conditions. Their studies have also used interview measures rather than self report measures and multiple informants in order to improve on the methodological problems of earlier studies.

In a series of studies testing their model, Thompson and colleagues have found some support for their hypothesised maternal adaptational processes and child adaptational processes in accounting for variance in adjustment (Thompson, Gustafson, Hamlett & Spock, 1992, Thompson, Gustafson, Gil, Kinney & Spock, 1999). For example, Thompson, Gustafson, Gil, Godfrey and Murphy (1998) showed that cognitive appraisal processes (stress appraisal, expectations of efficacy and health locus of control) accounted for a total of 37% of the variance in adjustment in children with cystic fibrosis and 33% of the variance in adjustment in children with sickle cell disease.

This model has also been criticised because of the lack of clarity about how the different elements in the model influence each other. Holmbeck (1997) points out that the diagrammatic representation of the model suggests a mediational model, and initial descriptions of the model did use the word "mediational" to describe the adaptational processes. However, Thompson et al. (1994) do not test a mediational model but a main effects model, using hierarchical multiple regression to determine the predictive value of each or their theoretical constructs.

Both the Wallander and Varni (1998) and the Thompson et al. (1994) models suffer from the lack of clarity in which the different processes are thought to be related to each other. This difficulty arises because of the way in which these factors are inter-related and the difficulty in isolating simple "cause and effect" pathways. For example, whilst cognitive processes may affect psychological adjustment, adjustment is also likely to affect cognitive processes. In addition, the link between maternal and child processes is not made explicit in the Wallander and Varni (1998) model, and only maternal and child adjustment are shown to be linked in the Thompson et al. (1994) model. However, it is very likely that the cognitive processes are related to each other and both impact on each other. Nonetheless they have both been useful models in terms of identifying specific processes that may account for variation in adjustment to chronic illness.

Coping Model

As described above, both these models draw on the stress and coping models originally described by Lazarus and Folkman (1984). This paradigm has been proposed as the most appropriate in terms of understanding variability in individual responses to managing the demands associated with chronic illness. According to this theory there are two stages in appraising a stressor. The first phase involves appraisal of the risk presented by the stressor (primary appraisal) and the second response phase involves the assessment of resources to effectively manage the stressor (secondary appraisal). Then, a specific coping process is used in response to the stressor. An important principle of the model is that the suitability of any particular coping strategy depends on the context and the nature of the stressor. Coping strategies are not inherently "good" or "bad", and their efficacy depends on the specific stressor.

There is very good evidence that individuals do appraise events in different ways and this has proved very helpful in understanding the impact of chronic illness. Illness has different meanings to different people, and the way in which a stressor is appraised will influence beliefs about how stressful the experience will be. For example, it has now been shown in many studies that the mother's appraisal of the impact of her child's illness is a more accurate predictor of psychological distress than the "objective" severity or burden of the illness itself (Davis, 1993; DeMaso, Campis, Wypij, Bertram, Lipshitz & Freed, 1991; Kovacs, Iyengar, Goldston, Obrosky, Stewart & Marsh, 1989; Sloper, 2000).

However, the description of different coping styles has been less helpful and less predictive of psychological outcome (de Ridder, 1997). Different coping style taxonomies have been described that move beyond the original "problem focused" or "emotion focused" styles described by Lazarus and Folkman (1984). For example, the COPE (Carver, Scheier & Weintraub, 1989) identifies 13 different coping dimensions. On the other hand Kidcope (Spirito, Stark & Williams, 1988) assesses 10 different categories of coping: distraction, social withdrawal, cognitive restructuring, self-criticism, blaming others, problem solving, emotion regulation, wishful thinking, social support and resignation. The lack of consensus about the number of coping strategies or the way in which these can be combined into "meta strategies" presents a problem in terms of interpreting different studies.

According to the theory, coping processes are specific to a stressor and to a context (Lazarus, 1993). For example, it is not possible to generalise from the way in which a child copes with injections, to the way they cope with being teased at school because of their illness and yet these are both aspects of "coping with a chronic illness". Lazarus (1993) claims that the coping model has been misinterpreted and misused in many studies because of the failure to appreciate that coping is a process or a style, rather than a trait. He gives an example of one of his studies investigating how students coped with the stressful experience of an exam. The students were evaluated at different time points during the exam process: the warning period when they knew they had to take an exam, the waiting period after the exam but before grades are given, and the period after the results were given out.

> "If the examination had been treated as a single stressful encounter, and coping had been summated across stages, there would have been great distortion in what might have been learned. To collapse what is happening over time is apt to produce findings that are at best uninterpretable and at worst misleading" (p 239).

However, most studies of coping styles in children or parents do make the assumption that it is possible to measure coping style in a general way. The findings from these studies have not been helpful partly because of the mismatch between the levels at which coping styles are assessed (i.e. in an abstract and global way) and the very specific events for which the coping style would be employed. There are a few examples of more contextualised and specific measures of coping strategies e.g. Quittner, Espelage, Opipari, Carter, Eid and Eigen (1998) but these are the exception rather than the rule.

The coping model was adopted with enthusiasm partly because it helped shift the focus away from the inevitability of maladjustment, towards a more positive and accurate description of the considerable differences between the way individual children, mothers and families adapt to chronic illness. However, it has only been of limited use because of the way in which coping styles have been assessed. In order to be clinically useful, it would be necessary to assess coping styles in a detailed way on an individual basis, but this has not been tested systematically yet (Tennen, Affleck, Armeli & Carney, 2000). However, the importance of the concept of appraisal that is a central feature of the model, has been shown to be important and helpful.

A Systemic Model of Adaptation to Chronic Illness

Rolland (1987) has developed a systemic model for understanding the impact of chronic illness, but this model has not been empirically tested to the same degree as the previous two models. Rolland's (1987) theory places more emphasis on the impact of illness within the developmental framework of the family. He developed a psychosocial typology to describe illnesses according to their psychosocial demands. Four key variables are used to describe illnesses, and although these are seen as continuous variables, categories are used as key anchor points. These are 1) onset of illness (acute or gradual), 2) course of illness (progressive, chronic or relapsing/episodic), 3) outcome (fatal, shortened life span/possibly fatal or nonfatal) and 4) incapacitation (incapacitating or non incapacitating). He also argues that it is important to understand the impact of the illness in terms of the developmental phase of the illness (e.g. crisis of diagnosis, chronic phase or terminal phase). Finally he argues that there is a third dimension consisting of the general development of the family, for example whether they are currently experiencing centrifugal or centripetal forces. Illness would usually exert a centripetal force on a family and this might cause conflict if the family were otherwise experiencing centrifugal forces, for example, during adolescence.

Whilst this model has not been extensively researched is does have certain strengths. It highlights the need to consider the illness within the context of both the child's family system, and the wider healthcare system, including the impact of working with ill children on health care staff (Rolland, 1994). It has been very influential in clinical work, because it explicitly looks at the meaning of illness for families on an individual basis. The research evidence however, does not support

Rolland's (1987) typology of illnesses, which results in rather odd combinations of conditions, such as spinal cord injury being grouped together with acute blindness.

Kazak (1989) has also developed a "systems and social ecological model of adaptation and challenge" to understand the impact of a chronically ill child on the family. She has demonstrated important differences in the support networks of families with a chronic illness compared to families of healthy children (Kazak, Reber & Carter, 1988; Kazak & Wilcox, 1984). In addition, her model draws attention to the wider system in which families live, and the importance of considering the social environment on the psychological impact of families with a chronically ill child. This model has some empirical support and has highlighted the need to consider the context, as well as the individual child and his or her family.

Psychodynamic Models of Adjustment to Chronic Illness

Although there are many descriptions in the literature of clinical work based on psychodynamic theories, there are very few empirical studies or evaluations of this work (Judd, 1989; Sourkes, 1995). Psychodynamic models place more emphasis on the relationship between the child and his or her mother than other models. In particular, attachment theory has been used as a theoretical framework to understand the impact of a child who has a chronic illness or disability (Marvin & Pianta, 1996). Van IJzendoorn, Goldberg, Kroonenberg and Frenkel (1992) carried out a meta-anlysis of all studies on attachment that have evaluated the effects of either child problems or maternal problems and included several studies on children with physical illnesses. They concluded that "child problems do not lead to significant decreases of secure attachments compared to the normal population" (p.854). Greenberg (1999) also reaches the same conclusion, however he acknowledges that there may be differences between different types of physical illnesses. In particular, a study of children with epilepsy (Marvin & Pianta, 1996) did find higher rates of insecure attachment amongst this particular group, which suggests that there may be some factors which do affect attachment, which are not well understood as yet.

Several authors have linked the mother's difficulty in adjusting to the birth of a child who is not "perfect" to her own unconscious difficulties about aspects of herself. For example Lax (1972) refers to the "narcissistic trauma" the mother experiences when a child is born with an obvious abnormality and links this with the mother's own difficulty in accepting imperfect aspects of herself. She argues that unless this difficulty is overcome, the mother may have difficulties separating herself from her child on a symbolic level, which may account for the patterns of over solicitous caring described by some clinical reports. Solnit and Stark (1961) first described the mother's reaction to a chronically ill or disabled child as "mourning for the perfect child" that had been expected and this analogy to grief has been widely applied to describe the response to having a chronically ill child.

In addition, some chronic illnesses cause considerable discomfort in the child, and the mother's ability to "contain" her child's discomfort is seen as very important (Bick, 1986; Bion, 1962). Part of the mother's task is not only to provide good physical care for her child but also to provide additional psychological support as a consequence of the child's physical illness.

Although there is little research evidence on the psychodynamic models, they have been very influential in clinical work and therapeutic interventions have been described in detail (Judd, 1989; Sourkes, 1995). This is partly because, like the systemic models, they place more emphasis on understanding the meaning of illness for a mother or family, rather than being able to predict outcome successfully. These models are therefore often seen as clinically useful, but there has been little systematic evaluation of the theoretical models underlying the therapy.

Evaluation of Therapeutic Interventions

Successive reviews have highlighted the importance of taking forward the theoretical research in order to develop effective therapeutic interventions (Eiser, 1990; Pless & Nolan, 1991; Wallander & Varni, 1998). However, there are very few evaluations of therapeutic approaches with children with chronic illness. As shown in the above section, there is a gulf between the theoretical models that have been empirically researched, and the clinical descriptions of therapeutic work that have been not been studied empirically. This reflects a gap between theory and clinical practice in psychology in general (Lazarus, 2000).

Results of preventative counselling programmes designed to reduce the likelihood of the development of psychological disorders among this "at risk" group have been disappointing. An early study by Pless and Satterwhite (1972) demonstrated improved psychological functioning and high levels of satisfaction among families of chronically ill children who received a generic, supportive intervention provided by non-professional, family counsellors. However, a large, randomised controlled study designed to test this intervention on a larger scale that used hospital social workers to provide a similar input failed to replicate these findings (Nolan, Zvagulis & Pless, 1987). They found no positive beneficial effect of six months of social work support to families attending general paediatric clinics. The study included a wide range of adjustment measures and included an analysis of sub-groups (for example, the families who had actively engaged in the social work input) and no significant differences between the group receiving this input compared to the control group were found. Given the methodological rigour of this study it seems unlikely that such general preventative interventions are effective in reducing psychological symptoms when applied in this way. Kupst and Schulman (1988) also found no long term benefit for families with a child with cancer of a coping skills programme, although there was some short term benefit to families using the programme just after the diagnosis period.

Pless et al. (1994) carried out a subsequent randomised study that demonstrated significant improvements in self esteem and symptoms of anxiety/depression in children who were assigned to a specialised nursing intervention, compared to children who received standard nursing input. Paediatric nurses were chosen to deliver the intervention because of the central role they play in the care of children with a chronic illness, and the nurses were given appropriate additional training in child psychology. Although the results in this study were quite modest, they do demonstrate that it may be possible to deliver a successful psychological intervention via existing nursing staff.

Psychological interventions focused on specific problematic symptoms associated with a chronic illness have been shown to be effective. A meta-analysis of 42 studies of psychological interventions for children and adolescents with chronic illness found an overall effect size for interventions of 1.12 (Kibby, Tyc & Mulhern, 1998). However, these were much more specific interventions based on disease management or targeting emotional or behavioural symptoms caused by chronic illness conditions e.g. relaxation training for headache or for painful medical procedures. The majority used behavioural interventions and only a small number of illnesses were included i.e. cancer, headaches, diabetes, asthma, cystic fibrosis, juvenile rheumatoid arthritis, renal disease and congenital heart diseases. Hence it is likely that behavioural techniques may be helpful for very specific problems such as pain management. The only non-behavioural therapy that was found to have a beneficial effect was described in Moran, Fonagy, Kurtz, Bolton and Borok's (1991) study of psychotherapy for diabetes, which did demonstrate a reduction in variation in biological indices of diabetes for children who received psychodynamic therapy.

The Psychological Impact of Skin Disorders in Childhood

As this review of studies and models has shown, the field of paediatric psychology has developed considerably over the past twenty years. The development of theoretical models has enabled the research to move towards more clarity in the exploration of the influence of different factors on the impact of chronic illness on children and their families. However, there is considerable variation in the depth in which particular disorders have been studied. Most studies have concentrated on rare or serious conditions, such as cancer, diabetes or cystic fibrosis. Skin disorders have rarely been studied and there is a lack of good studies on the impact of skin disorders on children and families in the literature. However, there are widely held beliefs about the reciprocal influences between the skin and the psyche (Papdopoulos & Bor, 1999).

There are several different ways in which the psyche and skin would be expected to be linked. Firstly, psychological problems may arise as a direct result of the experience of having a skin disorder. For example, someone with a very disfiguring skin condition may have low self esteem and difficulty with peer relationships which could lead to severe social anxiety and/or depression. Secondly, psychological difficulties may precipitate or exacerbate skin problems e.g. anxiety may precipitate psoriasis in someone who is prone to this condition. Thirdly, there may be some underlying biological mechanism linking both the skin and the psyche, and the most frequently cited candidate is the immune system (Hashiro & Okumura, 1998). Finally, psychological disorders do sometimes present as a skin problem, for example in the very rare cases of delusional parasitosis or dermatitis artefacta (Woodruff, Higgins, du Vivier & Wesseley et al., 1997).

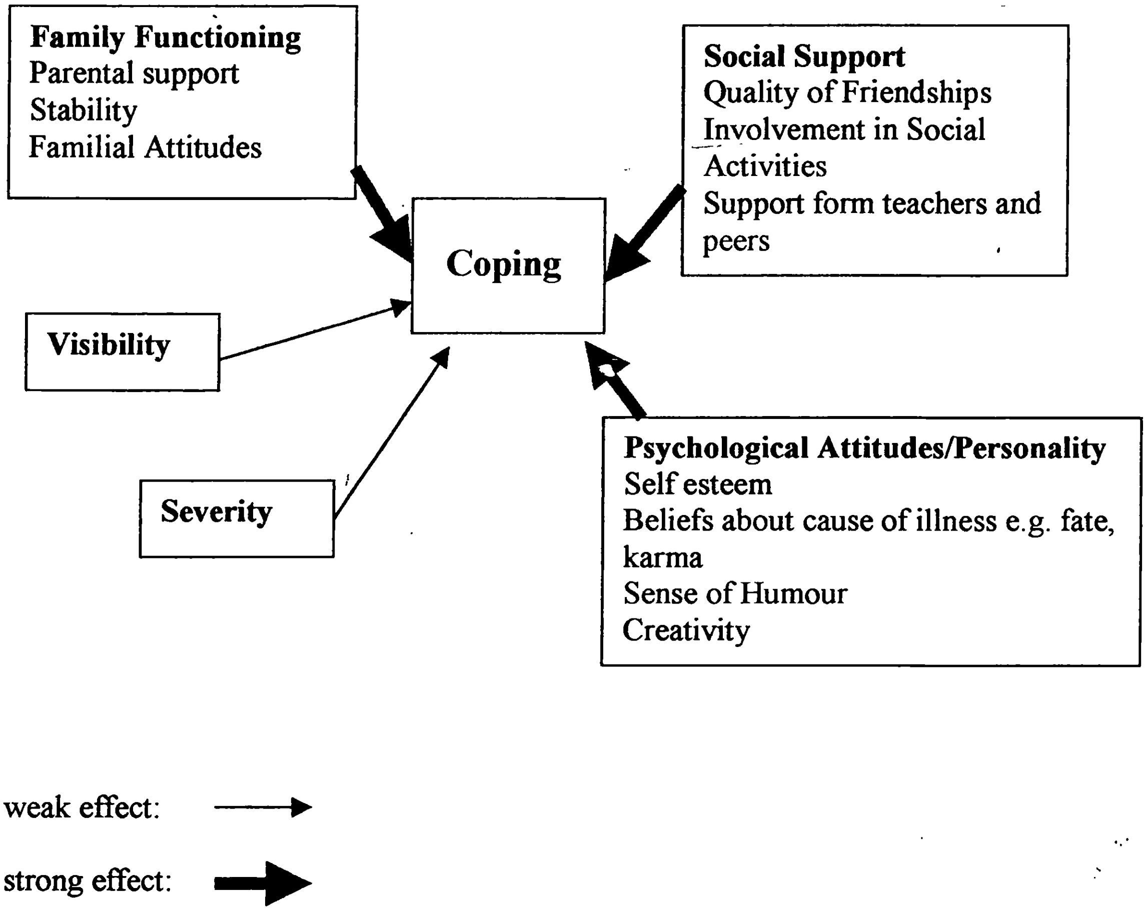
Papadopoulos and Bor (1999) suggest that a biopsychosocial model, which integrates the reciprocal relationships between physical, psychological and social factors is the most appropriate model for understanding the relationship between skin disorders and psychological adjustment (Engel, 1977). There is good evidence that skin disease causes social difficulties and psychological symptoms. For example, Hughes, Barraclough, Hamblin and White (1983) carried out a survey of psychological symptoms in adults attending dermatology services and demonstrated high levels of psychological symptoms amongst both groups. Jowett and Ryan (1985) carried out in depth interviews with adults attending dermatology out-patient services and revealed a high level of concern about the social impact of skin disease. This study is a good descriptive study of the self reported impact of skin disease, but because of the lack of a control group, the significance of the findings have not been demonstrated. There is also good evidence that patients referred to specialised dermatology-psychiatry clinics have a high rate of psychological difficulties (Woodruff et al., 1997). Some of these patients did have the skin conditions thought to be caused entirely by psychological features e.g. dermatitis artefacta or delusional parasitosis, but these were rare (4.7%). The majority had symptoms of anxiety or depression (58%). There is some evidence that stress is related to the exacerbation of vitiligo (Al'Abadie, Kent & Gawkrodger, 1994) and that cognitive-behavioural techniques can be helpful in reducing stress and associated symptoms of vitiligo (Papadopoulos, Bor & Legg, 1999).

However, all of these studies are with adults and there are very few studies of children and families. Several areas are considered to be of particular importance for children. For example, the impact of the child's physical appearance on the relationship between the child and his/her carers, the social context, particularly school, where the child may experience teasing or bullying, and the impact of the skin disease on the child's psychological development.

Papadopoulos and Bor (1999) describe a model of the impact of both disease, personal and contextual factors on the child's coping with skin disorders (Figure 1.3). As can be seen, the framework of the model is very similar to the Wallander and Varni (1998) model, except that coping is seen as the central outcome, rather than a process leading to adjustment. Characteristics of the disorder (severity and visibility), socio-ecological factors (family environment and social support) and personal factors (attitude and self esteem) are all seen as direct influences on adjustment.

Figure 1.3 Influences on Children's Coping with Skin Disorders

(Papadopoulos and Bor 1999)



However, unlike the Wallander and Varni (1998) model, this model does not specify the interaction of the predictor variables e.g. the impact of severity on psychological attitudes or self esteem. The strength of the relationship between the predictors and coping is also based on the Papadopoulos and Bor's (1999) assumptions rather than empirical studies. However, it does help clarify the personal, family and social influences on the child's reaction to the skin condition.

Children and adults with skin disorders are stigmatised by our society.

Appearance is very important in social interaction and studies have shown that children who appear less attractive are less popular than attractive children (Sigelman, Miller & Whitworth, 1986). Goffman's (1963) stigma theory identified social disapproval as the hallmark of stigma. Stigma theory also proposes that an individual who has a stigmatising attribute may have lower self esteem and have to develop strategies to manage disclosure of their condition. Stigma theory stresses that certain characteristics of a mark affect how vulnerable the individual is, for example, the more visible or visually disturbing the mark, the greater the associated stigma.

For some chronic illnesses the child can choose whether or not to disclose his or her illness to other people (Westbrook, Bauman & Shinnar, 1992). However, for many children with skin disorders it is not possible to conceal the disorder and hence they are "exposed" to the reactions of other people on a regular basis. Dealing with other people's reactions can be very difficult for a child or parent and other people's reactions are frequently experienced as hostile and intrusive. Richardson (1997) gives a vivid account of her experience of living with acne that illustrates some of the difficulties experienced by someone with a severe skin disorder. She describes some of the events she remembered hurting her as a child:

> "My best friend, when I was eleven, 'Your nose is just like a strawberry, and it's even got the black bits'; or later, my five year old brother refusing to kiss me because I was 'too spotsy'; children asking, 'Mummy, has that lady got chickenpox?'; some misdemeanour on my part, and my father's contemptuous rage, 'Look at you standing there with your spots hanging out.'" (p 62)

Visibility is one of the characteristics of a physical illness that has been seen as an important determinant of psychological outcome (Wallander & Varni, 1998).

Despite this, there are few studies demonstrating a straightforward link between visibility as a risk factor and psychological adjustment. It has been suggested that the relationship between visibility of the condition and psychological adjustment may not be a linear one. Pless and Nolan (1991) argue that visibility may be important because it influences the likelihood that the child is forced to accept himself or herself as a "disabled person". They conclude that such acceptance might actually contribute to better adjustment, and that a "marginal" state in which there is ambiguity about whether or not the child is seen as "normal" or as "disfigured" might be the most harmful situation.

Some studies have shown evidence that mothers are less responsive towards facially disfigured babies (such as babies born with cleft lip and palate) than towards normal babies (Barden, Ford, Jensen, Rogers-Salyer & Salyer, 1989). Speltz, Endriga, Fisher and Mason (1997) found that children with a cleft lip and/or palate did not have lower rates of secure attachment than a comparison group of healthy children on average, and that contrary to expectation, insecure infants were rated as more physically attractive than secure infants. They explain this finding by speculating that mothers of less attractive infants with cleft palate may perceive their infant to be more vulnerable and this could stimulate caregiving behaviour in a way that results in promoting more secure attachment. Hence, the link between the visibility of the condition and psychological adjustment may not be a straightforward one.

Parents of a child with a skin disorder have to manage not only their own reaction to the child's skin, but also the reaction of other members of the family, friends and strangers. Bradbury (1996) found that the reactions of grandparents are very important in this process. Teasing is extremely common and many children with skin disorders are affected by being bullied because of their appearance

(Papadopoulos & Bor, 1999). This can be a particular problem when the child has to move to a new context, for example going to school or moving on to a different school. Despite the considerable improvements in the awareness of the negative effects of bullying within schools and the implementation of anti-bullying policies, many children report repeated name calling and intrusive curiosity. Adolescence can be a particularly difficult time for young people with skin disorders. Most adolescents feel more self conscious during adolescence and peer acceptance is particularly important for this age group (Walters, 1997).

Bradbury (1996) and Kish and Lansdown (2000) describe interventions based on helping children who are visibly different to develop specific social skills to manage the reactions of other people. Some evidence suggests that these skills can alter the impact of these influences on the person with a disfigurement (Rumsey, Robinson & Partridge, 1996). This work is based on children with disfiguring conditions such as cranio-facial anomalies, as well as skin conditions. The interventions include strategies such as using an imaginary force field to deflect hurtful comments from other people, and being prepared to take the initiative in social encounters when faced with someone who is unsure and embarrassed about talking to someone who is disfigured.

Bridgett, Noren and Staughton (1996) describe a treatment programme for children with eczema that is based on behavioural principles. This focuses on managing the symptoms of eczema (itching) rather than the social consequences of a skin condition, but this approach has not been systematically evaluated with children as yet.

Summary

Empirical studies have demonstrated that children with a physical illness and their parents are at increased risk of developing psychological problems. However, there is considerable variation in the range of responses for both the child and the parents. Theoretical models based on the transactional model of stress and coping have been developed to determine the contribution of disease related, family, social and intrapersonal factors to psychological outcome. Studies have shown the importance of the individual's appraisal of the impact of the illness in determining psychological outcome. Systemic and psychodynamic models have placed more emphasis on the importance of the meaning of the illness and the impact of the illness on relationships. However, there are very few empirical studies of these models.

Several methodological difficulties have been identified. The noncategorical approach to illness is not supported by recent evidence. There are few qualitative studies of the impact of illness, and the reliance on quantitative methods have led to more emphasis on the predictive utility of the models than on understanding the psychological difficulties experienced by children and their families. The lack of research on clinical interventions in this area is partly due to the practical difficulties in this type of research, but also the theoretical rather than applied nature of the empirical studies.

Skin disorders have not been well researched, despite the obvious relevance of this research. The visibility of skin disorders and the discomfort associated with the conditions are thought to be important influences on adjustment.

Outline of Thesis

This thesis examines the impact of skin disorders on children and families for two different skin conditions. The first two studies are about the impact of the rare, genetic condition known as Epidermolysis Bullosa (EB). EB causes exceptionally fragile skin that blisters easily during normal activities. The next four chapters describe a quantitative study of the impact of EB on children and their parents (Chapters Two to Five). Then, a qualitative study of the experiences of mothers of children with EB is described (Chapters Six to Eight). The final study is about the more common condition of atopic eczema. This study explores further one of the themes from the qualitative study of EB and examines the link between severity of eczema and psychological adjustment (Chapters Nine to Twelve). The final chapter summarises the findings from the thesis and identifies areas where further research should be focused.

CHAPTER TWO

THE PSYCHOLOGICAL IMPACT OF EPIDERMOLYSIS BULLOSA

INTRODUCTION

Description of Epidermolysis Bullosa

The first two studies evaluated the psychological impact on children and families of the skin blistering condition known as Epidermolysis Bullosa (EB). EB is a rare, genetic disease that causes exceptionally fragile skin. Literally, it can be translated as breakdown (lysis) of the skin (epidermis) and blistering (bullosa). This is due to defects in the fibres that hold together the layers of the skin, resulting in separation of the skin layers and the development of blisters. There is considerable variation in terms of the effect of EB both in terms of the type of EB and even within each type. At the mild end of the spectrum, the disease results in blisters mainly on feet and hands that tend to be worse in hot weather. This may mean the person is only affected for the warmer months of the year. At the severe end of the spectrum, the damage is so severe it usually results in death within the first few years of life.

There are three major forms of EB - dystrophic, simplex and junctional (Atherton, 1997; Eady, 1990). In dystrophic (literally "scarring") EB, the defect is in the dermis (lower levels of the skin) and as a result the damage to the skin is serious. This form of EB can either be localised (affecting only a small area of the body) or generalised. When the blisters heal, the skin scars and the appearance of the skin changes over time. As a result of the scars and healing process, the skin can fuse between fingers and toes and cause contractures. In addition, the mouth and oesophagus can be affected leading to difficulties with swallowing solid food. The lower bowel and anus can also be affected, resulting in difficulties with passing stools. People with dystrophic EB have increased nutritional needs, due to the demands of wound healing and loss of fluid from the blisters. Severe dystrophic EB is associated with a reduction in life expectancy. Dystrophic EB can be inherited in a recessive or dominant form, and the recessive form tends to be much more severe than the dominant form.

In EB Simplex, the fault is in the epidermis (outer layer of skin) and in this form of the disease, the skin heals normally without scarring. EB Simplex is often thought of as the mildest form of the disease, but it can involve considerable pain and disability. Whilst the main areas affected are the hands and feet, the blistering can be severe enough to make it impossible for a child to walk around school. EB simplex is usually inherited as a dominant trait. There are several subtypes of EB simplex, for example, Dowling-Meara or Weber-Cockayne.

Junctional EB is the result of a fault in the adhesion between the epidermis and the dermis. As a result, the condition is life threatening, and babies born with the disease usually die within the first few years of life. Junctional EB is inherited as a recessive trait.

There is no cure for EB at present, although there are hopes that gene therapy may provide a treatment in the future. The management of the condition involves minimising the damage to the child's skin (Denyer, 1999; Foster, 1990). This usually involves protecting the skin by bandaging and dressing it with padded dressings, and limiting activities that may damage it. In order to reduce the damage caused by blisters, these have to be burst using a sterile needle and the wound dressed to help prevent infection. For a child with a severe form of EB, this procedure can take up to three hours daily. The rarity of the condition means that it can take parents a considerable amount of time before they find the most appropriate system for managing their child's skin. The management of EB has improved in the recent past (Lin, 1996). For example, infections are better controlled and can be treated more successfully. Nutritional management has also improved, and children who have experienced considerable difficulty with eating and hence with growth, are now given gastrostomies, which ensure adequate nutrition (Clayden, 1990).

There are no exact figures on the prevalence of EB, partly because many of the milder forms may never have been diagnosed. Davison (1965) calculated the minimal frequency as 1 per 50,000 for all dominant forms of EB and 1 per 300,000 for the recessive forms. National registers have recently been started in the UK and the USA. People with EB have been traced both through hospital and GP records and through informal contacts, such as family contacts and press advertisements. Evidence from the Scottish survey suggested that the incidence of EB simplex was 0.43 per million per year. For dystrophic EB the incidence was 0.2 per million per year, and for junctional EB about 0.05 per million per year (Horn, Priestly, Eady & Tidman, 1997). However, at least 30% of the people interviewed for this register had never seen a dermatologist, which suggests that there may well be more people in the community with the milder forms of EB who have never sought medical treatment for their condition.

Review of Studies on the Psychological Impact of EB

From a psychological perspective, there are several features of the condition that seem to be important in terms of the effect it has on children and their families. Firstly, the condition is genetic and although spontaneous mutations do occur, these are extremely rare. In the dominant forms of EB, the condition may have passed through many generations, although often the family will have a limited understanding of the condition, with many families even unaware of the diagnosis. In the recessive forms of EB, the parents will be unaware of their status as a carrier of EB, because carriers are totally asymptomatic. The likelihood of having any previous knowledge or experience of EB is extremely slight. Prenatal diagnosis is now available for couples who have had one child with EB, or who have a history of EB in their family. As with all forms of prenatal diagnosis, this can be extremely stressful for families, and many families find the decisions involved extremely difficult (Rodeck, 1990).

The blisters formed in EB are extremely painful for the child, particularly as they often form at points of friction or on joints. In addition the management of the blisters is in itself painful. The child therefore has to cope with pain throughout his or her life, and the daily unpleasant routine of treating blisters. For parents, both having to watch their child in pain, and having to inflict pain in order to manage the condition are the worst aspects of the condition.

As noted above, the dystrophic forms of EB are particularly disfiguring, both in the appearance of the damaged skin, which often looks like it has been burnt, but also in the fusion and distortion of the child's hands and contractures of other joints. Hoxter (1986), a psychotherapist, vividly describes the impact of working with a child with severe dystrophic EB on the members of staff and children at a school for disabled children where she worked:

> "Her raw state seemed to break through all their own defensive barriers, her screams and rages when being undressed to use the toilet, which often entailed tearing at her clothes stuck to thin tissues and clots, were unbearable to them. The disabilities of the other children seemed to become increasingly invisible to the teachers whereas they experienced this little girl as a constant

> confrontation with pain and trauma, frightening for the other children to perceive." (p. 100).

In the milder forms of EB and particularly for EB simplex, the EB is usually not disfiguring in the same way, although the blisters themselves may be visible. Paradoxically, this can make it very difficult for people with little experience of EB to understand why people with EB simplex have such difficult with mobility.

There are very few studies on the psychological impact of EB. Lansdown, Atherton, Dale, Sproston and Lloyd (1986) carried out a postal survey of the needs of families with EB. They sent a questionnaire to all 138 families who were members of the Dystrophic Epidermolysis Bullosa Research Association (DEBRA) and received 63 replies (45.5 % return rate). (DEBRA is a charity that helps sufferers of all types of EB by publicizing the effects of EB, supporting families with the condition and funding research into EB). Lansdown et al. (1986) demonstrated that families frequently felt dissatisfied with the level of information and practical support given to them by local services. 44 % of respondents reported that they had no emotional support at the time of their child's birth and 52 % of respondents reported practical difficulties in managing their child's skin.

Lansdown and Nabarro (1990) carried out a small qualitative study to look specifically at psychological difficulties in families with children with EB. They interviewed five families in their homes using a semi-structured interview. Whilst they found little evidence of overt psychological distress, they concluded,

> "There are indeed psychological difficulties both potential and actual, but they may often go unnoticed. They go unnoticed partly because we do not look for them and partly because people are often very good at hiding their feelings and denying difficulties." (p. 20).

Despite the lack of studies on the psychological impact of EB, there is recognition of the importance of psychological factors in the care of children with EB. For example, Atherton (1990) described counselling as an essential part of the medical care offered to families with EB. He described the main aim of the medical team as "helping the child and parents to cope, with the aims of making their lives more bearable and of reducing the handicap the disease causes" (p 17). This holistic approach to EB is reflected in the use of multi-disciplinary teams for treating EB.

Aims of the Study

The main aim of the study was to evaluate the psychological impact of EB on children with EB and their parents. Since so few studies have been done on the psychological impact of EB, this included a description of the level of psychological difficulties experienced by children and their parents. The study also assessed the relationship between the severity of EB and the psychological impact of the condition. Severity of the illness is hypothesised in both the Wallander and Varni (1998) and the Thompson et al. (1994) models to have an impact on adjustment. This study tested whether there was a direct relationship between severity of the EB, and the adjustment of the child or parents.

In addition, the relationship between family functioning and psychological adjustment to EB was explored. Family processes have been shown to be associated with adjustment in other studies of children with chronic illnesses (Drotar, 1997) and are seen as resistance factors in the Varni and Wallander (1998) model and as indirect influences on adjustment in the Thompson et al. (1994) model. This study tested whether there was an association between family functioning and psychological adjustment in the child or parents.

The study also examined the child's understanding of EB and their view of the difficulties they experienced. This involved a brief interview with the child with EB in order to determine their knowledge about the condition and to investigate the coping strategies used by the children in both general contexts and situations related to their EB.

The study was designed to address some of the methodological weaknesses identified in the review of previous studies in Chapter One. For example, multiple informants were used including the mother, father, child and child's teacher. Interview methods were used where possible to overcome some of the difficulties of relying on self-report measures. A measure of psychosocial adjustment was used rather than a measure of psychiatric symptomatology in order to obtain a broader measure of the impact of the EB on the child. The study used both quantitative and qualitative methodology in order to obtain descriptive data but also to gain some understanding of the psychological processes underlying the child and family response to EB.

Research Questions

1) Is the severity of the child's EB associated with the child's psychological adjustment or parental psychological distress?
2) Is family functioning related to the child's psychological adjustment or parental psychological distress?
3) How do children with EB view their illness? What coping strategies do children with EB use to manage the difficulties they experience?

The method, results and discussion for this study are covered in Chapters Three, Four and Five. The second part of the study was a qualitative study of the experiences of mothers of children with EB. The method, results and discussion for this study are covered in Chapter Six, Seven and Eight.

CHAPTER THREE

THE PSYCHOLOGICAL IMPACT OF EPIDERMOLYSIS BULLOSA

METHOD

Overview

The parents of children with EB completed questionnaires to assess the child's psychological adjustment, parental psychological distress, family functioning, severity of EB and disability caused by the child's EB. Children with EB were interviewed about their experience of having EB and, if appropriate, completed a self esteem questionnaire. For children of school age, the school also completed a brief questionnaire about the child's psychological adjustment at school.

(Mothers were also interviewed about their experience of having a child with EB for the qualitative study, and the method, results and discussion for this are covered in Chapters Six, Seven and Eight).

Participants

Participants were recruited from the monthly EB clinic at Great Ormond Street Hospital, and from families contacted by the Dystrophic Epidermolysis Bullosa Research Association (DEBRA), who indicated they would be willing to participate in the research. All of the ten families approached at the EB clinic at Great Ormond Street Hospital agreed to take part in the research. Out of 25 families approached by DEBRA, 17 (68%) returned a consent form indicating they would be prepared to take part in the study. All the families recruited at Great Ormond Street Hospital were also members of DEBRA. Written consent was obtained for all families (Appendix 1).

Ethical approval for the study was obtained from the Research Ethics Committee at Great Ormond Street Hospital.

The initial sample consisted of 31 children with EB from 27 families. Of these families, three were not interviewed and did not complete questionnaires although they had consented to the research, because of difficulties they were experiencing at the time of interviews. One family had returned to their country of origin because of difficulties coping in this country, one child was the subject of care proceedings by the local authority, because of concerns about his welfare and in one family, the child died. All these three children were recruited via Great Ormond Street Hospital and had recessive dystrophic EB. The final sample therefore consisted of 28 children from 24 families.

Of the 28 children with EB, 16 (57%) were girls and 12 (43%) boys. Seven of the children (25%) were recruited from Great Ormond Street Hospital and 21 (75%) from DEBRA volunteers. Table 3.1 shows the frequency of the different types of EB and Table 3.2 shows frequency of type of EB by source of referral. The average age of the children was 8 years 10 months, the range was 2 years 6 months to 16 years 3 months. 21 (75%) of the children were aged 5 or above.

There were 18 (75%) families with both biological parents living together or married, 3 (12.5%) families where the parents had separated but the child was living with his or her mother and her new partner, and 2 (8.3%) families where the parents were separated and the child lived with their mother. In one family the mother was a widow.

Table 3.1 Type of EB by Sex of Child

Type of EB	Girls	Boys	Total
Recessive Dystrophic	4	7	11
Dominant Dystrophic	4	0	4
Simplex	7	3	10
Unknown [a]	1	2	3
TOTAL	16	12	28

Table 3.2 Type of EB by Source of Referral

Type of EB	GOS	DEBRA	Total
Recessive Dystrophic	5	6	11
Dominant Dystrophic	1	3	4
Simplex	1	9	10
Unknown [a]	0	3	3
TOTAL	7	21	28

Note: [a] Although these children had not had skin biopsies to confirm the type of EB they had, these 3 families had been told that it was EB simplex, and thus they are assumed to have EB simplex in this study.

Table 3.3 shows the socio-economic status of the families, based on the occupation of the main earner. The figures for national sample based on the General Household Survey 1988 are given as a comparison (O.P.C.S., 1990). Although the EB sample included all socio-economic groups, there were relatively more families from social group IV than would be found in the general population and fewer families from social group III.

All the families were of White British ethnic origin except one boy of mixed race, whose mother was White British and whose father was Afro-Caribbean.

Table 3.3 Socio-Economic Status of Sample

Socio Economic Group	EBSample (%)	National Sample (%)
I Professional	8.7	7
II Managerial and Technical	21.7	22
III Skilled Manual and Non-Manual	39.1	52
IV Partly Skilled	21.7	14
V Unskilled/State Benefits	8.7	5

Procedure

The interviews were conducted in the family home with both the mother and the child affected by EB. The father was also present for some of the interviews. After the interviews, the questionnaire measures were left for the mother, and if possible, the father to complete and return by post.

Measures

Interview with the child

If the child was aged five or over, he or she was also interviewed using a semi-structured interview (Appendix 2). Children under five were not interviewed because it was felt that they would be too young for this sort of format to provide reliable results. These interviews were tape recorded and transcribed. There were four sections to the interview. Firstly, the child was asked about their understanding of EB and his or her knowledge of the condition was assessed. Secondly, the child was asked to describe their experience of EB, and asked about the three worst aspects of EB. Thirdly, general coping strategies were investigated. Three commonly experienced stressors (a tummy ache, a friend cancelling an arrangement to meet and being teased) were used as standard stressors to investigate coping strategies. A brief scenario incorporating each of the three standard stressors was read to the child. The children were asked whether there was anything they could do to make themselves feel better under these circumstances. They were then questioned about how successful their nominated strategy was and how they felt it worked. Finally, the child was then asked about the specific coping strategies they used for the three worst features of EB they had nominated.

The strategies used by children for the three standard stressors were coded using the scheme described by Ryan-Wenger (1992) (Appendix 3). She developed this scheme by analysing all the strategies identified in previous studies of children's coping responses and combining them into one system with 15 different categories, such as aggressive activity, cognitive avoidance, cognitive distraction, social support. This scheme was chosen because it is one of the few multi-dimensional schemes described in the literature that is suitable for self reported material from an interview. The author and one independent rater completed the ratings (See Appendix 3 for Coding Instructions). The overall percentage agreement was 90% for the main strategy used (Cohen's Kappa 0.87).

Children aged over eight years old also completed one brief questionnaire about self esteem, described in the questionnaire section below. This was introduced and explained by the author at the end of the interview. This questionnaire is designed for children aged eight or over therefore younger children did not complete this.

Questionnaires Completed by Parents

Family Environment Scale (FES: Moos & Moos, 1986). This self report questionnaire contains 90 items answered either "true" or "false". The full version consists of 10 sub-scales with 9 items each and is quite time consuming to complete. Three sub-scales were excluded (intellectual-cultural orientation, active-recreational orientation and moral-religious emphasis). The remaining 7 sub-scales were reduced to 7 items each and the resulting scores pro-rated for comparisons with normative data. This was done because the original questionnaire is very lengthy and the excluded scales were not felt to be of great importance to this study.

The sub-scales used were cohesion (e.g. "family members really help and support one another"), expressiveness (e.g. "we say anything we want to around home"), conflict (e.g. "we fight a lot in our family"), independence (e.g. "we think things out for ourselves in our family"), achievement orientation (e.g. "we feel it is important to be the best at whatever you do"), organisation (e.g. "being on time is very important in our family") and control (e.g. "there are set ways of doing things at home").

Both parents independently completed the FES according to how they saw the family. The FES has normative data based on a large community sample for a combined group of mothers and fathers. Studies have shown no significant differences between mothers' and fathers' ratings of their family (Moos & Moos, 1986; Vostanis & Nicholls 1995).

Rating of severity of EB. As there is no standard way of rating the severity of EB, a simple scale was devised (Appendix 4). This involved the parent scoring the severity of their child's EB on a six point scale for each of 10 body areas, ranging from 1 (not affected), to 6 (very severely affected). These scores were summed to give a total score reflecting the extent to which body surface was affected. This measure is referred to as the "Extent of Body Area Affected". Parents were also asked to give an overall rating of the severity of their child's EB using a five point scale ranging from 1 (very mildly) to 5 (very severely). This measure is referred to as "overall severity".

In addition, the impact of the child's EB on three areas of functioning (mobility, visibility and lifestyle) were also separately rated on 5 point scales ranging from 1 (no impact) to 5 (very severe impact).

Functional Disability Inventory (FDI: Walker & Greene, 1991). This scale consists of 15 items assessing the child's functional disability, i.e. their difficulty in normal activities such as walking up stairs, being at school all day and going shopping. Each item was scored on a five point scale, by rating it as no trouble, a little trouble, some trouble, a lot of trouble and impossible. The items were summed to give a global measure of the child's disability. This scale was included because of the previous literature suggesting that disability may have a specific impact on psychological distress, independently from chronic illness (Cadman et al., 1987).

Personal Adjustment and Role Skills Scale III (PARS III: Stein & Jessop, 1990). This questionnaire was completed independently by both parents and assesses a global measure of the school-aged child's psychological well-being or psychosocial adjustment. It was included because of the concerns discussed in the introduction of using simple symptom based measures or psychiatric diagnostic categories (Eiser, 1990). In addition, the commonly used Child Behaviour Checklist (CBCL:

Achenbach & Edelbrock, 1983) contains several items measuring somatic symptoms, which may lead to higher scores for children with chronic physical illness and hence inflate estimates of psychological problems.

The PARS III consists of 28 items rated on a four point scale corresponding to never or rarely, sometimes, often, always. The items reflect six factors - peer relations, dependency, hostility, productivity, anxiety/depression and withdrawal. It has normative data on children aged 5 to 16 with physical illnesses as well as comparisons with healthy children.

General Health Questionnaire-28 (GHQ-28: Goldberg & Williams, 1988). Both parents independently completed the 28 item version of the General Health Questionnaire. This assesses general well-being and psychological symptoms and is composed of four factors; somatic symptoms, anxiety and insomnia, social dysfunction and severe depression. These were rated using a four point descriptive scale, and scored according to the GHQ scoring system. This system assigns 0 if the symptom is reported as absent or no more than usual, and 1 if it is present. A threshold of 5 is most commonly used to indicate psychiatric "caseness". Extensive validity and reliability studies have been published (Boardman, 1987; Cox et al.,1987).

Child Questionnaire

Self Perception Profile for Children (SPP: Harter, 1985). This is a self report questionnaire for children which assesses five domains of self-worth as well as a global self worth scale. The five domains (scholastic competence, social acceptance, athletic competence, physical appearance and behavioural conduct) are each composed of six items. For the purpose of this study, the questionnaire was shortened to include only three questions for each of the domains. This was done because some of the items are very repetitive, and the questionnaire is very long for children to complete.

For each question the child is presented with two descriptions (e.g. "some kids find it hard to make friends" compared to "other kids find it's pretty easy to make friends"). The child then decides which of the two descriptions he or she is most like, and then whether the statement is "really true for me" or "sort of true for me". The questionnaire begins with one sample item to introduce this format to the child. This format appears to work well in terms of facilitating responses that are less socially desirable. This questionnaire is designed for children aged eight or over.

School Questionnaire

If the child was of school age, the school was contacted with the parents' permission, and asked to complete a questionnaire about the child's coping at school (Appendix 5). This covered the teacher's rating of the child's functioning at school, absences from school over the previous term and information given to the school about EB. The child's teacher also completed the Rutter B Scale (Rutter, 1967). This scale consists of 26 problem behaviours which are rated by the teacher on a three point scale (0 = doesn't apply, 1 = applies somewhat and 2 = certainly applies). This was scored on two factors: emotional disorder and conduct disorder. A total score of 9 is used to indicate whether the child's behaviour reaches a threshold of probable caseness. This scale has been extensively researched and shows good reliability and validity (Elander & Rutter, 1996; Rutter, Tizard & Whitmore, 1970).

CHAPTER FOUR

THE PSYCHOLOGICAL IMPACT OF EPIDERMOLYSIS BULLOSA

RESULTS

The results for each of the quantitative measures are presented and where possible, comparisons made with existing normative data. The research questions are then addressed in turn.

Descriptive Results from the Questionnaires

The full sample consisted of 24 families, with a total of 28 children. One family failed to return their questionnaires after the interview, and they then moved house so it was not possible to contact them. Therefore, all of the results from the questionnaires completed by parents are based on the 23 mothers and 20 fathers who returned questionnaires. The results on the standardised questionnaires are compared with the published norms for the measure.

The results for all the measures were reviewed by plotting out the data to determine whether it could be assumed there was an underlying normal distribution. Unless it is stated otherwise in the results for a particular measure, it was assumed that there was a normal distribution.

Family Environment Scale (FES)

The results of the mothers' and fathers' ratings on the Family Environment Scale are given in Table 4.1. When compared to the norms published for healthy families, mothers of children with EB rated their family as more cohesive ($t(22) = 2.98$, $p < 0.01$) and lower on control ($t(22) = 2.67$, $p < 0.05$) than parents of healthy children. There were no significant differences between the fathers' ratings of family environment and the norms.

Table 4.1 Mean Scores on Family Environment Scale

Sub-scale	Mothers (n = 23)		Fathers (n = 20)		Norms [a] (n = 1125)	
	M	SD	M	SD	M	SD
Cohesion	7.53**	1.48	7.02	1.74	6.61	1.36
Expressiveness	5.87	2.28	5.21	2.01	5.45	1.55
Conflict	3.31	2.81	3.12	2.39	3.31	1.85
Independence	6.36	1.71	6.08	1.94	6.61	1.19
Achievement	4.97	2.04	4.73	1.64	5.47	1.61
Organisation	5.31	2.71	4.33	2.46	5.41	1.83
Control	3.10*	2.22	3.35	2.19	4.34	1.81

[a] Separate norms for mothers and fathers not available

* $p < 0.05$, ** $p < 0.01$

GHQ - 28

On the GHQ-28, 30 % of mothers (n=23) and 40 % of fathers (n=20) scored above the threshold indicating psychiatric caseness (5 or more). The mean score on the GHQ was 4.3 for mothers and 4.5 for fathers. The mothers' scores on the GHQ are comparable with those obtained in a community sample by Cox et al. (1987), who found that 33% of women scored above the threshold. However, the fathers' levels of distress are somewhat higher than the community sample figure of 27%. However, this difference was not significant on a χ^2 test. The scores on the GHQ had a highly skewed distribution with 52 % of the mothers and 50% of the fathers scoring 0.

Functional Disability Inventory and Severity of EB

Parents' ratings of their child's disability on the FDI, and their ratings of severity are shown in Table 4.2. Fathers' and mothers' ratings on the FDI were strongly correlated ($r = 0.84$). Non-parametric statistical tests were used to compare the means because the sample did not have a normal distribution.

There was a significant difference between ratings given to children with different types of EB (Table 4.3). (Because of the high correlation between mothers' and fathers' ratings, only mothers' ratings are shown for simplicity). Children with EB simplex were rated by mothers as significantly more functionally impaired on the FDI than children with dystrophic EB ($U = 45.5$, $z = 1.9$, $p = 0.05$). However, children with dystrophic EB were rated as affected over a greater surface area of their body than children with EB simplex by both mothers and fathers ($U = 36.5$, $z = 2.6$, $p < 0.01$ and $U = 10.5$, $z = 2.5$, $p = 0.01$ respectively). Fathers rated the visibility of the EB as greater for children with dystrophic EB compared to children with EB simplex ($U = 14$, $z = 2.3$, $p < 0.05$). This is consistent with the difficulties reported by many families. EB simplex is much less visible, but the impact of the condition on mobility is often underestimated whilst dystrophic EB is more visible and extensive.

Table 4.2 Parents' Ratings of Severity of EB

Measure	Mothers (n = 27) M	 SD	Fathers (n = 19) M	 SD
Functional Disability	9.9	8.2	10.8	9.1
Extent of Body Affected	25.7	8.5	26.9	10.2
Overall Severity	1.6	0.9	1.9	0.9
Mobility	1.6	1.0	1.6	1.0
Visibility	1.4	1.2	1.6	1.3
Impact	1.6	1.0	1.8	1.0

Table 4.3 Mothers' Ratings of Severity for Dystrophic EB and EB Simplex

Measure	Dystrophic EB (n = 15) M	 SD	EB Simplex (n = 12) M	 SD
Functional Disability	7.2*	8.4	13.5	6.8
Extent of Body Affected	29.6**	8.2	20.8	6.4
Overall Severity	1.6	1.1	1.6	0.9
Mobility	1.3	1.1	1.8	0.9
Visibility	1.6	1.1	1.1	1.2
Impact	1.4	1.0	1.8	1.0

* $p<0.05$, ** $p<0.01$

PARS III

The parents' ratings of their child's adjustment as measured by the PARS III are shown in Table 4.4. These figures are based on 21 children aged 5 or above. Mothers' and fathers' ratings were strongly correlated (r = 0.79). Mothers' ratings on the PARS were statistically significantly associated with SES (r = - 0.57, p < 0.05), and although the fathers' ratings were not significantly associated with SES, there was a moderate size correlation (r = - 0.41).

Fathers rated their children higher on the dependency (t(15) = 2.64, p < 0.05), anxiety/depression (t(15) = 2.53, p < 0.05) and withdrawal (t(15) = 2.43, p < 0.05) scales than the norms, but there was no difference on total scores. (Higher scores represent better adjustment on this scale). Mothers also rated their children as higher on the dependency scale than norms (t(20) = 2.07, p = 0.05).

Table 4.4 Mean Scores on the PARS III

PARS Sub-scale	Mothers (n = 21)		Fathers (n = 16)		Norms [a] (n = 444)	
	M	SD	M	SD	M	SD
Peer relations	10.0	3.2	9.6	3.7	11.4	2.8
Dependency	12.6*	2.0	13.1*	2.2	11.7	2.9
Hostility	19.4	4.1	20.6	4.1	19.1	3.9
Productivity	11.1	3.3	12.1	3.2	11.0	3.1
Anxiety/Depression	19.7	2.7	20.9*	2.9	19.0	3.5
Withdrawal	14.6	1.7	15.1*	1.4	14.3	2.2
Total Score	87.5	12.7	90.8	13.5	86.6	11.9

* $p < 0.05$ compared to norms

[a] Separate norms for mothers and fathers are not available

Self Perception Profile

The ratings given on the Self Perception Profile are shown in Table 4.5. These are based on the fourteen children aged 8 and above. Although there were no differences on the global self esteem measure, children with EB rated themselves as lower on the athletic (t(13) = 4.05, $p < 0.01$) and physical (t(13) = 2.47, $p < 0.05$) sub-scales, than healthy children.

Table 4.5 Mean Scores on the Self Perception Profile

Sub-scale	Children With EB (n = 14)		Norms[a]	
	M	SD	M	SD
Athletic	2.12 **	0.83	3.02	0.73
Behaviour	2.95	0.67	2.80	0.55
Scholastic	3.10	0.65	2.85	0.67
Physical	2.33 *	0.69	2.79	0.71
Social	3.26	0.71	2.94	0.65
Global	3.29	0.55	3.02	0.67

[a] Harter (1985) presents her norms separately by gender for each year age group. For the purpose of this study the norms calculated by Radcliffe, Bennett, Kazak, Foley and Phillips (1996) from Harter (1985) were used. These norms are the averages of the norms given by Harter (1985), across all the age groups and sample groups.

* $p < 0.05$, ** $p < 0.01$

The children with EB simplex had higher self esteem ratings on all scales except for the behaviour scale than the children with dystrophic EB but none of these differences were statistically significant.

School Questionnaire and Rutter B Scale

Two schools failed to return the Rutter B questionnaire and the school questionnaire, hence the data is based on 19 children. On the Rutter B scale which was completed by the child's teacher, 3 children (15.8 %) scored above 9, the threshold for psychiatric caseness on this measure. Of these, two children had predominantly conduct symptoms and one child had predominantly emotional symptoms.

On average, children had missed 4.6 days of school the previous term due to sickness, the range being from 0 to 17 days. (The length of a school term is approximately 60 days). The mother's ratings of the amount of the child's body affected by EB and her rating of overall severity were strongly correlated with the frequency of absence from school ($r = 0.64$, $p < 0.01$ and $r = 0.68$, $p < 0.01$ respectively).

Correlations Between Measures of Adjustment

Table 4.6 shows the relationship between the parent, child and teacher ratings of adjustment. The level of distress in the fathers was significantly negatively related to the level of child psychological adjustment as rated by the mother. The child's rating of self esteem was not significantly related to the parents' or school rating of adjustment. The father's rating of child adjustment was significantly related to the teacher's rating of child adjustment.

Table 4.6 Correlations Between Measures of Adjustment

	Mother PARS n = 21	Father PARS n = 16	Child SPP n = 14	School Rutter B n = 19	Mother GHQ n = 23	Father GHQ n = 20
Mother PARS	1.0	0.79**	0.35	- 0.39	- 0.27	- 0.62*
Father PARS		1.0	0.45	- 0.56*	- 0.36	- 0.41
Child SPP			1.0	- 0.24	- 0.41	- 0.07
Rutter B				1.0	- 0.10	0.36
Mother GHQ					1.0	0.02

* p < 0.05, **p < 0.01

Research Question One – Is the severity of the child's EB associated with the child's psychological adjustment or parental psychological distress?

The correlations between the measures of severity of EB (FDI, Overall Severity and Extent of Body Area Affected) and the measures of child adjustment (PARS III, SPP, Rutter B) and the measures of parental distress (GHQ) are shown in Table 4.7. To reduce the number of comparisons made, the mothers' ratings only were used in the subsequent analyses. The mothers' and fathers' ratings were highly correlated for all three measures of severity (Functional Disability Inventory ($r = 0.84$), overall severity ($r = 0.95$) and the extent of body area affected by EB ($r = 0.95$)).

There was some correlation between the different measures of severity. Whilst overall severity was significantly associated with FDI and the extent of body area affected ($r = 0.58$, $p < 0.01$ and $r = 0.67$, $p < 0.01$ respectively), FDI was not significantly related to extent of body area affected.

None of these three measures of severity were related to any of the measures of child adjustment.

The overall severity of the child's EB and the extent of body area affected were significantly related to father's distress. However, the severity of the child's EB and the extent of body area affected were not related to the mother's distress. FDI was not significantly related to mothers' or fathers' distress.

Table 4.7 Correlations Between Measures of Severity of EB, Child Adjustment and Parental Distress

	Mother PARS N=21	Father PARS N=16	Child SPP N=14	School Rutter N=19	Mother GHQ N=23	Father GHQ N=20
FDI	- 0.33	- 0.33	0.15	0.35	0.22	0.38
Overall Severity	- 0.33	- 0.11	0.26	- 0.06	0.24	0.63**
Extent of Body Affected	- 0.33	- 0.01	- 0.19	0.07	0.08	0.73**

*p < 0.05, ** p< 0.01

Therefore, no relationship was found between child psychological adjustment and severity of EB, or between mother's psychological distress and severity of EB. However, father's psychological distress was related to both the overall rating of severity and the extent of body area affected by EB.

Research Question Two – Is family functioning related to the child's psychological adjustment or parental psychological distress?

Table 4.8 shows the correlation coefficients between the sub-scales of the FES and the measures of severity of EB, child adjustment and parental distress.

Mother's rating of family cohesion on the FES was related to child's psychological adjustment ($r = 0.56, p < 0.05$). Family conflict was negatively related to the child's psychological adjustment ($r = - 0.50, p < 0.05$). Expressiveness was negatively related to disability ($r = - 0.46, p < 0.05$).

Table 4.8 Correlations Between Mothers' Ratings on the Family Environment Scale, Severity and Adjustment to EB

FES Sub-scale	FDI n = 23	Extent of Body Area Affected n = 23	PARS n = 21	SPP n = 14	Mother GHQ N = 23
Achievement	0.25	0.25	-0.42	-0.30	-0.05
Cohesion	-0.08	0.12	0.56*	0.01	0.04
Conflict	-0.16	-0.21	-0.50*	-0.46	0.16
Control	0.20	-0.20	-0.25	0.26	0.18
Expressiveness	-0.46*	-0.33	0.44	0.06	-0.33
Organisation	-0.06	-0.30	0.26	0.09	-0.13
Independence	0.08	-0.18	0.11	0.06	-0.09

* p< 0.05

There were no significant correlations between the fathers' ratings on the FES and any of the measures of adjustment above. Mothers' and fathers' ratings on the FES were strongly correlated for cohesion (r=0.53, p<0.05), conflict (r=0.63, p<0.01), independence (r=0.50, p<0.05) and organisation (0.77, p<0.01), but not for achievement (r=0.25), control (r=0.34) or expressiveness (r=0.38).

Therefore, whilst family cohesion was positively related to child psychological adjustment and conflict was negatively related to child psychological adjustment, for the mothers' ratings, no associations were found for fathers' ratings.

Research Question Three: How do children with EB view their illness?

What coping strategies do children with EB use to manage the difficulties they experience?

Interviews with the Children

The tape recorded interviews with the children over 5 were transcribed and analysed. Out of 21 children aged 5 or over, one mother did not want the interview recorded and for one child the tape was too faint to be used. Four children, aged 5, 6, 6 and 14 either refused to complete a part of the interview or gave replies that were inadequate or incomplete and hence they could not be included in all of the analyses.

Knowledge and Understanding of EB

All children were asked about the name and type of skin disease they had. They were also asked about the cause of the fragility of the skin, the treatment they used and how they had contracted this condition. These questions were scored to give an estimate of the child's knowledge and understanding of EB and were scored from 0 - 8.

Overall, the average score was 4.76. For the children with dystrophic EB (n = 7) the mean score was 5.63. For the children with EB simplex (n = 10), the mean score was 4.0. This was despite the fact the children with EB simplex were on average older (mean age 9 years 6 months) than the children with dystrophic EB (mean age 8 years 3 months). This difference was not significant, but showed a trend toward significance (t (15) = 1.7, p = 0.10). This reflects the pattern observed amongst the families of children with dystrophic EB who appeared to have greater knowledge about their condition than the families of children with EB simplex.

Whilst all the children were able to describe the treatment for their skin, only three of the children with EB simplex knew the full name “epidermolysis bullosa”, and these same three were the only ones who knew the form they had was known as simplex. Out of the seven children with dystrophic EB, six knew the name "epidermolysis bullosa" but only three knew that they had a type known as dystrophic. Three of the children with dystrophic EB and four of the children with EB simplex knew that EB could be passed on through the family, or that it was a genetic condition.

There was a small correlation between the age of the child and their knowledge score (r = 0.38) but this was not significant (p = 0.13).

Table 4.9 shows the responses the children made about the worst three features of having EB. Three features accounted for 60% of all the replies. These were pain (22%), restrictions on activities (20%) and feeling self conscious or embarrassed (18%).

Table 4.9 The Three Worst Things About Having EB

	Total	(%)	Worst	Second Worst	Third Worst
Pain	10	(22)	4	5	1
Restrictions on Activities	9	(20)	2	4	3
Self Conscious/Embarrassed	8	(18)	5	3	0
Feeling Wary or Cautious	3	(7)	1	0	2
Lack of Understanding	2	(4)	1	1	1
Blisters Spreading	2	(4)	1	1	0
Name Calling/Bullying	2	(4)	1	0	1
Hospital	2	(4)	0	0	2
Unfairness	1	(2)	0	0	1
Tongue Tied	1	(2)	0	0	1
Worry About Exams	1	(2)	0	1	0
Nothing	4	(9)	0	0	4
TOTAL	45		15	15	15

Examples of some of the main categories are given below.

Pain: "When it stings and hurts."

> "Popping them... because when you take it out and turn it round, it hurts so much."

Restrictions on Activities:

> "You see the other children run about and have all this fun, you know, and I know that I can't get up and join in, and you know that sometimes, it's a bit, it's a bit horrible."

Feeling self-conscious or embarrassed:

> "I mean some people say we look scabby, and horrible things, and other people just look, like you're really weird or something."
>
> "Having scars on my ankles form blisters. I find them embarrassing and people make comments."
>
> "I don't like limping when I'm walking down the road. And sometimes you see people looking, you know, what's wrong with her? I think that's the worst thing."

Feeling wary or cautious:

> "I mean, whatever you do you always have to go back and think before you do it, 'Could this be a problem?' and obviously sometimes you make mistakes and that's when things go wrong. But you really do have to be careful."
>
> "Having to worry about it. Even now, if you're going to do something now with a risk of pain, of hurting yourself then you know, it's a worry. Running I find a bit scary because I always think I'm going to fall over. And take the skin off my hand."

Lack of understanding:

> "My games teacher at school, we did have several kind of arguments really, about it. Because she just won't understand... She makes me play in games, and gives me dirty looks and tries to make me feel really small."

Coping Strategies

The strategies used by children for the three standard stressors were coded using the scheme described by Ryan-Wenger (1992). For the purpose of simplicity, the main strategy used is analysed in Table 4.10, although some children did describe more than one strategy for one stressor.

As is apparent from the table, different strategies dominated the responses for the three different stressors. For example, for the pain stressor the predominant strategy used was stressor modification, followed by self-controlling activities. However, for the disappointment stressor, the predominant strategy was clearly behavioural distraction. Finally, for the teasing scenario, the predominant strategies were social support and cognitive avoidance. This marked difference in the strategies used by children reflects the importance of the stressor itself in the determination of the strategy used.

Table 4.10 Coping Strategies Used in the Three Standard Situations

Strategy Used	Pain	Disappointment	Teasing
Aggressive Activity	0	0	1
Behavioural Avoidance	1	0	0
Cognitive Avoidance	0	0	3
Behavioural Distraction	0	9	1
Cognitive Distraction	1	0	0
Cognitive Restructuring	0	1	2
Self Controlling Activities	4	0	0
Social Support	1	0	6
Stressor modification	6	0	0
No Strategy	0	3	1
Other	2	2	1
TOTAL	15	15	15

The strategies used by the children for the stressors due to EB are shown in Table 4.11. Because of the influence of the type of stressor on the strategy used, these results are presented according to the type of stressor. As can be seen for the pain stressor, the strategies used are very similar to the ones nominated for the standard scenario about pain and stressor modification is the most common coping strategy.

This study demonstrated that the type of coping strategy reported by the children was largely determined by the type of stressor nominated for both the standard stressors and the stressors related to EB. This reflects the importance of context on coping and emphasises the need to ensure that coping strategies are investigated for specific stressors, rather than as a general style.

Table 4.11 Coping Strategies used for the Stressors due to EB

Type of Stressor	Strategy	Frequency
Pain	Stressor Modification	8
	Behavioural Distraction	1
	Nothing	1
Restrictions On Activities	Behavioural Distraction	3
	Cognitive Restructuring	2
	Social Support	1
	Cognitive Distraction	1
	Nothing	1
	Uncodable	1
Self Conscious/ Embarrassed	Aggressive Activities	2
	Nothing	2
	Cognitive Avoidance	1
	Cognitive Restructuring	1
	Behavioural Distraction	1
	Social Support	1
Wary	Endurance	1
	Cognitive Avoidance	1
	Nothing	1
Lack of Understanding	Cognitive Restructuring	1
	Nothing	1
Blisters Spreading/ Unable to burst	Endurance	1
	Stressor Modification	1
Name Calling/ Bullying	Aggressive Activities	1
	Behavioural Distraction	1
Hospital	Social Support	1
	Behavioural Distraction	1
Unfairness	Endurance	1
Tongue-tied	Nothing	1
Exams	Cognitive Problem Solving	1
TOTAL		41

CHAPTER FIVE

THE PSYCHOLOGICAL IMPACT OF EPIDERMOLYSIS BULLOSA

DISCUSSION

Summary of Results

This study found that there was no significant relationship between the severity of the child's EB as rated by his or her parents and the child's psychological adjustment or the level of psychological distress reported by the mother. However, there was a relationship between the severity of the child's EB and the level of distress reported by fathers. The study found some support for a link between family environment and the child's psychological adjustment, which has been shown in other studies of children with a chronic illness (Drotar, 1997).

The children with EB nominated the three worst things about EB. These were pain, restrictions on activities and feeling self conscious or embarrassed. The coping strategies used by the children for both the standard stressors and the stressors associated with EB that they had nominated, were shown to be strongly related to the nature of the stressor itself.

Evaluation of Results

The relationship between severity of EB and psychological adjustment

This study showed there was no relationship between the severity of the child's EB and the child's psychosocial adjustment or self esteem. There was also no relationship between the severity of the child's EB and the mother's level of psychological distress. This is in keeping with the majority of studies of chronic illness in childhood (Wallander and Varni, 1998).

However, there was a strong relationship between the severity of the child's EB, in terms of the amount of their body affected and the overall severity rating, with the level of distress in the fathers. There are now several studies that illustrate that different aspects of having a child with a chronic illness are perceived as stressful by mothers and fathers. For example Dahlquist, Czyewski and Jones (1996) found that fathers' distress was related to the child's health status and did not reduce over time. Sloper (2000) found that the mother's appraisal of the strain of the illness, and her own ability to deal with this were related to mother's distress, but that employment problems and frequency of hospitalisation were related to father's distress.

The results from this study have to be interpreted cautiously given the small sample size involved. The power of the study was limited by the small sample size and it would therefore only be possible to detect differences representing moderate to large effect sizes. In addition, the ratings of severity were based on parental report (rather than being rated by a nurse or doctor) and the measures of severity used were constructed for this study and therefore not standardised. This must be taken into account when interpreting these findings. Nonetheless, this study is consistent with other studies that have found a difference between the effects of severity of illness on mothers and fathers.

The relationship between family functioning and psychological adjustment

The results from this study are consistent with results from other studies of children with chronic illnesses. Drotar's (1997) review of the relationship between family variables and child function showed that several studies identified a similar pattern of family cohesion being related to good adjustment in the child and conflict being related to poor adjustment.

However, some differences between mothers' and fathers' ratings did emerge. There was no relationship between the fathers' rating of family functioning and child adjustment, self esteem or parental distress. Given the small numbers of fathers included in this study, it may not have been possible to detect a significant relationship between adjustment and family functioning in this study.

Mothers rated their family as higher on cohesion on the FES than mothers of families in the general population. This has also been shown to be the case in other studies of children with a chronic illness (e.g. Midence, McManus, Fuggle & Davies, 1996). Mothers also rated the family as lower on the control scale of the FES than the norms. This scale reflects the use of rules and set procedures in family life so a lower score indicates greater flexibility about rule setting within the family.

However, there were no differences between the fathers' ratings on any of the FES sub-scales compared to the norms for this measure. Fathers have not been included in most studies of children with chronic illness, so less is known about their perception of the effects on the family. The FES does not provide separate norms for mothers and fathers because no gender differences were found in the studies on which the norms are based, and this has been supported by further studies (Vostanis & Nicholls, 1995). In this study there were high correlations between mothers and fathers ratings on the FES for four of the sub-scales (cohesion, conflict, independence and organisation) but not for the remaining three.

Most studies, like this one, are cross sectional in design and it is not possible to determine the direction of causation. In other words, does family functioning predict adjustment, or is it a consequence of adjustment to the illness? The few longitudinal or predictive studies that have been carried out give somewhat conflicting results. In the study by Varni, Katz, Colegrove and Dolgin (1996) they were able to show that cohesion and expressiveness measured soon after diagnosis for cancer did predict child adjustment up to 9 months later. However, Gustafsson, Bjorksten and Kjellman (1994), who used a different measure of family functioning (The Family Adaptability and Cohesion Evaluation Scales (FACES): Olsen, Sprenkle & Russel, 1979) found that the family dysfunction they observed occurred as a result of the onset of physical symptoms, rather than the other way around.

In addition, very few studies include informants other than the mother and at least some of the association found between family functioning and child adjustment may be due to shared method variance because of the reliance on the mother's report for both family functioning and child adjustment.

The measure of cohesion in the FES has been criticised within the literature because of the possible overlap between two separate dimensions. One of these is "closeness-caregiving" which reflects the level of positive, mutual support within a family (Green and Werner, 1996). The other is "enmeshment" or "intrusiveness" which could be seen as potentially limiting, if this reflects unnecessary restrictions on a child's individuality or independence. Cohesion, as measured in the FES, could potentially confound the two separate concepts of "closeness-caregiving" and "intrusiveness" which reflect these different aspects of cohesion.

The FES has also been criticised because of the low levels of internal consistency found in some studies for some of the sub-scales (Roosa & Beals, 1990). It can also be problematic to interpret because of the variability in perceived normality of family functioning across different cultures and different age groups (Casella & Kearins, 1993; Kazak, McCannell, Adkins, Himmelberg & Grace, 1989). Nonetheless it is still the most widely used measure of family functioning (Piotrowski, 1999) and has been shown in some prospective studies to have some predictive validity for adaptation to chronic illness (Varni, Katz, Colegrove & Dolgin, 1996).

Levels of psychological distress in parents of children with EB

The level of psychological distress, as measured by the GHQ, indicated that, on average, the level of distress reported by the mothers of children with EB was about the same as that found in the general population (Cox et al., 1987). Although, the level of distress reported by the fathers was higher than in the general population, this difference was not significant. Both the direction of this gender difference and the relatively low level of distress are unexpected findings. Most studies do demonstrate higher ratings of distress amongst mothers of children with a chronic illness compared to fathers (Mastroyannopoulou et al. 1997; Silver, Westbrook & Stein, 1998; Sloper, 2000; Timko, Stovel & Moos, 1992). However, Sloper and Turner (1993) also reported higher rates of distress among fathers. They interpreted the difficulties the fathers encountered with practical care tasks, such as feeding, as due to the fact that they were less familiar with the task and had not had to come to terms with the difficulties in the way the mothers had.

This was a very small sample and these findings may be the result of the characteristics of this particular sample. In addition, the GHQ focuses on symptoms over the last few weeks, and the respondent has to compare these with their "usual" state so it does not accurately reflect more long term or chronic symptoms. The GHQ is a global measure of psychological distress and does not measure the adjustment to the child's illness specifically. It will therefore be influenced by many other stressors as well as coping with a child with a chronic illness. The design of this study does not make it possible to separate out these other factors in order to look just at the impact of the illness itself.

Psychological adjustment of children with EB

The PARS was used in this study because it measures child psychosocial adjustment, rather than psychiatric symptoms or disorders. In addition it does not contain any somatic items unlike other commonly used measures, such as the CBCL (Achenbach & Edelbrock, 1993). Using the PARS meant that it was possible to measure the associations with severity and family functioning against a broader measure of adjustment, rather than presence or absence of psychiatric symptoms. However, the comparisons with the norms for the instrument, which are based on children with chronic illnesses, do not provide information on overall levels of psychiatric problems. The children with EB were rated as more independent (by both fathers and mothers) and as less withdrawn and less depressed (by fathers only) compared to the norms. This is surprising considering the disabilities of many of the children, but might be explained by the characteristics of the samples on which the norms for the measure are based, which include children with severe medical conditions e.g. cardiac anomalies, cerebral palsy and seizure disorders (Stein & Jessop, 1990).

There have been some criticisms in the literature about the PARS. For example, a study by Harris, Canning and Kelleher (1996), which compared the CBCL, the PARS III and the Columbia Impairment Scale (CIS: Bird, Shaffer & Fisher, 1993) with diagnoses obtained by the Diagnostic Interview Schedule for Children (DISC-2: Shaffer, Fisher, Piacentini, Schwab-Stone & Wicks, 1990) demonstrated that the PARS had low sensitivity, particularly for identifying children with internalising disorders. Given that the meta-analysis carried out by Lavigne and Faier-Routman (1992) identified that in this population, emotional disorders are more prevalent than conduct disorders, this may result in the under recognition of emotional problems.

Self Esteem

The Self Perception Profile scores illustrate that the children with EB had lower self esteem than the norms for this measure on the specific scales relating to athletic ability and physical appearance, as might be expected for this particular condition. However, the global self esteem scores were about the same level as the norms. This suggests that the children may have been able to "compartmentalise" the different aspects of their self to some extent, limiting the impact of EB on their global self esteem. Given the SPP can only be used with children over eight, the number of children in this study was very small and these results must be seen as tentative. In addition, the norms used here were based on Radcliffe et al.'s (1996) composite norms calculated from the norms originally given by Harter (1985). These had to be used because of the small numbers of children in any particular age group in this sample. Also, Cramer (2000) points out the difficulties with relying on self report of self esteem because of the influence of experimenter demand effects as well as the possibility of defence processes influencing the child's response. Therefore, although it is encouraging that these children reported self esteem scores that were not significantly different from the norms for the global self esteem measure, these results have to be interpreted cautiously.

Differences between the different types of EB

As would be expected, the two different types of EB, simplex and dystrophic, had different effects on the children. EB simplex caused more functional disability, although it was rated as less severe than dystrophic EB in terms of how much body area was affected. Disability has been shown in some studies to be an important influence on adjustment to illness, independently of other measures of severity (Cadman et al., 1987; Silver et al., 1998). However, no association between disability levels and child adjustment was found in this study.

Children with dystrophic EB had lower self esteem scores on the SPP than children with EB simplex. Although this difference was not significant in this study, this may be partly due to the small numbers in each group who were old enough to complete the SPP.

Knowledge of EB

The findings about the children's knowledge about their EB revealed that the children had quite limited understanding of their skin condition. Although all the children were able to describe the effects of the EB on themselves and the treatment, a high proportion did not know about the different forms of EB or have a good understanding of the genetic transmission of EB. There was a trend towards older children having a greater understanding of their condition, but this was a very small sample and any results have to be understood in that light. Many of these children did not receive medical attention regularly for their EB, because the condition was fairly static, and many of the families reported how limited the understanding about EB was in the family as a whole.

Early studies on children's understanding of illness did suggest that the child's acquisition of knowledge followed Piagetian developmental stages (Bibace & Walsh, 1980). However, it has since been shown how important experience is in determining the acquisition of knowledge and that young child can gain a higher level of understanding if the knowledge is relevant and serves a purpose for them (Paterson, Moss-Morris & Butler, 1999). Rushworth (1999) reviewed studies investigating children's understanding about illness and showed that early studies had therefore probably underestimated the ability of children to develop understanding about their illness. Newer theories incorporate Vygotsky's (1962) model rather than a Piagetian model, which proposes there is more of a range of abilities that a child can be expected to achieve at any particular age group, depending on their experience and the way in which they are assessed. Hence the children in this study had very little need to know the type of EB they had, or to use the full name for the condition, and this is reflected in their scores. However, they did understand about the treatment because this was a constant part of their lives.

Coping with EB

The findings from the coping study illustrate the importance of the stressor itself in determining the appropriate coping strategy rather then the child's own coping style or preference. This was apparent when the responses to three standard stressors (pain, disappointment and teasing) were analysed which revealed that different strategies were used for the three types of stressors. It is likely that the strategies described to cope with the situations nominated by the children as the worst aspects of EB, would probably be related to the type of stressor as well, although the small numbers involved meant it was not possible to analyse these in detail. However, the strategies used for the three main features of EB nominated by the children (pain, restrictions on activities and feeling self-conscious) did reveal different strategies for the different types of stressors. It was apparent that the majority of strategies described by the children used to cope with the pain associated with EB were the same as those when presented with the pain scenario as one of the three standard stressors.

This has also been demonstrated in a study by Spirito, Stark, Gil and Tyc (1995) and has important implications for the study of coping with chronic illness in children. This supports the argument put by Lazarus (1993) that coping is a style rather than a trait and that it is not possible to generalise across situations in a meaningful way. He suggests that one way forward is to explore coping strategies within the individual across different situations, which may also prove to be a more successful way of helping someone to alter their coping strategies if they have specific difficulties in any particular situation. Tennen, Affleck, Armeli and Carney (2000) describe methodology that they have developed which uses a within subject design rather than a between subjects design. However, this too has its limitations because of the likelihood that reporting coping strategies over several occasions may well influence the strategies under study.

Given the clear influence of situation on the type of strategy used, it is important to consider what features of the situation influence the strategy. Band and Weisz (1988) conclude that the controllability of the situation is an important determinant of the type of stressor. If it is possible to alter the stressor itself then under most circumstances a child will prefer to do this than to alter the effects of the stressor. This would be consistent with the use of stressor modification strategies in the pain scenario used in this study, when the most frequently cited strategy was to take painkillers to reduce the pain. It would be possible to relate how controllable a stressor is perceived to be by a child to the type of strategy used to tackle it.

It was possible to code the replies given by children both to the standard stressors and the self nominated stressors associated with EB, according to the multi-dimensional descriptive coding scheme developed by Ryan-Wenger (1992). High inter-rater reliability was obtained indicating that this coding scheme is reliable using the simple descriptions of categories given by the author. However, at times it was difficult to determine a single major strategy and some children described several strategies for a single stressor. For example, when describing her response to the teasing scenario, one child replied,

> "Well, I think if I was being teased I would probably cry. And then my friends would give me a cuddle and then that would cheer me up...They try and tell awful jokes to try and make you feel better, and in the end you end up laughing because it's so bad."

This reply indicates three strategies - ventilating emotion, seeking support from others and finally distraction. Whilst a judgement can be made about the main strategy used, just recording one strategy does not fully reflect the complexity of the child's report of coping.

The quality of the replies given by children varied enormously, from children who were very articulate and willing to discuss what were often distressing topics, to children who found the experience much more difficult and were reticent, and from whom it was difficult to obtain full responses. As noted above, four children refused to participate in this task or gave replies that made it clear they were not fully participating. The child's ability to reflect on his or her own coping style is essential if the reported strategy is likely to be strongly related to the actual strategy used in a given situation. These results may therefore be limited by the child's limitations in reflecting on their own behaviour, and then reporting this to someone else (Cramer, 2000).

Limitations of the Study

There are several important limitations to this study. Firstly, the sample size was small, due to the rarity of EB, and this has limited the power of the study and the conclusions that can be drawn from the results. In addition, given the limited knowledge about the prevalence of different types of EB it is difficult to determine how representative the study is and to what extent these findings can be generalised. All the study participants were members of DEBRA and this sample may not be representative of the wider population of children with EB. For example, people often join support groups such as DEBRA because of a level of concern about their condition that may not be present in the population as a whole. For example, the sample does not reflect the ethnic diversity in the population as a whole. Only one mixed race child was included in the sample and the remaining children were of white British origin and this may be an indication of a selection bias. However, it may possibly reflect the genetic distribution of EB within different ethnic groups, but there is not enough information about the distribution if EB in the population to determine whether this is the case.

There were three families included in the research who had two children with EB, which would be expected given that EB is a genetic condition. If data from both children were used, this would have resulted in non-independent data. Therefore it was necessary to use only the data from one child in the analyses that combined data from both the mother and child in order to reduce the impact of this non-independence on the results. However, this does not overcome the problem totally because it would be expected that a mother with two children with EB would experience a higher level of stress than a mother with one child and this could not be taken into account in the analyses.

No control group was used in this study. In their meta-analysis of this field, Lavigne and Faier-Routman (1992) identified that studies which used comparisons with test norms showed larger effect sizes than those that used ad hoc control groups based on matching only a few variables and argued that small control groups may sometimes be misleading. From previous studies in this field we know a considerable amount about the impact of chronic illness in children compared to healthy children, and the primary question becomes how we explain the variability in the response of children and families, rather than repeating research on these differences (Pless & Nolan, 1991). However, there are limitations in relying on the study norms as well, in that many of the measures used e.g. Family Environment Scale and Self-perception Profile have norms based on American children and these are not necessarily appropriate for English children.

Three families dropped out of the study after having consented to take part. These three families were all experiencing considerable difficulties, which was the reason they withdrew from the research. It is therefore possible that the study did not include some of the most severely affected children and families, which will have influenced the results of the study.

CHAPTER SIX

A QUALITATIVE STUDY OF THE EXPERIENCES OF MOTHERS OF CHILDREN WITH EB

METHOD

A semi-structured interview was used to elicit the mother's experiences of having a child with EB from the period immediately after her child was born to her current concerns (Appendix 6). This methodology was used partly because of the difficulties referred to by Lansdown and Nabarro (1990) in their study of families with a child with EB. They suggested that whilst their study had found that there were no overt psychological difficulties among children and families with EB, this did not reflect the high level of concerns within families. Therefore, in-depth interviews were carried out in order to gain detailed descriptions of the way in which caring for a child with EB had affected the child's mother. In addition, it was felt that this methodology offered the opportunity to understand some of the psychological processes that underlie adaptation to a chronic illness, which is particularly important for rare conditions when sample sizes are small and quantitative methods may be limited (Eiser & Twamley, 1999). This qualitative study therefore complemented the previous study by providing more information about the experiences of the mothers from the participants themselves.

Participants

All the mothers who took part in the previous study were also included in this study. A description of the participants can be found in Chapter Three. However, one mother did not consent for the interview to be tape recorded and therefore this study is based on a total of 23 mothers.

Method

The interview lasted approximately one hour to one and a half hours and was carried out in the family home. It was audio taped and then transcribed. The interview was semi-structured and covered six areas: information about the family and history of EB in the family, current management routine for the child's skin, the mother's initial reaction when the child was first born, her current strategies for coping with EB, other psychological difficulties experienced by the child and the impact of EB on family life (Appendix 6). The interview questions were chosen to encourage the mother to talk about her own experiences and to reflect on the way that these had developed from the time that the child had been born to the current time. The initial section was more factual and structured and progressed to a more open-ended format to encourage the mother to talk more freely about her own experience.

The transcripts of the interviews with the mothers of children with EB were analysed qualitatively, using interpretative phenomenological analysis (Smith, Jarman & Osborn, 1999). An example is shown below of this procedure as it was applied to one section of one transcript.

The first stage of this involved reading and re-reading all the transcripts to become familiar with the material. Then, for a smaller number of transcripts (initially eleven), comments and initial concepts were noted onto the left side of the margins of transcript. These comments included associations, connections, summary comments and some interpretations. As this study involved 23 transcripts, this was done at quite a broad level because of the large number of transcripts involved so each comment or association related to a larger part of the transcript than would be the case for a single case study.

The next stage involved identifying key words and possible theme titles. This is usually done by re-reading the transcripts and using the right side of the margin to list emerging theme titles and identifying key words to summarise the data in that part of the text. However, because this study involved a large number of transcripts, this stage was done by copying the comments from the first stage onto a summary sheet for each transcript, so it became possible to look across the different transcripts for clusters or connections between the emerging themes and ideas. At this stage four initial "themes" were identified. In order to ensure that these themes were an accurate reflection of the material in the transcripts, it was then necessary to go back to the original transcript to identify examples and illustrations of each theme. At this stage one of the original themes (mother's containment of her child's distress) was dropped and another theme was identified (diagnosis phase).

Having checked these throughout the small group of transcripts, it was then necessary to go back through all 23 transcripts with a "more focused lens" (p 231, Smith et al., 1999) to identify all the extracts that were relevant to the four themes, and to ensure that the themes adequately reflected the data in the transcripts. It was then possible to clarify the sub-themes and the connections between the four identified themes. The four themes are presented in Table 7.1 with the sub-themes listed next to them.

Example of Analysis

In order to illustrate the methodology used an extract from one transcript is shown below. This shows part of the initial transcript together with the notes in the left hand margin that indicate important ideas or concepts illustrated in the text. The passages relating to these notes are underlined in the text. This extract is taken from an interview with a mother with two children with EB Simplex. This section of the interview asked about the emergence of difficulties and what sort of support was or would have been helpful at that time.

Extract from Transcript 17

	Mother: When she started walking I suppose, actively, from the age of about two, two and a half onwards, it was just…it started.
	Interviewer: And do you think that was the most difficult period?
School years difficult	Mother: No. The most difficult period has always been the school years. Um, for both of them definitely.
	Interviewer: What's been difficult about the school years?
Preventing blisters - hard to control when older Letting go	Mother: I think because they were, you see, up until the time when they were, well when they were still at home, I could control it, it wasn't very hard. I could stop them walking, I could stop them getting bad. But once they go off to school, all sorts of things happen at school in the course of the day. Repeatedly they come back, you know, lame as anything and I've got to keep them home a few days and so it's really when they're out of my care.
	Interviewer: Thinking back to those early years, would you, I don't know if you did have any support. Did you know any other families with EB?
Wanted contact with EB family Acceptance	Mother: No. I didn't. I've always wanted to, yes, I would have like to have met another family. Just to feel less isolated. Um, but I've never really looked for any help. Because I've always accepted point blank that there isn't any, there isn't anything that you can do about it, you've just got to get on with it…So I wouldn't look for any sort of miracles…
	Interviewer: And then when you say that going to school was difficult because they got worse, was it also difficult to get them to accept it at school?
Lack of understanding	Mother: Very difficult. Yes, very difficult. I think the lack of understanding it like they said, to me it's always been the biggest problem. Other people's perception of it. And trying to get it across to people, and it's like a constant battle…

Because of the relatively large number of transcripts in this study, it was necessary to copy key ideas from each transcript onto a separate summary sheet in order to be able to look across transcripts. The summary sheet for this case is shown below. The section of the summary sheet that relates to the above extract is underlined. The themes to which the sections relate are shown in the right hand margin.

Summary of notes from Transcript 17

Initial Comments (copied from margin of transcript)	*Emerging Themes*
Wariness when child born – looking for symptoms	*Diagnosis*
Hopefulness with no.2 / disappointment	
When younger can control activity therefore easier to manage.	
Harder when older at school.	*(Different Stages)*
"Letting go"	*Normal vs Special*
Would have liked contact with EB family	
Acceptance "no miracles"	*Resolution*
Other people - lack of understanding	
- cruelty, criticism	
- minimisation "just a blister"	
Physical care - Got to "get on with it",	
Used to it because does self	*Coping with caring*
Practical and financial support with mobility difficulties	
Gender difference – worse for boys because more physical -	
Difficult to watch children struggle and suffer	
Worries re future e.g. grandchildren	
Downward comparison	*Resolution*
Frustration with "minor" mobility problems – can't they be overcome?	

To protect the anonymity of the participants, each mother was given an identifying number. Table 6.1 shows the mothers' ID together with the age, sex and type of EB of her child.

Table 6.1 Mother's ID and Type of EB in Child

Mother's ID number	Age of Child	Sex of Child	Type of EB	Family Member With EB
1	4	Boy	Dystrophic	None
2	9	Girl	Dystrophic	None
3	6	Girl	Dystrophic	Father
4	13	Girl	Dystrophic	None
5	3	Boy	Dystrophic	None
6	4	Girl	Dystrophic	None
7	8	Boy	Dystrophic	None
8	4	Girl	Dystrophic	None
9	9	Boy	Dystrophic	None
10	2	Boy	Dystrophic	None
11	14	Boy	Simplex	None
12	3, 6	Girls	Simplex	Father
13	15	Girl	Simplex	Father
14	15	Girl	Dystrophic	Father
15	3	Girl	Simplex	None
16	14	Boy	Simplex	None
17	10, 16	Girls	Simplex	Mother
18	7	Girl	Simplex	None
19	7, 9	Girl, Boy	Simplex	Mother
20	7	Boy	Simplex	None
21	10	Boy	Simplex	None
22	5	Boy	Simplex	None
23	15, 15	Twin girls	Dystrophic	Father

Qualitative researchers acknowledge that the investigators own perspective, the context in which the research is carried out and the researchers experience of the area being investigated may influence the interpretation of the results of the study (King, 1996). In order to allow the reader to take these factors into account, a statement describing these influences is usually made by the researcher, a process that is known as reflexivity.

These interviews were carried out as part of a project funded by DEBRA and although the participants knew the results would be presented anonymously, they knew that the results for the group as a whole would be summarised for DEBRA. The interviews were analysed after the author had had several years clinical experience, working within the area of chronic illness in a general paediatric setting. The author did not have any personal experience of EB, but she had worked with one family with a child with EB while training as a clinical psychologist.

CHAPTER SEVEN

A QUALITATIVE STUDY OF THE EXPERIENCES OF MOTHERS OF CHILDREN WITH EB

RESULTS

Themes

Four core themes emerged from the interpretative phenomenological analysis, and these are described in turn below. Each theme was chosen because it helped to clarify a process that could be identified within the transcripts which was relevant to the group as a whole and which was important in terms of understanding the mothers' experiences of having a child with EB. The four core themes can be seen as significant processes that were important for this group of families in adjusting to their child's EB. However, as each individual transcript varied with respect to the importance of each theme and to how it was expressed for that individual, the themes can best be understood as dimensions, and examples are given of statements which reflect different positions on the dimension for each theme.

An identifying number was given to each transcript and Table 6.1 lists the ID numbers, giving details of the child's age, sex, type of EB, and family history of EB. Each quotation is followed by a number indicating the mother's ID number, and details of the child's sex, age and type of EB. All names and other identifying details in the quotations have been changed to protect the anonymity of individual participants.

Table 7.1 Summary of Themes from Qualitative Analysis

Theme	Sub-Themes
1. Difficulty with Diagnosis	Quality of medical care
	Transmission within the family
2. Resolution of the Diagnosis	Changes in the illness
	Changes in child's development
3. Coping with Caring	Support
	Child's temperament
	Support of health care system
4. Treating the Child as "Normal" or "Special"	Other people's perceptions
	Impact on mother

Theme 1. Difficulty with Diagnosis

This dimension reflects the level of difficulty experienced by the mother from the time that the child was born until she obtained accurate information about the diagnosis and management of EB. At one end of the spectrum were those families who did not have a traumatic experience at the birth and who were easily able to obtain a diagnosis and/or appropriate information about management. At the other end of the dimension were those families who either had a very difficult initial experience because of the severely damaged physical state in which the child was born, or because of difficulties obtaining the diagnosis and appropriate advice about treatment.

It is important to remember that this dimension reflects the mother's report of her experience of this period. As a result there is not necessarily a direct relationship between actual time spent before obtaining an accurate diagnosis and the level of distress experienced by the mother. Instead, this dimension reflects the psychological difficulty as described by the mother during this period.

Some families easily obtained a diagnosis and advice about management or recognised what the condition was, and this was a very straightforward phase.

> "She was a month old, and they started popping up then. And her blisters came out then, and that's when I first noticed it. And that's when I took her to the doctor's and had it checked and found out that is what she had...Well, I knew S (husband) had it... But we knew that it was a mild strain. So, we weren't that worried." (14: girl, 15, dystrophic)

> "When she was crawling she got a blister on one of her toes when she was crawling around on the floor. I sort of spotted it and thought, well that's it, she's got it...I just accepted it. When they got really bad, eventually I did take her to the doctor just to show him and have him confirm." (17: girl, 16, simplex)

The majority of children in this category were children with EB simplex but two families with children with dystrophic EB were also in this category.

At the other end of the spectrum, several mothers had had extremely traumatic experiences and described these vividly. Children born with dystrophic EB frequently have a lot of skin damage as a result of the birth and as a consequence the child's appearance is quite shocking for many parents.

> "It was a real shock you know… They just pulled the blanket off him and it was raw, there was no skin." (7: boy, 8, dystrophic)

> "I kept breaking down in floods of tears… I can remember a couple of times sitting on the lap of the nurse and I'd cry my eyes out saying, I can't stand it, what am I going to do? And it wasn't that I was being selfish that I didn't want the baby or I couldn't be bothered. I just couldn't stand seeing her in pain." (3: girl, 6, dystrophic)

> "Those weeks were just nightmares…For a long time, for many years, it's less now but I still get, I still have nightmares about it" (6: girl, 4, dystrophic)

> "I didn't know what to think. In the back of my mind I was thinking it was something dreadful. It had to be something dreadful. My child was going to die. And when I got back home I wouldn't talk to anybody for a week. You know, people were phoning "How's the baby?" and you know there's this lovely little baby that you think is going to die. You just feel totally, totally lost." (5: boy, 3, dystrophic)

Quality of Medical Care

In addition to how traumatic the first symptoms were for the mother, there was a lot of variation in the manner in which the family received the diagnosis and information about management of EB. At the "straightforward" end of the dimension were those families who were fortunate enough to have someone diagnose the condition accurately themselves, or who made the appropriate contact with DEBRA, Great Ormond Street Hospital or St Thomas' Hospital. In these cases, children with severe EB were diagnosed within days, and offered support from specialist clinics immediately.

> "When you listen to other mum's who couldn't get their children to specialist hospitals like Great Ormond Street...you realise how lucky you were. That she just suffered like that for two days. Because once we got to Great Ormond Street it was really different." (3: girl, 6, dystrophic)

However, several of the families had very difficult experiences of the medical care they received. The appearance of a child with severe EB is not just shocking for parents but also for some professionals, because most will never have come across EB before. The difficulties experienced by some professionals were sometimes apparent to the mothers.

> "Then she started to suck her fingers when she was hungry and came up in blisters. And they were horrified in special care. They were really horrified. They gave us a social worker who used to go into hysterics. I asked her not to come anymore. She was useless, 'How dreadful, how dreadful, I feel funny' she used to say". (23: girls, 15, dystrophic)

> "The nurses drugged her at the hospital... because they couldn't stand the screaming. I mean one of the sisters said, 'We nurse sick children but they're not really in any pain' and it was quite a shock to the nurses." (8: girl, 4, dystrophic)

EB is so rare that most families had to wait to get a referral to the DEBRA specialist nurses or an EB specialist at St Thomas' Hospital or Great Ormond Street Hospital before receiving correct information. As has been reported in many other studies, several families felt they were managed very badly by the medical system that treated them in an inconsiderate way. The trauma that some of these mothers reported clearly had an impact on themselves and sometimes left them with unresolved anger or suspicion of the medical system.

> "I felt extremely isolated and alone. And I felt ... very much some kind of case for the record books, the way I was treated... all of a sudden the doctor left the room, called in another one and then they called in another one. I was like a freak in a show, you know. Everybody would come in and look. They weren't talking to me, they were talking about me. And no one asked me how I felt, no one minded." (19: boy, 9, simplex)

> "I heard the doctors coming down the corridor. And they were talking about this poor child that had been born with what they thought was Harlequin's disease. And I was thinking 'Oh, poor thing'. Little did I know it was my baby they were talking about outside the door." (4: girl, 13, dystrophic)

Some families also remembered being given very misleading information. Understandably, this often scared parents and they could give vivid descriptions of their experience several years later.

> "And we had a friend take us down (to London). We walked in and we saw a very, very young doctor. He stripped R down and he looked at R and he said, 'Oh, he's got Epidermolysis Bullosa. He'll be dead by the time he's two.' And that was all he said to us. And we both, D (husband) and I came out and we were devastated. We didn't know what to do. I cried. And D went very quiet". (9: boy, 9, dystrophic)

> "He wrote it down on a piece of paper. He told me that it was something that was, in all the medical textbooks, that was very rarely seen. Um, that there was nothing that he could do for T. He would never be able to take part in normal life and sport. He would always have difficulty walking... he really painted quite a bleak picture." (16: boy, 14, simplex)

> "And there's me a wreck, having been told, after waiting for two and a half days for somebody who could tell us what the problem was. Um, I was very, very

> upset. And having been told in that way, imagined the absolute worst." (5: boy, 3, dystrophic)

Knowledge about how to manage EB within the medical profession is quite limited and some children were clearly managed inappropriately, despite knowledge that they may have EB. This mother knew her twins were at risk of EB because the father had a dominant form of EB and despite ensuring she had the babies at a hospital where there was some knowledge of EB, felt they were inappropriately managed.

> "They were forceps. Can you believe? I mean I can't believe that I let them do it. Talk about ignorance is bliss... They should never have done that...I put my whole trust in them, they had all the knowledge. They should have had the answers...I just couldn't believe it. I gradually realised, you know, that I had to take this on myself, because they know nothing in reality." (23: girls, 15, dystrophic)

There were also examples of some families who felt that their GP or local paediatrician did not take their concerns seriously enough, and who found it very difficult to access specialist advice.

> "I didn't think it was normal. And they just kept saying that her skin would toughen up, and I couldn't make them understand ... And I'd take her down to the GP's and I would say, 'Look, she got blisters from walking on a carpet in bare feet, indoors.' You know, you don't get blistered feet from walking round on a carpet... I just didn't get any sort of help." (18: girl, 7, simplex)

The worst example of this was of a boy with dystrophic EB, who was treated by the local paediatrician, who had not been able to diagnose EB and who had not referred the child to a specialist dermatology clinic until he was four. However, for one period of six months when he was about 18 months old, he had been kept as an inpatient in hospital because she felt this was the most appropriate treatment for him.

For the majority of families, the specialist help provided by DEBRA, in particular the specialist nurses, was invaluable. They were able to offer both specialist knowledge of how to manage EB and emotional support to families who were very emotionally vulnerable at this stage.

Transmission within the family

None of the mothers of children with a recessive form of EB, or whose EB was caused by spontaneous mutation, had prior knowledge of the condition because it is so rare. The extent of knowledge about EB in the other families was very variable and often very limited. There were five mothers who had a partner who had EB, whose knowledge of EB was through him and his family. There were two mothers who had EB themselves and so had direct experience of the condition. Surprisingly, having prior knowledge about EB did not necessarily mean that this phase was an easy one. Within the group of mothers who did have some knowledge of EB beforehand there was considerable variation as to how difficult the mother found the diagnosis phase.

Several of the mothers with a partner with EB talked openly about the guilt the father experienced as a result of passing on EB and whether they thought this had affected the father's relationship with the child. One mother described how the father absolutely refused to be involved in the child's care, despite obviously having to manage his own blisters, which she attributed to his guilt.

> "I became quite obsessive about it. Because I'm like that...I think the more I became like that, the more guilty he felt. Because he'd never talked about it. And he never wanted to talk about it". (12: girls, 3 and 6, simplex)

Another mother described how she felt the father's family resented her opening up old wounds by exploring the way EB had been passed on within the family.

> "They knew the name. As to how it was caused they had no idea. They thought some really peculiar things. And I think they resented this person coming into the family from the outside...because I wanted to know". (23: girls, 15, dystrophic)

One of the two mothers who had EB herself did not know what her condition was called, nor that it was a genetic condition. She was only diagnosed herself when her first child was diagnosed at about one year, and also had a very difficult early experience.

> "I didn't know what could be done. It wasn't a very rosy future at all. It was quite bleak really...I used to keep him very restricted. We'd leave him in a pushchair. And I've never really let him play..." (19: boy, 9, simplex)

Some of the mothers did not feel prepared for the impact that the EB would have on a baby or child that is very different from the impact on an independent adult. Although they did not find the diagnosis period difficult, they subsequently had difficulties when they realised the implications of the child's EB.

> "I was upset and I was disappointed that she had it. But I still didn't think that it was going to be as bad. And it was the summer after she started to walk. Her feet got awful. Then I realised how terrible this was, you know. It was going to affect the rest of your life." (12: girl, 6, simplex)

> "The fact that she had a few blisters when she was little, didn't really bother me. It was when it started affecting her, when she was trying to do things, wanted to do things...when she was with the other children every day, and wanting to do the things that they could. Even things like walking home from school. And that's when it started to hit me."(13: girl, 15, simplex)

Theme 2. Resolution of the Diagnosis

This theme reflects the process by which the mother has made sense of and adapted to having a child with EB and incorporated the demands of EB into her lifestyle. At one end of this dimension the mothers could be described as "resolved". Whilst these mothers may have experienced considerable emotional turmoil during the diagnosis phase, they now had a way of managing the difficulties they experienced and had a realistic and constructive viewpoint. These mothers did not deny difficulties but demonstrated realistic expectations of their child and themselves. At the other end of the dimension, the mothers could be described as "unresolved". For these mothers, the emotional impact of the child's EB was close to the surface during the interview, and they sometimes continued to experience considerable emotional trauma. They felt unable to accept the difficulties caused by the condition and continued to experience emotional distress.

This theme overlaps with the concept described by Pianta and Marvin (1992) as "resolution" of a child's diagnosis. Pianta and Marvin (1992) describe the time when a child is diagnosed with a serious illness as a "crisis for a parent, which threatens (destabilises) their view of themselves as parents, and parenthood". Following the crisis of diagnosis, resolution is the process by which the parent adjusts to the child's illness. Resolution is defined by Pianta and Marvin (1992) as,

> "the process of the integration of this information/emotion within the parents' representational systems of themselves as parents, of their child, and their relationship with their child. The cognitive task of resolution is for the parents

> mental representational systems to integrate this information about the child and incorporate it without distorting reality". (p 3)

However, this is not limited to the time of diagnosis and can rather be seen as an ongoing process;

> "Resolution is viewed as a process and the extent of resolution will vary from time to time depending on the child's and parent's circumstances, as well as developmental history." (p 4)

Pianta and Marvin (1992) define two discrete categories of reaction to a diagnosis: "resolved" and "unresolved". However, it was evident from these interviews that "resolution" could be best understood as a dimension with the two categories of "resolved" and "unresolved" representing the different ends of the dimension. Both the content and the emotional tone of the interview provided important indicators of resolution or non-resolution.

At the resolved end of the spectrum were mothers who acknowledged some difficulties and who may have experienced considerable emotional turmoil but who now had a way of incorporating the difficulties encountered by themselves and/or their child into a realistic and constructive viewpoint. Whilst not minimising the difficulties encountered, the mother did not continue to feel traumatised by her experience, and some could even reflect on some positive aspects of the experience. She was able to show realistic expectations of her child, whilst not denying possible difficulties in the future. For example, this "resolved" mother, who had a child with recessive dystrophic EB, described an extremely difficult and traumatic reaction when her child was first born.

> "I didn't do anything else but cry. I think I cried solidly for about, it must have been for about four days. I don't think I stopped crying...and I kept thinking 24

> hours and our life had just fallen apart. Um, we just didn't know what was to become of her"

However, she was able to describe her current situation in a very different light.

> "I can accept things now, but I mean when S was a baby, I'd never had dealing with anybody that was disabled...I think in some ways you get to know other people and then you realise that what you thought at first was such a major tragedy, your life isn't as bad as other people's lives. I know you shouldn't be helped along by that, but there is always somebody worse...It puts your whole life into perspective... It makes you value things less, a lot of things I think...Before I had S we wouldn't have been on holiday but the house would have been done. Whereas now you know, I've got no doors in the kitchen but what the heck, I'll get them one day" (8: girl, 4, dystrophic)

Another mother, who had a child with recessive dystrophic EB, described the initial trauma very vividly:

> "And I remember looking at this little thing, feeling absolutely cut up inside...You know, I remember T (husband) coming in and telling him to pull the curtains and then I just bawled my eyes out...

However, the way she described her current situation indicated that she was resolved now.

> "I get a lot of pleasure from, you know, hurdles he gets through...I suppose that's what it is, yes, you don't take everything for granted, and what he gives back to us is amazing...I feel so proud of him when I pick him up from the play group." (5: boy, 3, dystrophic)

She was still able to acknowledge current difficulties and her feelings of frustration at his physical care, but did not feel overwhelmed by these and was able to reflect on some positives in the experience.

At the unresolved end of the spectrum some mothers spoke and behaved as if the emotional impact of EB was still very raw and emotional turmoil was close to the surface during the interview. They might be tearful at times and show clear signs of re-experiencing some of the original emotions during the interview. They may have an unrealistic view of their child or of the future. This could include having unrealistically high expectations of what the child could do, but also could include minimising or denying the difficulties the child experienced. (Pianta and Marvin (1992) also include within this group parents who are still searching for an explanation for their child's condition (because they have never really accepted the diagnosis) and parents who (unjustifiably) blame the medical system for their child's condition. However, there were no examples of these two categories in these interviews.)

This mother with a child with recessive dystrophic EB described some superstitious thoughts that reflect her underlying sense of doom about the future,

> "And now if anybody says to me, 'She's doing well with food now, she's putting on weight', I say, ' Don't say that.' Because to me, I might be over-superstitious, it's tempting fate. As soon as you get complacent and start thinking that things have improved, then it goes again. And I won't do it now. I tend to look a little bit on the black side all the time... Because EB causes such a lot of problems. You know, with the eyes, with the throat, with the mouth, with the skin. You know it's just all she's ever known is pain". (3: girl, 6, dystrophic)

Another mother, with two children with EB simplex, described being able to mask her emotion in the day but finding the evenings very hard to cope with,

> " Later on when they go to bed, that's when I cry, and sort of get really upset about it. I just worry about everything. I just worry about their whole future. How it's going to effect their whole...I'm that kind of person, mind, you know. And, I, you know, about their jobs and everything." (12: girls, 6, 3, simplex)

This mother of a child with recessive dystrophic EB also described on ongoing struggle with coping with her child's EB:

> "It's been getting to me lately. I think, that's what you do, you think....you've got through one thing and then another problem comes up. And then they can be doing well, and then he'll fall flat on his hands, and thats another set back. It's always something...I've had feelings for a long, long time of feeling very sorry for myself, very sorry for myself. And maybe because I've never allowed myself to be honest before. I think about it, we really didn't ask for this. Why shouldn't I feel sorry for myself? But then another part of me gets so cross with myself." (1: boy, 4, dystrophic)

Changes in the Illness

This dimension is clearly a fluid one, in that different events or developmental stages could re-evoke some of the initial trauma, and the mother's emotional reaction would then shift as well. One factor that might cause such a shift was related to difficulties encountered directly as a result of the illness itself. For example, one mother whose daughter had dystrophic EB and was currently experiencing extreme difficulties with swallowing food, was understandably preoccupied by this difficulty which had re awakened some of her original difficulties in coping with her daughter's skin. Whilst she had become "resolved" with respect to her previous physical difficulties, these new difficulties meant she was now more "unresolved".

Resolution appeared to be closely linked to the physical state of the child's EB. Several mothers described phases in the illness that had been particularly difficult for them, and at these times they were more unresolved. It is important to note that this can be a helpful process to a certain extent. The emotional tension the mother described at these times often did help motivate her to seek further treatment or resolve the difficulties her child was encountering. The mother's ability to pick up on her child's distress can be seen as a essential way of ensuring the child gets the physical care he or she needs. However, obviously the emotional distress that this causes can also be very destructive and interfere in both the physical and psychological well being of the child.

Changes in Child's Development

The significance of the difficulties experienced as a result of EB can also vary depending on the developmental stage of the child. A mother of a child with dystrophic EB found it harder to accept the EB once her daughter was older because of the conflict between controlling her daughter's activities and allowing her more independence:

> "Well it's all changed now because I'm finding it harder now than I did when she was younger, because when she was younger you were looking after her and ...you could say where she was, so you could keep her at home if you wanted to...And nowadays she's obviously got to go to school everyday, and you want her to become more independent. And it's just about how you go about doing it. In that respect I find it very hard." (4: girl, 13, dystrophic)

Downward Comparison

There were several examples of "downward comparison" used by the mothers in this study to explain their attitude towards EB (Taylor & Lobel, 1989; Taylor, Wood & Lichtman, 1983). This process did seem to serve a useful purpose, in that it helped to place the mother's own difficulties within a different context, but does not necessarily indicate that the mother has "resolved" the diagnosis.

> "The thing I think about all the time is the children that I have seen with junctional EB, dystrophic EB that I saw at the Christmas party. I think of them kids and I think that we have been lucky...It may sound terrible but it does help me." (18: girl, 7, simplex)

> "The other thing that keeps me going is that I know she's not the only one going through it. I wouldn't wish it on anybody else's child, because I know that they're

> all as upset as I am and I feel sorry for the kids. But it's nice to think that you're not the only one. And they do understand how you feel. And you know their kids go through it and come out the other side, sort of thing." (3: girl, 6, dystrophic)

Theme 3. Coping with Caring

This theme reflects the degree of difficulty the mother experienced in providing the care for her child. One end of the dimension were those who were easily able to provide the physical care and who experienced little distress at the nature of the care that was required. At the other end of the dimension were mothers who found it extremely distressing to manage the treatment regimes and who sometimes felt overwhelmed by the child's needs. The amount of physical care a child needed from his or her mother varied from virtually none, to up to three hours a day. For example, some of the older children were totally responsible for their own blister care, although they occasionally needed some help. For other younger children with more severe forms of EB some mothers estimated that they spent a total of 3 hours a day on the child's care. However the burden of the care on the mother was not a simple reflection of the amount of care that was required. The perceived burden was affected by many different factors, for example, how distressing the mother found the treatment, how co-operative the child was with the treatment and the level of support the mother received.

The most dominant of these was the level of distress experienced by some of the mothers at the physical care she had to provide for her child. The management of EB is particularly difficult for mothers because of the pain and distress the child experiences as a result of the physical care for example, removing dressings, bathing and bursting blisters.

All mothers had to cope with a time of transition where she took up the physical care of the child. A few easily tolerated the demands of the physical regime. However, many talked openly about feelings of fear, distress and also disgust at the physical care involved. Many felt that they simply could not believe that they would be able to manage the physical care, because of feelings of "squeamishness" and feeling overwhelmed by the physical demands. However, the experiences of many mothers illustrate how the practical reality of having to get the treatment done forced them to overcome their own difficulties to a certain extent. Whilst all of these mothers did make this transition, in spite of their initial difficulties, this was not necessarily true of their partners or other family members. Mothers gave examples of partners whose level of distress was such that they refused to participate in treatment, even on a respite basis. The mothers had had to overcome this difficulty, despite the emotional impact on themselves and many described this very vividly.

At one end of the dimension were mothers who had taken up the physical care easily and had not found this distressing. Some of the mothers with children with dystrophic EB did fall at this end of the dimension despite the fact that objectively the child's skin would be rated as severely affected.

> "Having a medical background I could grasp and understand it better...I think I could cope with it better. And I, you know, dressings and things don't bother me. I have no problem, you know, doing something practically. And I remember (the DEBRA specialist nurse) saying, 'How will you cope if you've got to do something that you think might hurt him?' You know, that type of thing I can cope with. But, you know, G (husband) is not so good.". (10: boy, 2, dystrophic)

> "I've always thought I'm glad it was the first baby and not the second…So I am glad that it was the first one. Even though it was a shock. Because, obviously you expect a healthy baby…I wasn't too bad actually. I seem to take things as they come, and don't worry about things." (2: girl, 9, dystrophic)

However, the majority of mothers expressed difficulty with the management of their child's skin, especially initially.

> "And other members of the family come to me "Oh I don't know how you cope, I couldn't do it, bursting all those blisters, and having to do those dressings. I don't know how you cope" And I thought, well I cope because I've got to. You either love the baby, and you do it. You don't like doing it. You don't want to do it. But you do it…What choice have you got? You haven't really got a choice have you? You've got to do it. And all those people that said it to me, they would do it too if it was their child. They say they wouldn't, but they would." (3: girl, 6, dystrophic)

> "I feel like a nurse…I burst the blisters, I mean I can, I hurt him, it's awful, but I've learned to just about switch off, I don't think you ever do inside, it's awful, but you just have to do it." (1: boy, 4, dystrophic)

> "I hate it, but what can I do. I suppose if anyone saw me they'd think 'Oh she's a cold cow'. Because with the blisters I shut myself off completely." (12: girls, 3 and 6, simplex)

> "I'm doing something awful to them which they know that I have to do. Even so it is horrible. I mean sometimes I'll end up in the bedroom in tears…it really is having to be cruel to be kind, but it's constant…I mean sometimes I get incredibly depressed." (19: boy and girl, 9 and 7, simplex)

> "There are days when I think well if I could afford six pounds I'd phone up the Medicare people to come out here and do it. But I don't I just say, you know,

> we're going to have your bath. We're going to get it done. And if I sit there in tears, I'll do it." (9: boy, 9, dystrophic)

> "You ask yourself why, and how can he have this when I haven't had it and no one else has had it. And you know, I used to get so upset when I used to have to prick his blisters. I used to actually feel sick that I had to do it." (21: boy, 10, simplex)

Support

Some mothers felt totally unsupported with the practical care and were unable to relinquish the physical care of their child to anyone, because there was no one else who could take on this difficult task for them. Several mothers acknowledged that it is hard to relinquish care when you know the child would be more comfortable with their own mother, which increases the pressure on the mother to provide the care herself. However, other mothers were fortunate to be able to afford a nanny, au pair or other childcare that enabled the mother to have more choice about whether or not she did all the care. In a small minority of the families the father carried out the physical care. There was considerable variation between families as to the level of support they wanted or chose to use.

> "They said to us they'd supply us with one of the, what do you call it, one of these home help people, you know, to do his bandages. And we sort of said that no we'd do them ourselves." (7: boy, 8, dystrophic)

> "They didn't come in to do the dressings. I got it into my head that I wanted to do it, and that's very much, you get that feeling that you're the only one that can do it." (1: boy, 4, dystrophic)

> "We then got a nanny and trained up a nanny to help. Just to take the physical...I say the physical, but actually in a way it's sort of the emotional pressure. The burden of it. So we've been so much luckier than others." (6: girl, 4, dystrophic)

Some mothers were aware that their own expertise had developed over time and that this enabled them to become more accustomed to the physical difficulties encountered in EB in a way that their partner, who had not been so involved with the physical care, had never experienced. Some mothers even expressed a sense of satisfaction or pride that they had overcome what had initially felt overwhelming.

> "But my husband finds it hard. Because he's not dealing with it day to day, he finds it more difficult to come to terms with. You know, when he falls over I just say, 'Oh come on, get up.' You know whereas D gets really upset. And I think that really, it's just because he's out of the house all day and he just sees, well he's not really dealing with it day to day so much." (5: boy, 3, dystrophic)

> "It would probably be interesting to see what G would say because I guess...you'd get a different picture, because they watch, but they...and I always think sometimes, you know, if you can do things practically then you don't feel so hopeless." (10: boy, 2, dystrophic)

Child's Temperament

The child's temperament clearly had an effect on how easy or difficult it was to carry out the treatment. It had a direct impact on the experience of providing the care because all the mothers found their own child's distress very hard to tolerate and some children did experience and act out a high level of distress. This was most marked in a family with two children with EB simplex, one of whom would sit meekly while the treatment was carried out and the second of whom would consistently resist the treatment. This need to carry out the treatment despite the protests of the child will clearly have an impact on the mother and child relationship and many mothers commented on this. Whilst this may be of particular significance for EB because of the particularly unpleasant form of management of the condition, this is also a difficulty for many other forms of chronic illness.

> "Then he has his bath which can take anything up to two hours. Depending on the length, the amount of blisters he's got. And how reliable and co-operative he can be. It can be one of those days where he doesn't want a bath. And you literally end up fighting with him, and he ends in tears. I end in tears. Massive arguments." (9: boy, 9, dystophic)

Support of Health Care System

Another factor which is relevant is the support provided by the health care system and the parent's mastery of this system. The standard of care received by these families varied, but in addition, some families were more able to make assertive use of the health care system and obtain the help they needed. Many mothers had considerable difficulty obtaining the dressings, needles and bandages they needed and had to be very persistent and determined in order to obtain them. Some mothers appeared to rise to this challenge and became experts in using the system, but others, understandably, found it very daunting to challenge the medical system and this added to their difficulties.

> "You've just got to grow a very thick skin, you yourself have to grow a very thick skin, and just smile. And say, 'Oh no, I really do think I want it done that way.' They try and persuade you that you're an over-protective mother" (11: boy, 14, simplex)

> "You have to be quite firm. And in actual fact, it's always been difficult. They've always given me the dressings I want. This time I got different dressings and different bandages. And my comment was, 'Are these cheaper?' which the health visitor thought was quite funny because she said, 'Probably J, yes.' And we actually found that they are cheaper. And now I think they are going to write to Great Ormond Street to find out if actually that is necessary." (10: boy, 2, dystrophic)

At the other extreme were a family who followed inappropriate advice and felt unable to challenge the doctor's decisions even when it was clear she did not know what the child's condition was.

> "She phoned social services up and she told that that unless the house was carpeted all the way through that T couldn't come out because so much as a splinter could kill him. So we followed by her rules. She hadn't got a clue what it was, but we still followed her rules." (20: boy, 7, simplex)

Theme 4. Treating the child as "normal" or "special"

This dimension reflected the dilemma parents faced when trying to give their child as "normal" a childhood as possible whilst being sensitive to their needs as child with a chronic illness. At one end of the dimension would be a parent who makes no concessions as a result of the child's illness, and tries to treat the child in the same way as a healthy child. The other end of the spectrum would be a parent who is excessively protective and unnecessarily restricts the child as a result. Mothers of children with EB were very conscious of this dilemma, maybe because of the need to address how protective to be from a very early age, in terms of the child's physical care. Children with EB are both physically vulnerable (literally) but also psychologically vulnerable because of their appearance and the stigma associated with skin problems. It is very easy to understand the instinctive desire to protect a child who is vulnerable, and this desire can be very functional, in that it does enable the mother to provide the necessary practical care which to an outsider, often seems an impossible or unmanageable task.

The appropriate balancing point between treating the child as special or as normal, varied depending on the nature of the child's EB and the severity of his or her symptoms. Children with EB simplex often look normal and yet have particular difficulties with mobility, which may make it harder for them to lead a "normal" life than a child with dystrophic EB, who may appear very abnormal or disfigured. Both mobility and visibility of the condition appeared to strongly influence this dimension for children with EB.

Other people's perceptions

Most parents feel very protective towards their own children, and this is increased when the child is vulnerable. The difficulties experienced by the mothers of children with EB were based on both the physical restrictions on the child and the restrictions due to other people's perceptions of their child. This was due both to a lack of understanding of the child's physical difficulties and attitudes towards children who look very different.

One feature that several mothers talked about, but particularly the mothers of children with EB simplex, was the lack of understanding of the difficulties with mobility, which frequently caused problems for the children in school. These children were often seen as avoiding school or using the illness to their advantage.

> "I think our main problem has been trying to convince teaching staff that there's a problem. It's all very well that you go and talk to the schools before you go...But when it actually comes down to it, they (the children) get very little help

> whatsoever. Because there's nothing to be seen, it's not noticeable." (13: girl, 15, simplex)

> "I think the lack of understanding is like they said, to me it has always been the biggest problem. Other people's perception of it. And trying to get it across to people, and it's like a constant battle to try and get teachers to understand how difficult it is." (17: girls, 10 and 16, simplex)

However, some teachers were able to be helpful and find constructive ways of solving a child's difficulty with mobility.

> "And the teacher said that it was amazing how, rather than treat her like a freak, ...they would walk around watching the floor all the time. And if they found (something) they'd pick it up and say, 'I picked this up so that Y wouldn't tread on it.' It was amazing how caring they got. But not to the point where they made her feel like something different." (18: girl, 7, simplex)

The children with dystrophic EB who were visibly very different were frequently stared at in public and several mothers talked about the distress that teasing and rejection at school had caused their child and themselves.

> "And I'm sure it's something that any mother whose got a child with something wrong with it feels. And you're standing in the supermarket queue while people are looking at him. And his scars are such that, they're little white scars and they could almost look like cigarette burns. And you're standing in the queue and there's someone looking at you or looking at him and then they don't say anything." (5: boy, 3, dystrophic)

> "How you cope with other people. Sort of, people who don't know A, sort of looking at him, and feeling very protective towards him. But I feel sure a lot of the new mums feel like that. But I've learned to cope with it. You either give them a leaflet or you sort of, if they don't look, frankly they just stare, people are

> like that, you just turn a blind eye to it and think 'If only you knew, A is ten times whatever to you.'" (1: boy, 4, dystrophic)

> "He took Christmas cards into school for his friends at school and he came out and I said 'Did you give the kids your Christmas cards?' And he said, 'No, I threw them in the bin.' And I looked and I said 'Why?' And he said 'They said they don't want them, they might catch something."(20: boy, 7, simplex)

> "By that time, he was really bad. He had both his hands tied up. Both feet tied up. His face was bad. And you'd start to get the weird talk down in the town that I was abusing him. I had the police round to the door because there had been a report of me beating him up. I stood in the town and got accused of scalding him." (9: boy, 9, dystrophic)

All the mothers were involved at times in explaining to other people about their child's condition, and this could also prove quite difficult for the mother herself.

> "There was one time where she really fell at school and they couldn't find the information because they'd lost it. And I had to go and get her. I was furious that they'd actually made a big thing out of it. And for the first time, E realising that she was different in a big way. That really upset me. We didn't want that" (14: girl, 15, dystrophic)

> "Because before she goes anywhere or does anything...you've got to explain it. And it's very difficult, because they just look at you with an open mouth and some of them think you've been over-reacting. And others, once they understand a bit more, think, 'Oh my God, how does she cope?'" (6: girl, 4, dystrophic)

> "There's nothing worse than somebody trying to be sympathetic but they don't really understand. They say, 'Oh but won't it get better?', 'Have you tried different washing powder?', 'I knew someone who had this, that and the other'.

> It's not the same thing. And they're not listening you know. There's no ease on the frustration that you have discussing with people."(15: girl, 3, simplex)

Some mothers explicitly talked about the possibility of her child using their difficulties to avoid tasks they didn't want to do. This is a normal feature of childhood, but can make it quite difficult in terms of judging how lenient to be with a child with a chronic illness.

> "A few times if I ask him to do something and he doesn't want to do it he'll make a big thing about it. Like go up the stairs...it's like it's too tiring for him, which is understandable. But there again I can't let him away with it all the time. Not when he can go out and play and then he comes in and won't do something." (21: boy, 10, simplex)

> "Because sometimes come lunchtime, their feet are just blistered completely and they can't cope with the afternoon. But if they're going every morning at least it's continuity, because otherwise children get it into their minds that they can just stay off whenever they feel like it. I mean they're no angels. They'll play on it" (19: boy and girl, 9 and 7, simplex)

Impact on Mother of Treating Child "Normally"

Mothers also identified the importance of being able to separate her own feelings from the child's feelings, and to try and conceal her own level of distress, if necessary. In fact, mothers sometimes took strength from her own child's resilience, which actually helped the mother to feel more resilient too.

> "But there are times when he will fall and it's such a bad fall, and the trousers off, and the little knee pads he wears. He's just absolutely raw. And I have to turn away because I feel myself crying and I mustn't let him see that...Then he gets up and runs around and that's all you need, really to stop. He just gets up. He

> hasn't seen me starting to cry and he starts talking about tractors or something and you just, you just get over it and try and forget it" (5: boy, 3, dystrophic)

> "You have a choice. You either...go greyer and greyer, and let him be happy. Or you wrap them up in cotton wool and make them miserable, and you're happy. You can't have both. And you've got to choose which is best." (11: boy, 14, simplex)

> "I've never mollycoddled her, I mean friends of mine say, you know, you've had a good attitude. But a lot of that is her attitude. You know, that's determined my attitude because there'd be no holding S back if she wanted to." (8: girl, 4, dystrophic)

Despite the mothers attempts to give the child as much freedom as possible, it was apparent how anxious this made them. Unfortunately, the child's experience is limited because of their EB, even when the mother is trying hard to contain her own difficult feelings and allow the child as much freedom as possible.

> "I mean, we let him be as normal as possible. I tend to watch him more. I don't let him go out of my sight. I mean, I won't let him go off and go for a walk on his own or go off with his sister. He has to be in my sight so that I know where he is. I mean he's very restricted in what he can do. Because I'm very wary of letting him out in case anything happens. I mean if he was to fall off his bike and I'm not, I didn't know about it, I'd never forgive myself." (9; boy, 9, dystrophic)

> "He fell off a motorbike when he broke his arm...and he didn't have one blister at all. We couldn't believe it. We run over there...and it was like we were on like a Sunday out and he was on this little thing and expected him to be like blistered and torn everywhere. And we picked him up and he said, 'My arm'. And we said, 'But he ain't got a blister!' The poor child was sitting there with his arm broken!

We was just sort of standing him to see if there was a blister." (7: boy, 8 dystrophic)

- Two mothers who had girls with EB identified gender differences as important.

They felt that because of the physical restrictions of EB boys would find it harder to cope.

"If R (girl) hadn't had it and N (boy) had I think that we might have been different, had a lot more rules. N will fight and kick and have a great time" (14: girl, 15, dystrophic)

"I definitely wouldn't want a boy with it...I think it would be much worse. I think because boys are, because their nature is, you know, sort of football and outside and sort of running around. I think it would be worse with boys." (17: girls, 10 and 16, simplex)

However, one mother of a boy with EB identified the reverse gender difference because she saw the impact on the child's appearance as very important.

"I mean little girls are different. It's bad enough for a boy. But, you know. Especially a little girl like S. You know she's very pretty and I just can imagine how it would affect her more so." (21: boy, 10 simplex)

Summary

There are four key themes that this study has identified which are important in understanding a mother's adjustment to her child's EB. Firstly, there is the difficulty experienced between when the child is born and obtaining correct information about the child's condition. Secondly, there is the resolution of the diagnosis, which may vary over time depending on the child's current level of physical difficulty and their developmental stage. This reflects the degree to which the mother has accepted the condition and overcome any initial trauma to develop a positive but realistic view of the difficulties the illness poses. Thirdly, there is the burden experienced by the mother at the physical care she has to provide. There is considerable variation in the amount of care needed by a child with EB, but this dimension reflects the psychological difficulty in managing this care, and is not therefore directly related to the amount of physical care. It does seem to be affected by the amount of support the mother feels she receives and the child's temperament and response to the treatment. Finally, there is the balancing act between treating the child as "normal" and making allowances for their illness. Children with EB are physically vulnerable, so all mothers have to find a balance between protecting their child and allowing them to develop as normally as possible.

CHAPTER EIGHT

A QUALITATIVE STUDY OF THE EXPERIENCES OF MOTHERS OF CHILDREN WITH EB

DISCUSSION

Evaluation of the Themes

The themes identified in this study have also been shown to be relevant in other studies of children with a chronic illness. Each theme will be considered in turn and the findings of this study will be discussed in relation to existing literature on the impact of chronic illness on children and families.

Difficulty with Diagnosis

Obtaining the diagnosis has often been described as a key moment for any parent of a child with a chronic illness (Davis, 1993). However, there are several features of EB that appear to make this stage particularly crucial. Firstly, EB is very rare, and very few doctors have direct experience of diagnosing or managing children with EB. As a consequence of this, getting access to the appropriate information and services can be particularly hard for families of children with EB. As in many other studies, the mothers who had a struggle to obtain this information or who were given incorrect information, remembered this stage, often in great detail, for long periods of time (Quine & Rutter, 1994). The emotional impact of this is clear from their descriptions of these events. Many saw their first contact with DEBRA as a turning point, and DEBRA's services were universally praised. Obtaining the very specialised services needed for rare conditions can be difficult, and self-help groups and the voluntary sector can play an important role in this (Partridge & Nash, 1997). Many mothers clearly valued the opportunity to have contact with other mothers of children with EB. The rarity of EB means that mothers of children with EB are usually in the position of having to explain their child's condition to other people, and rarely feel that others understand their dilemmas. Support groups, such as DEBRA, provide a very important link between such families, and this informal support is perceived as very helpful by parents.

Secondly, the initial symptoms of EB can be very frightening for parents (and professionals) particularly for the most severe forms of EB. Most of these severely affected children have a recessive form of EB so it is extremely unlikely that they will have had any prenatal knowledge of EB. Bradbury and Hewison (1994) showed how much variability there is in a mother's adjustment over time to having a baby with an obvious facial disfigurement, but all the mothers experienced some initial adjustment difficulties. This is likely to have important influences on the mother's ability to be responsive and sensitive to her child, and hence on the child's attachment status. This highlights the need to be able to provide sensitive support to mothers at this stage in order to minimise the impact of these initial difficulties on the mother-child relationship (Walters, 1997).

Finally, EB is a genetic condition and there were seven families in this study who already had some experience of EB within their family. However, because EB is very rare, only a few of these families had a good understanding of both the inheritance of EB and the impact of EB on a child, rather than an adult. The attitude of the parent with EB towards their own EB will have an effect on their reaction to the child's EB. The increased understanding of the genetic causes of EB together with improved pre-natal testing procedures may mean that the understanding of families does increase over time. However, the difference between existing family beliefs about the illness and newer genetic theories can in itself sometimes cause some difficulties. Rolland (1999) describes this conflict in this way: "Family myths and narratives about a vague disorder that may have evolved over generations collide with high technology medicine." (p 127). This conflict was already apparent in some of the families, but may become more prominent as the genetic causes of EB become better understood.

Resolution of the Diagnosis

The theme of resolution identified in this study is very similar to that discussed by Pianta and Marvin (1992) and Pianta, Marvin, Britner and Borowitz (1996), who emphasise the importance of looking at the shift between the mother's description of her emotional reaction when the child was diagnosed, compared to her current experience. This shift reveals the extent to which the mother has "resolved" the difficult emotions associated with the realisation that the child has a serious problem and found a positive way of managing the illness. Some difficulties with the concept of resolution were apparent in this study. This concept is clearly a dimensional one, although Pianta and Marvin (1992) do describe two discrete categories of resolved and unresolved. Hence the dividing point between these two categories is to some extent arbitrary and relies on a very specific coding scheme.

Another problem arises because of the difficulty in separating the mother's mood from her state of resolution. The rating of resolution depends entirely on the mother's description of her feelings and thoughts about her child's illness and if the mother is depressed, her description will be, by definition, more negative and this influences her rating of resolution (Beck & Emery, 1985). It was not possible to assess the relationship between resolution and mood in this study, but it would be important to clarify how separate these two concepts are in future research.

The concept of resolution is a very helpful one in terms of understanding the variation in levels of distress expressed by the mothers of children with EB. There is no doubt that EB is a painful and difficult to manage disorder, but it was noticeable that the level of distress expressed by the mothers was not directly related to the severity of the child's EB. This is supported by the findings of Study One. Resolution is a broad concept that reflects the mother's appraisal of the difficulties presented by EB and may be a useful measure for identifying mothers who feel unable to cope with the child's illness. Thompson et al. (1994) have shown that the mother's appraisal of a situation is a strong predictor of levels of psychological distress. The concept of resolution has been shown to be a useful one in this qualitative study, but further studies are needed to determine how valid and reliable this concept is.

Coping with Caring

The mothers' accounts of the physical care they provided for their children reveal the emotional cost of having to manage the child's illness. Whilst the treatment for EB is particularly unpleasant and causes very clear distress for many mothers, other chronic illnesses in childhood (e.g. diabetes, cystic fibrosis) also involve physical treatments that are unpleasant for the child and hence difficult for the mother to provide.

Some mothers in this study did describe a sense of mastery and achievement at providing good physical care for their child, but this did not in any way compensate for the distress they also experienced. Folkman and Moskowitz (2000) describe a similar pattern amongst carers of people with AIDS and reflect on the role that positive reappraisal of difficult tasks plays in enabling caregivers to maintain their support over long periods of time. "The sense of mastery and control engendered by successful problem focused efforts helps explain caregivers' reports of positive affect in the midst of their distress" (p. 650). They argue that one of the limitations of much research on coping has been the assumption that coping is solely directed at reducing distress. However, there can be multiple and competing goals in any situation, and coping strategies are not always directed by the simple motivation of reducing distress.

Several studies have shown that the mother's appraisal of the burden of caring for chronically ill child is related to the level of maternal distress reported (Canning, Harris & Kellehar, 1996). Maternal distress is more strongly related to the mother's subjective rating of the burden she experiences than to an external rating made by a doctor or a nurse (Silver et al., 1998). However, these results may be partly accounted for by the measures of appraisal of burden and measures of maternal distress, which are often confounded and the relationship becomes rather circular. For example, a mother who is distressed is by definition more likely to report experiencing feeling burdened than a mother who is less distressed. Whilst this study did not specifically measure burden of care, the accounts given by mothers of the difficulties they experienced are likely to be influenced by factors such as the mother's mood, as well as the physical demands of the care she has to provide.

Treating the Child as "Normal" or "Special"

Many of the mothers interviewed in this study were themselves acutely aware of the need to balance their protective feelings towards the child with the needs of the child to have as normal a life as possible. The nature of EB means that this question has to be addressed very early on, and medical professionals working with children with EB are aware of the importance of helping the parents find the appropriate level of protectiveness (Atherton, 1990; Foster, 1990). However, it is relevant for all types of chronic illness in childhood and has been identified as an important influence on parenting children with chronic illnesses which is particularly relevant in adolescence when the young person assumes more responsibility for his or her own care.

Several studies have shown differences between parents and professionals views on parenting styles, with professional predicting higher levels of protectiveness than are reported by parents themselves (Noll et al., 1998). The underlying reason for this difference is not clear. It is possible that parents are more protective than they report themselves to be and that they under report protectiveness, either to present themselves in a favourable light or because of a level of denial about their own anxiety. However, it has also been suggested that they interpret standardised questions in a way that already takes into account the child's level of difficulty, and hence use these scales in a different way than professionals. Alternatively, it may be that professionals, who have not had to adjust to the demands of having a child with a chronic illness are reporting the level of protectiveness they would expect to feel in this situation.

There have been some suggestions in the literature that children who are clearly and markedly "different" from a healthy child have fewer adjustment problems than children who are less obviously different (Pless and Nolan, 1991). This may be related to the expectations of the children and how they are treated by their peers and other adults, as well as their families. Many of the children with EB simplex felt their difficulties were not taken seriously and that other people did not understand their difficulties, partly because their disability was not obvious. On the other hand, some of the children with dystrophic EB were clearly very different from their peers and had all the difficulties associated with the stigma of disfigurement and consequent social difficulties (Walters, 1997).

Links Between the Themes

There are links between the different themes described here. Resolution can be seen as a higher level or meta theme because if a mother is resolved about the child's illness, it is likely that she has fewer difficulties with the practical care she has to provide. And vice versa, any mother who is still struggling with providing the physical care cannot be said to be resolved about her child's illness. However, resolution appears to be independent of the difficulty experienced in the diagnosis phase. Some of the mothers who had very little emotional reaction to the diagnosis did remain unresolved about the disorder, whereas some of the mothers who experienced the most difficulty at the time of diagnosis did become resolved about the condition over time. It is likely that the mother's level of resolution is also linked to her management of the child's needs and her ability to treat her child "as normally as possible". A mother who is resolved is more likely to be able to tolerate the anxiety generated by allowing her child to take some risks, whereas an unresolved mother might find this intolerable, and hence protect her child more.

Putting it back Together: Different Perspectives on EB

The quantitative and the qualitative studies of EB have provided different perspectives on the impact of EB on children and their families. The quantitative study has enabled comparisons to be made between this sample of families with EB and other samples of children without physical illnesses. In addition, the study showed an association between fathers' ratings of the severity of the child's EB and psychological distress. The family variables of cohesion and conflict that have been shown to be associated in other studies have also been shown to be relevant in families of children with EB. However, the qualitative study has been able to convey the experience of mothers who have children with EB and to give some insight into the psychological processes that may be of importance in adjusting to having a child with EB.

At first sight the results of the two types of study may appear to give rather different views of the impact of EB on children and families. The levels of maternal distress measured in the quantitative study were not high when compared to the norms for the measure, and yet the descriptions given by mothers of children with EB illustrate the marked impact this condition has on the families. These two studies were however measuring different concepts and there are some difficulties with the particular measure of psychological distress used in the quantitative study. The level of psychological distress within this group may not be more than average as measured by the GHQ, but this is a very general measure of stress and not a specific measure of the impact of having a child with a chronic condition like EB. Many authors (e.g. Bradford, 1997; Eiser, 1990) have argued that more specific measures of the impact of illness need to be developed in order to more accurately assess which predictors are associated with adjustment to chronic illness.

The qualitative study may shed some light on the finding that the fathers' levels of distress were related to severity of the illness, whereas the mothers were not. As reported in the qualitative study, many mothers reported a form of adjustment over time to the physical demands of EB because of the necessity of providing the practical care. However, many fathers had never had to overcome these difficulties because they weren't so involved in the practical care of the child so they continued to find the child's symptoms and care more difficult. This is consistent with Sloper and Turner's (1993) interpretation of their study that found that mothers and fathers found different aspects of caring for a chronically ill child stressful.

The two studies also illustrate the advantages of using these two different methodologies to study the impact of chronic illness. If the self report questionnaires were the only source of data, these would give only a very limited picture of the experience of these families and provide very little information about the psychological impact of the condition in terms of understanding the experiences of these families. However, if only qualitative methodology had been used, it might have been assumed that far high levels of distress were experienced than were actually reported. In addition, it would not have been possible to evaluate the links between severity of illness or family functioning and psychological functioning. The insights from both types of study complement each other and support the use of methodological pluralism (Barker, Pistrang & Elliott, 1994).

Limitations of the Study

The qualitative approach is also subject to several limitations. The transcripts used in this study are the mother's report of her experience, and not necessarily factual summaries of events. The aim of the interviewing process was to allow mothers to describe their experience in their own words and the interviewer aimed to encourage the mother to reflect on these experiences (Silverman, 2000). However, these were brief research interviews, and the results have to be interpreted within that context. There are undoubtedly issues that mothers felt were too sensitive to discuss in a brief interview even though the interview was carried out at home and the results were to be reported anonymously. There is a lot of variability in how comfortable individuals feel in the context of an interview like this one; whilst some mothers appeared to enjoy the opportunity to talk about their experience, others appeared much less comfortable. In addition, mothers knew that the research was funded by DEBRA, and will have been influenced by their views about DEBRA. In addition, the interviews can only provide a picture of adjustment to EB at one particular time and this is an inevitable simplification of a much longer term process of adjustment.

There are limitations to the generalisability of this study. This sample of families may not accurately represent the population of families with EB. They are likely to be those who are more concerned about their condition, and hence have joined DEBRA, and are willing to participate in research. In addition, three families dropped out of the study after having consented to take part. These three families were all experiencing considerable difficulties, which was the reason they withdrew from the research. It is therefore possible that the study did not include some of the most severely affected children and families.

Qualitative research cannot be assessed using the familiar tests of reliability and validity that are used in quantitative studies (Fiese & Bickham, 1998). Therefore other methods were used in this study in order to address reliability and validity (Elliott, Fischer & Rennie, 1999). First, the analysis started with a smaller number of cases in order to develop preliminary themes and then the analysis was extended to a larger group in order to determine whether the themes were accurate descriptions of the data. This method, known as constant comparison, ensures that the reported findings are not confined to a few interesting or striking examples that do not reflect the data as a whole (Silverman, 2000). All the transcripts were examined to find examples that either confirmed or refuted the themes.

In order to assess the validity of the themes, some authors recommend that the results of the study are fed back to the participants in order to confirm the relevance of the identified themes (Stiles, 1993). This is not always a straightforward task because the analysis does include some interpretation that may be valid, but which may not be "approved" by the participants. In this study, a preliminary report was sent to all participants in the research to comment on before finalising the report for the funding organisation.

Some authors have suggested that it should be possible for an independent audit of the data and the process of selecting the themes to be carried out, in order to ensure that the selection of themes and examples do accurately reflect the data (Lincoln & Guba, 1985). This can also be done in a modified form by giving enough detail about the methodology used and sufficient examples of text to enable another person to assess the relevance of the themes. Smith (1996) recommends that it is a useful exercise to make it possible for another person to "follow the paper trail" from the original data to the write up, even though this will probably never actually be done.

The process of triangulation refers to the use of other sources of data to verify the findings of the research (e.g. some quantitative data, or interviews with other connected people). The different methods would not be expected to produce the same findings, but would be expected to be consistent with one other, or to help explain each other (Lyons, 1999). In this study this has been done by reflecting on the combined findings of the quantitative study and the qualitative study to compare the picture of the impact of EB on children and families given by the two different methods.

Finally, qualitative researchers accept that the researcher's perspective will influence the interpretation of the data. In order to make this explicit and to allow the reader to take this into account when interpreting the results of the study, many authors stress the importance of the author declaring their own experience or interest in the phenomena being explored which is known as reflexivity (King, 1996). This statement is included in Chapter Six, as part of the description of the methodology for this study.

It has often been pointed out that many of the issues that are relevant to the evaluation of qualitative research are equally relevant for quantitative research (Woolgar, 1996). However, the procedures by which such issues are addressed have become more established within the quantitative paradigm. In addition, the different epistemological stances taken by the two paradigms do result in different questions that need to be answered. For example, many qualitative researchers do not accept that there is an external reality that can be measured meaningfully in any "pure" way but that all psychological events are socially constructed. In which case, it would be impossible for any measure to accurately reflect "reality" and hence comparing results with the objective truth is not possible. This does not mean however, that any interpretation is legitimate and hence the task for the qualitative researcher is to demonstrate the relevance of and evidence for, their particular interpretation of the data.

To end this chapter, the following quote from one of the mothers interviewed for the study, summarises the impact of EB on children and their families. When asked if there was anything else she would like to add, this mother, who had a young boy with dystrophic EB, said, only half jokingly, "I think we ought to get medals." (10).

CHAPTER NINE

THE PSYCHOLOGICAL IMPACT OF ECZEMA

INTRODUCTION

This study focused on the common condition of childhood eczema. EB and eczema are very different skin conditions and this study is not intended as a direct comparison with the study on EB. However, this study will develop further two of the findings from the studies on EB, namely, the importance of the concept of "resolution" and the nature of the relationship between severity of the illness and psychological adjustment.

Resolution was one of the themes identified in the qualitative study of children with EB which appeared to be an important indication of the level of difficulty experienced by mothers in coping with her child's illness. This study used the standardised interview measure of resolution (The Reaction to Diagnosis Interview: Pianta and Marvin, 1992) in order to determine whether mothers who were "resolved" had lower levels of psychological distress than mothers who were "unresolved".

In most studies of the impact of chronic illness on children and families, the severity of the illness has not been directly related to the level of psychological distress experienced (Eiser, 1993). However, in the EB study, severity was associated with the fathers' level of psychological distress, but not with the mothers' level of psychological distress. It has been suggested by Thompson et al. (1994) that it may be the individual's appraisal of the difficulties or burden presented by the illness that is the important predictor of distress important rather than the level of objective severity. This study tested the hypothesis that the mother's resolution of her child's diagnosis acts as a moderating variable on the relationship between the severity of eczema and psychological adjustment. It was predicted that the mothers who were resolved show the same level of psychological difficulty whether or not the child has mild or severe symptoms of eczema. However, for the mothers who are unresolved, and who therefore continue to perceive eczema as a burden, there will be a correlation between severity and psychological adjustment.

Although eczema is a common condition in childhood, there are very few studies about the psychological impact of eczema. In the meta-analysis carried out by Lavigne and Faier-Routman (1992) of studies conducted on the impact of chronic illness on children and families, there were no studies about childhood eczema. Since then, few studies have been published.

This chapter will begin with a review of the clinical features of eczema and the treatments for it. Then the existing studies on the psychological impact of eczema on children and their families, and the impact on the mother-child relationship will be summarised. Finally, the aims of the current study and the research questions will be described.

Description of Eczema

Atopic eczema is defined according to the following diagnostic guidelines (McHenry, Williams & Bingham, 1995):

Diagnostic Guidelines for Atopic Eczema

Must Have:

An itchy skin condition

Plus 3 or more of the following:

1. History of involvement of the skin creases such as folds of the elbows, behind the knees, fronts of the ankles or around the neck (including the cheeks in young children).
2. A personal history of asthma or hayfever (or history of atopic disease in a first degree relative in children under 4).
3. A history of general dry skin in the last year.
4. Visible flexural eczema (or eczema involving the cheeks/forehead and outer Limbs in children under 4).
5. Onset in the first two years of life.

Atopic eczema is associated with the other atopic conditions of hay fever and asthma and is thought to be caused by an interaction between environmental, immunological and genetic or familial factors. The word "atopic" is used to distinguish the most common form of childhood eczema, from other types of eczema, such as contact eczema (Atherton, 1994). The words "eczema" and "dermatitis" have been used interchangeably in the past, although atopic eczema is now the most common term. For simplicity, the word eczema will be used to refer to atopic eczema in this study.

Eczema is a common condition in childhood, affecting between 5 - 15 % of children (McHenry, Williams & Bingham, 1995). The prevalence is highest amongst infants, falling to 8.1% in 2 – 11 year olds and 2.2% in 12 – 15 year olds (Herd, Tidman, Prescott & Hunter, 1996). However, most of these children have very mild eczema and are seen and treated by their GP. Only a small minority, about 4 % of all children with eczema, are referred to a dermatologist (Emerson, Williams & Allen, 1998). There is strong evidence that the prevalence of eczema has increased over the last 30 years, even allowing for increased awareness of the condition (Williams, 1992). The cause of this increase is unknown although several factors are thought to be of relevance e.g. the increase in air pollutants, reduction in child infection, and an unventilated indoor climate. The rates of eczema are not uniform across all ethnic groups and for example, London born black Caribbean children have an increased risk of eczema (Williams, Pembroke, Forsdyke, Boodoo, Hay & Burney, 1995).

Currently there is no cure for eczema, although several treatments have been shown to improve symptoms. The current guidelines produced by the British Association of Dermatology (McHenry, Williams & Bingham, 1995) recommend the use of emollients (moisturisers), topical corticosteroids, avoidance of provoking factors, antibiotics when necessary, and antihistamines. Other, more controversial, treatments include traditional Chinese herbal medicines, allergen avoidance through a restricted diet or house dust mite avoidance, photochemotherapy (PUVA) and evening primrose oil. New treatments, such as tacrolimus ointment, are currently being developed and show some effectiveness (Ruzicka et al., 1997). In addition to the established treatments, a large number of complementary therapists offer treatments for eczema for example, acupuncturists and homeopaths, and many families use "over the counter" treatments and complementary therapies in addition to prescribed treatments (Ernst, 2000).

Eczema does resolve spontaneously with age for the majority of children (Williams & Strachan, 1998). For most children affected it is a mild condition, which causes some discomfort, but which does not have a major impact on their lifestyle. However, it can cause considerable distress for both the child and carers. The main symptom, itching, can be very distressing for a child and results in scratching. Scratching can cause considerable skin damage itself, and the combination of the eczema and damage to the skin caused by scratching can be quite disfiguring. Many children with eczema have bouts of scratching at night, which are distressing and disruptive for both the child and his or her family. In addition, the typical pattern of the condition is of considerable fluctuation in severity, with periods of manageable skin inflammation followed by acute flares or exacerbation of the condition. The condition is also extremely frustrating to manage partly because there is no cure, but also because it is so unpredictable.

Many families are reluctant to use long term steroids, which are the mainstay of conventional treatment for eczema (Charman, Morris & Williams, 2000). Fischer (1996) describes the low level of compliance to prescribed treatments in families of children with eczema. He suggests this is due to the widespread distrust of steroids, and/or a belief that the eczema must be due to an underlying cause (such as allergy) which has not yet been identified, in addition to the time and cost of treatment. Despite attempts to allay some of the fears about the use of steroids (Long, Mills & Finlay, 1998) it is widely acknowledged that steroids are not acceptable to many parents as a long term treatment. Frustration with conventional medicine leads to the use of other, unregulated treatments. However, in one study of Chinese herbal treatments that families had obtained from complementary therapists, most of the creams were found to contain inappropriate amounts of steroids, unknown to the parents (Keane et al., 1999).

Review of Studies on the Psychological Impact of Childhood Eczema

The impact of childhood eczema on the quality of life of both the child and his or her family has been documented in several studies. For example, Lawson et al. (1998) identified 11 key areas of concerns for parents: practical care issues, psychological pressure on the parents, effect on family life-style, sleep disturbance, problems at school, effects on child behaviour, impact on social life, impact on relationships, inadequate medical support, additional financial burden and impact on holiday arrangements. Long, Funnell, Collard & Finlay (1993) found a similar range of difficulties amongst members of the National Eczema Society. Their postal survey showed that the following were the most common effects reported by parents of children with eczema: sleep disruption (60%), restrictions on sports (27%), difficulties at school (27%), difficulties with holiday arrangements (25%) and impact on playing with other children (14%).

Very few studies have specifically assessed the psychological impact of eczema on children and their families. Several studies have shown high rates of anxiety or distress in adults affected by eczema (Hughes, Barraclough, Hamblin & White, 1983) although it has not been shown how these high rates of distress relate to those for adults affected by any other physical illness. Jowett and Ryan (1985) carried out a structured interview with 100 adults affected by acne, psoriasis and eczema and vividly describe the social impact of skin disease. The interviews demonstrated the high levels of discomfort experienced by the respondents and the negative impact of their skin condition on personal relationships and self confidence. However, studies with adults with eczema cannot be generalised to children. As well as the usual difficulties in generalising across the age span, eczema itself is a very different condition in childhood than in adulthood (for example; it is much less common in adulthood and less likely to be self limiting).

As part of a large epidemiological study of child mental health problems Meltzer, Gatward, Goodman and Ford (2000) found that 1289 out of 10310 parents (12%) reported that their child had eczema. It was the second most common illness reported, with asthma being the most common (16%). Of this group of children with eczema, 11% were found to have a "mental disorder", defined using ICD-10 definitions of disorder with strict impairment criteria, compared to 10% of the healthy population. In comparison to all the other chronic illnesses studied, the children with eczema had the lowest proportion of mental health problems. This is an important finding because it is based on a very large community sample and because the study used very clear diagnostic criteria for mental disorder. However, it did rely on parental report of eczema and very high numbers of affected children were reported compared to other studies of the prevalence of eczema, which suggests that some of the eczema may have been extremely mild.

A study by Absolon, Cottrell, Eldridge and Glover (1997) found high levels of psychological difficulties among children attending dermatology out-patient clinics. However, there were no significant differences in the rate of mental health problems among children attending the clinic for the treatment of eczema, compared to a control group of children attending the same clinic for other mild conditions such as warts. When the children with mild eczema were excluded however, the children with moderate or severe eczema did have a significantly higher rate of psychological difficulties compared to the control group. High levels of distress were found in the mothers of both the children with eczema and the control children, but there were no significant differences between the two groups of mothers. Therefore, although mothers of children with eczema did report high levels of distress in themselves and their children it is not clear if this was due to eczema. On average, families attending out-patient paediatric clinics for any condition are likely to report high levels of concern, but this is partly due to the characteristics of the sample, which is highly selective.

This study also suggests that there may be a link between the severity of the child's eczema and the child's psychological adjustment. Although this has intuitive appeal, few studies of children with other types of physical illnesses have shown any link between the severity of the illness and psychological adaptation (Wallander & Varni, 1998).

Psychological Treatments for Eczema

There is some evidence that psychological intervention may be helpful in the management of chronic eczema. For example, behavioural techniques have been shown to help manage the symptoms associated with eczema, particularly scratching. Relaxation and guided imagery (De Horne, Taylor & Varigos, 1999) and habit reversal (Bridgett, Noren & Staughton, 1996) have been shown to be effective in the short term for adults with chronic atopic eczema. One study with children showed some reduction in lichenification (thickening) and surface damage to the skin in children taught relaxation and hypnotic techniques to control their eczema, compared to a no treatment control group (Sokel et al., 1993). Unfortunately the study had a high drop out rate, so these results are only preliminary. However, it may be possible to use some of the symptom management techniques developed for adults with older children, although it would be hard to do so with the youngest children.

Review of studies on Eczema and the Mother–Child Relationship

In addition to the psychological impact on the mother and child individually, it has long been assumed that the relationship between a mother and her child must both affect and be affected by the child's eczema. Eczema most commonly starts during the first few months of life and early studies suggested abnormalities in the developing relationship between the mother and her child. For example, Miller and Baruch (1948) described a pattern of either maternal rejection or overprotection towards allergic children (including children with asthma and hay fever as well as eczema). Bick (1986) described the analysis of a child with severe eczema and developed a psychodynamic theory of the importance of the skin in early object-relations. Her, controversial, descriptions of the importance of the mother's ability to "contain the infantile unintegrated state" (p 292) led to an interest in the skin as a symbolic, as well as a physical boundary, and the possible disruption to this function in cases of childhood eczema or other skin conditions.

Koblenzer (1990) has drawn attention to the importance of touch for normal development and describes how this becomes disrupted in children with severe eczema. She also describes clinical cases where the mother-child relationship has become very disturbed as a result of the child's eczema (Koblenzer & Koblenzer, 1988). In the clinical cases she describes, mothers went to extreme lengths to prevent their child scratching, for example, agreeing to unreasonable demands from the child and attempting to reduce the child's discomfort by constant soothing or avoiding conflict. The children are described as irritable and controlling with delayed developmental milestones. A common feature described was the child sleeping with the mother in an attempt to soothe the child and reduce disruption. As a result of constant exhaustion however, the mothers found it even harder to manage her child's behaviour. Koblenzer and Koblenzer (1988) interpret the mother's over solicitous reaction to her child's eczema as due to the mother's unconscious ambivalence towards her child and her unconscious fear that she herself may harm her child. As well as treating the eczema aggressively using conventional treatments, Koblenzer and Koblenzer (1988) used regular appointments to elicit these conflicting feelings towards the child to enable the mother to respond more appropriately to her child. They interpret the observed improvement in the child's skin as due to the mother's increased understanding of her need to "infantilize, overindulge and overstimulate the child" and the resulting change in her relationship with her child.

Although this formulation is based on psychoanalytic principles, the clinical picture is one also described by more behavioural clinicians, although the formulation is very different. In a behavioural formulation, scratching is seen as a habit, which may have initially developed as a consequence of itching but has become established as an automatic, habitual behaviour (Bridgett, Noren & Staughton, 1996). The child's scratching behaviour is reinforced by the attention the child attracts as a result of scratching, which most parents find hard to tolerate. The child is then often described as "manipulative" because of the way in which he or she scratches when distressed and succeeds in controlling the mother's response by this behaviour. Habitual scratching is then treated by constant monitoring and substitution of more appropriate activities, e.g. distraction or play.

These clinical descriptions are based on selected cases and no attempt has been made to assess whether they can be generalised to the whole population of children with eczema or whether they only apply to a small minority of memorable cases. However, studies that have used standardised assessment methods and more systematic sampling have failed to substantiate these claims for most mothers of children with eczema. For example, Solomon and Gagnon (1987) showed that whilst there were differences between dyads of mothers with healthy children and mothers of children with eczema, these differences did not fit with the descriptions in the literature. There was no evidence that the mothers of children with eczema were colder, more hostile or aggressive with their children or that they were more anxious than the control mothers. They touched, held, soothed and stroked their babies as much as mothers with healthy children. However, they did observe that mothers of children with eczema were less likely to be involved in exchanges of "spontaneous positive reinforcement" than the controls. This was described as fewer episodes where mother and baby would both spontaneously find enjoyment in something, as opposed to the mother responding to the child's overtures, or vice versa. Although this was a small study involving only 14 mother-child dyads, it is the only study to use a standardised observational scheme in a systematic way and not just rely on clinical anecdote. It is however limited because it is based on limited observation periods which may not generalise to natural interaction.

Daud, Garralda and David (1993) developed this work further and tested the hypothesis that young children with eczema are more insecurely attached to their mothers than healthy children, using the Strange Situation procedure (Ainsworth, Blehar, Waters & Wall, 1978). The study again showed a higher level of behavioural symptoms in the children with eczema compared to healthy controls on self report measures. In addition, the mothers of the children with eczema reported more difficulty in parenting tasks. However, they found no differences in the security of the child's attachment to his or her mother between the children with eczema and their control group of healthy children. Indeed, they observed that far from being rejecting and negative towards their children, the mothers showed a "positive, empathic" attitude towards them. This study suggests that mothers of children with eczema experience parenting tasks as more difficult than mothers of healthy children. In addition, the study found that both child behaviour problems and maternal distress were more common when the child had more severe eczema, which is in keeping with the Absolon et al. (1997) study. However, this study used a non-standardised measure of parenting difficulties that was constructed for the study, so it is difficult to determine the validity of the finding about difficulties with parenting. In addition it was based on a small sample size with a control group matched on only a few variables, so these results have to be interpreted with theses limitations in mind.

Many of the earlier psychodynamic models assume that the development of the child's eczema is at least partially, if not totally, caused by abnormalities in the mother child relationship. However, these explanations, have now largely been replaced by the more complex biopsychosocial model that allows for both biological and psychological processes as well as the reciprocal influences these are likely to have on each other (Engel, 1977). For example, Taylor (1985) proposes that the bodily condition both affects and is affected by the emotional state, with the child's relationship with the main carer (usually the mother) influencing both the child's physical and emotional health. This model acknowledges that whilst children with eczema do have physiological abnormalities that cause the eczema, the condition of the child's skin will also both affect and be affected by the child's emotional state and the child's relationship with his or her mother.

Howlett (1999) integrates some of the early object-relations theories with the biopsychosocial approach to the impact of eczema on a mother and child's relationship. He draws attention to the differences in sensation that a baby with eczema must experience, for example "intensive waves of itching" (p 382) and the importance of the mother's response in modifying these experiences. More importantly, he acknowledges the additional physical demands of caring for a child with eczema, which are frequently overlooked by psychological theories of eczema.

> "The impact on the child's appearance, the additional restlessness and irritability, more disturbed nights, the time-consuming nature of the skin-care routine and the mother's inability to cure or soothe her child, will all add to the stresses and exhaustion of motherhood. In addition, the special demands of caring for such a child make it more difficult for her to get a break by surrendering its care to others" (p 383).

The balance of evidence suggests that many of the descriptions of difficulties in the mother-child relationship were based on a small sample of clinical cases rather than a representative sample of most children with eczema. The few, systematic studies that have been carried out suggest that whilst some mothers do experience some difficulties in their relationship with their child, these are partly due to the demands of the condition itself, and that there is little evidence that difficulties in the mother-child relationship cause atopic eczema. However, there are few good studies which describe the difficulties experienced in parenting a child with eczema and there is some evidence that mothers of children with eczema do find parenting more difficult.

Pianta and Marvin's (1992) Resolution Theory

There have been few attempts to develop a model that attempts to explain a mother and child's psychological adjustment to illness in terms of the mother's beliefs about the illness. However, illness beliefs have been shown to be important in determining psychological adaptation to illness (Weinman, Petrie, Moss-Morris & Horne, 1996). It is likely that the mother's beliefs about the illness will affect her ability to manage the care required for her child, and will be important determinants of the extent to which her relationship with her child is affected by the illness. It is also more likely that these beliefs will be more important determinants of the psychological adjustment to illness, than features of the condition itself, e.g. the severity of the illness.

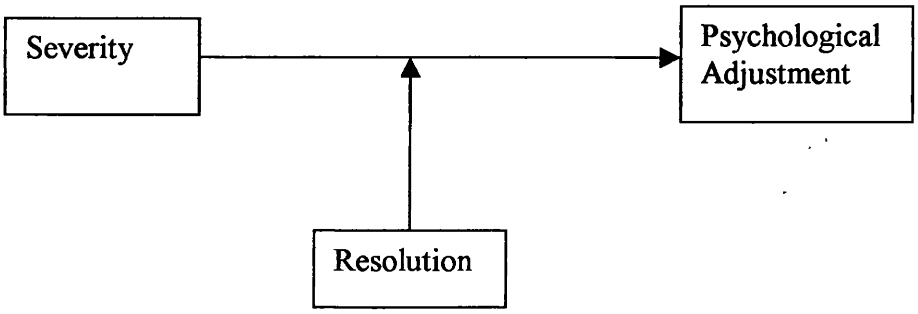
Pianta and Marvin (1992) have developed one such theory about the importance of the mother's adaptation to the diagnosis of chronic illness in a child and the impact this has on her relationship with her child (Pianta, Marvin, Britner and Borowitz, 1996). Their model is based on attachment theory (Bowlby, 1980), and focuses on the mother's internal representation of the caregiving system that regulates her behaviour towards her child. In normal development, the caregiving system develops in a complementary and highly integrated way with the child's attachment. However, being informed that your child has a chronic illness is a crisis that disrupts the caregiving system and challenges the mother's existing beliefs, feelings and expectations, and can be seen as analogous to a grief reaction or mourning. Resolution is defined as the process of adjustment to the diagnosis and the extent to which the mother has moved away from feelings of crisis to feelings of coping or acceptance of the child's difficulties (Pianta, Marvin, Britner & Borowitz, 1996). It is viewed as a process, because the extent of resolution is likely to vary over time, depending on the child's development and the family context.

This concept of "resolution" is very similar to the one that emerged as one of the four main themes in the qualitative study of children with EB (See Chapter Seven). Pianta and Marvin (1992) have developed a brief interview measure, the "Reaction to Diagnosis Interview" (RDI), designed to assess the extent to which a mother has become resolved with respect to her child's illness. The interview is usually videotaped and rated using a standardised coding scheme described in Pianta and Marvin (1992). They have demonstrated good inter-rater reliability for their coding system, as well as showing that it is strongly associated with parenting stress (Sheeran, Marvin & Pianta, 1997), and with the child's attachment status (Marvin & Pianta, 1996). Despite this, few studies have been carried out using this measure other than by the authors and hence replication is important.

The methodology used in the RDI is also sensitive to the mother's own viewpoint, because it is based on her narrative, and attempts to understand her own experience of the difficulties of caring for a child with eczema (Greenhalgh & Hurwitz, 1999). Many of the previous descriptions of the mother-child relationship have been very critical of the mother's attempts to care for her child, and have not explored the mother's view of why she has encountered such a high level of difficulty in managing her child. The RDI is designed to assess the mother's own view of impact of the diagnosis on herself and provides the opportunity for her to express her own difficulties.

The RDI was used in this study in order to test the hypothesis that mothers who are resolved about their child's illness have lower levels of psychological symptoms themselves, as well as lower levels of parenting stress. Resolution may also help explain the lack of consistency across studies in the relationship found between severity of illness and psychological outcome. It seems likely that the mother's beliefs about her child's illness may be more important determinants of psychological distress than factors related to the disease itself, such as severity. It is hypothesised that the mother's resolution may act as a moderating variable on the relationship between severity and outcome, as shown in Figure 9.1 below. It was predicted that the mothers who were resolved show the same level of psychological difficulty whether or not the child has mild or severe symptoms of eczema. However, for the mothers who are unresolved and who therefore continue to perceive eczema as a burden, there will be a correlation between severity and psychological adjustment.

<u>Figure 9.1 The Relationship Between Severity and Psychological Adjustment</u>



In addition, the study explored further some of the issues that have been raised by previous studies and highlighted in this review. These are whether the high levels of psychological difficulties reported by mothers of children with eczema are different from those reported by mothers of children attending paediatric clinics for other types of chronic health problems. In addition, the study explored whether mothers of children with eczema do experience different levels of parenting stress than other mothers, using a standardised measure of parenting stress. The strength of the relationship between severity of eczema and psychological adjustment in both the child and mother was measured. Both of the two main conceptual models discussed in Chapter One propose a link between disease severity and psychological outcome either directly or indirectly, and the strength of this association was measured in this study.

The study was designed to overcome some of the methodological limitations outlined in Chapter One. The measures used included an interview with the mother rather than relying only on self report measures. The child's own view of the impact of their eczema was included where possible by using a quality of life measure. All children develop eczema during the first two years of life and therefore the age of diagnosis did not vary much within the sample and a restricted age range was used to avoid some of the difficulties of generalising across a wide age range. Finally, the study focused on one specific medical condition to avoid the difficulties of the noncategorical approach.

Research Questions

The study addressed the following research questions:

1. Do children with eczema and their mothers experience different levels of psychological distress than children with other chronic, but non-life threatening, physical illnesses?
2. Do mothers of children with eczema experience different levels of parenting stress than mothers of children with other chronic, but non-life threatening, physical illnesses?
3. Is the severity of the child's eczema related to the child's psychological adjustment or to maternal psychological distress?
4. Do mothers who are resolved report lower levels of psychological symptoms than those who are unresolved?
5. Does the mother's resolution of her child's illness act as a moderating variable on the relationship between severity of the illness and psychological adjustment?

CHAPTER TEN

THE PSYCHOLOGICAL IMPACT OF ECZEMA

METHOD

Overview

This study consisted of a comparison between two groups of children with a chronic illness. In one group, the children were attending dermatology out-patient clinics because of chronic childhood eczema. In the other group, the children were attending general paediatric out-patient clinics for other long-standing but non life-threatening physical illnesses. The study compared the two groups on standardised measures of parenting stress and psychological distress in the mothers, and psychological symptoms in the child. In addition, the mothers of children with eczema were interviewed about their experience of having a child with eczema using a standardised interview, and these interviews were rated according to what extent the mother had become resolved about her child's condition. A measure of eczema severity was completed by an expert rater, and the child also completed a measure of the impact of eczema on their quality of life.

Participants

Eczema Group

Participants were a consecutive patient series recruited from the paediatric dermatology clinics at the University Hospital, Lewisham, and St John's Institute of Dermatology at St Thomas' Hospital, London. All children with a diagnosis of eczema, aged between four and 11 years, with a parent who was fluent in English, were eligible.

All the main carers in the study were mothers, although fathers also accompanied the mothers for six children.

Based on the previous literature a power calculation was carried out on the assumption of a medium effect size and it was calculated that 40 families would be needed per group (Cohen, 1992). Four families (8%) declined to participate in the research. Five families (10%) agreed to take part in the research but subsequently failed to return questionnaires, so were excluded from the sample. 40 families (82% of those approached) completed all stages of the research. 22 (55%) of the children with eczema were recruited from University Hospital, Lewisham and the remaining 18 (45%) from St John's Institute of Dermatology.

Comparison Group

Participants were recruited from the paediatric medical out-patient clinics at University Hospital, Lewisham. Families of children aged between four and 11, who were fluent in English and who were consulting a paediatrician about a chronic but non life-threatening illness in their child were eligible to take part in the study. Four mothers declined to take part in the research.

Table 10.1 summarises the demographic information about the participants in the eczema group and the comparison group. The eczema and comparison groups were very similar on most variables. The children in the comparison group were on average slightly older than the children with eczema, and this difference approached statistical significance (t(78) = 1.9, p = 0.06). However, age was not associated with any of the psychological measures (PSI, SDQ or GHQ) and therefore it was not necessary to enter it as a covariate in the analyses.

The comparison group had a relatively high number of unskilled families in social class 5, reflecting the high levels of economic deprivation within Lewisham, but was not significantly different from the eczema group. There was also a high proportion of single mothers within the comparison group compared to the eczema group. However, this difference was not significant on a χ^2 test. There was a high proportion of ethnic minority families in both comparison and the eczema groups, reflecting the multi-racial community in South East London.

Table 10.1 Demographic Characteristics of the Eczema and Comparison Group (Number and (%) unless stated otherwise)

	Eczema Group N = 40	Comparison Group N = 40
Age in Months		
Mean (SD)	79.93 (27.14)	91.25 (25.48)
Sex : Boys	13 (32.5)	15 (37.5)
Girls	27 (67.5)	25 (62.5)
Ethnic Group:		
White British	23 (57.5)	25 (62.5)
Afro-Caribbean	9 (22.5)	7 (17.5)
African	1 (2.5)	3 (7.5)
Asian	4 (10.0)	0 (0.0)
Other	3 (7.5)	5 (12.5)
Socio-Economic Group:		
1 - Professional	2 (5.0)	2 (5.0)
2 - Managerial	7 (17.5)	8 (20.0)
3 - Skilled	21 (52.5)	13 (32.5)
4 - Semi-Skilled	3 (7.5)	6 (15.0)
5 - Unskilled	7 (17.5)	11 (27.5)
Marital Status:		
Single Mother	7 (17.5)	13 (32.5)

Procedure

Eczema Group

The parent or parents accompanying the child were shown the Information Sheet and written consent was obtained (Appendix 7). The parent who was the main carer for the child was either interviewed in the clinic, or a time was arranged to carry out in the interview in the family home. All the main carers in the study were mothers. The questionnaires were either completed in the clinic or the mother was given a stamped addressed envelope to return them at her convenience. One reminder letter was sent if she then failed to return the questionnaires. The measure of severity of the child's eczema was completed by the research nurse from the skin unit or one of the Consultant Dermatologists at the time of the appointment (see measures section for further details).

Comparison Group

The mother accompanying the child was shown the information sheet (Appendix 8) and if they agreed to the research, they were given the questionnaires to complete while waiting in the clinic.

Ethical Approval

The Ethics Committee at University Hospital, Lewisham gave ethical permission for the study (Appendix 9).

Measures

Eczema group

The study included an interview with the child's mother, three questionnaires completed by the mother, one questionnaire completed by children over six years old and an objective rating of the severity of the child's eczema carried out by the research nurse working in the skin unit, or a Consultant Dermatologist.

Demographic Information: The interview began by obtaining demographic details, history and current treatment regime for the child's eczema and details of any family history of eczema (Appendix 10).

The Reaction to Diagnosis Interview (RDI: Piantä & Marvin, 1992) was used to assess the mother's adaptation to the child's eczema, in an adapted form. The RDI is a semi-structured, standardised interview, originally designed to assess the parents' resolution of the difficulties associated with receiving a chronic medical diagnosis for their child. The original interview consists of five major questions with supplementary probes. The modifications were made to reflect the nature of eczema, because for the vast majority of these families the specific diagnosis period is not a significant moment. Most children are diagnosed by the family GP, but many parents already recognise the symptoms of eczema prior to diagnosis, either because they themselves or someone else they know, has had eczema. The question prompting for detail about the experience of receiving the diagnosis itself was therefore deleted. Instead, a question was added asking about the course of the child's eczema, and whether it had got better, got worse or fluctuated over time since the diagnosis. An additional question was added which asked whether the parent felt that their relationship with the child had been affected by their child's illness. This was added to elicit information about the impact of the illness on the child's most important relationship, which would reflect the significance of impact of the illness on the child (Appendix 11).

The interviews were tape-recorded and transcribed and coded according to the coding scheme described by Pianta and Marvin (1992). The two main classifications (Resolved or Unresolved) were used, but the nine sub-classifications were not used. All transcripts were coded twice: by the author and by an independent rater who was also a clinical child psychologist (Appendix 12).

Pianta, Marvin, Britner and Borowitz (1996) report very high levels of inter-rater reliability for the coding system using the two main codes "resolved" and "unresolved" (92 – 96% agreement) as well as high levels of inter-rater reliability for the nine subcategories (84 – 97% agreement). In addition, they have shown that rates of resolution did not differ for two different types of chronic illness (epilepsy and cerebral palsy) and the resolution was not related to severity of the illness or time since diagnosis.

General Health Questionnaire (GHQ – 28: Goldberg & Williams, 1988). The GHQ assesses psychological symptoms and general well-being and is composed of 4 factors: somatic symptoms (e.g. "Have you recently been feeling rundown and out of sorts?"), anxiety and insomnia (e.g. "Have you recently lost much sleep over worry?"), social dysfunction (e.g. "Have you recently been managing to keep yourself busy and occupied?") and severe depression (e.g. "Have you recently felt that life is entirely hopeless?"). These are rated using a four point descriptive scale and scored according to the GHQ scoring system of 0 for no symptoms and 1 for mild or severe symptoms. A threshold of 5 is most commonly used to indicate psychiatric "caseness". This scale has been used extensively and many validity and reliability studies have been published (Boardman,1987; Cox et al., 1987).

Parenting Stress Index – Short Form (PSI: Abidin, 1995). This is a 36 item questionnaire which is designed to assess stress within the parenting system. It consists of three sub scales, Parental Distress (e.g."I often have the feeling that I cannot handle things well"), Parent-Child Dysfunctional Interaction (e.g."My child rarely does things for me that make me feel good") and Difficult Child (e.g."My child seems to cry or fuss more often than other children"). The parent chooses from a five point scale ranging from 1 = "strongly disagree" to 5 = "strongly agree" for each question. The original long form of the PSI, with 101 items, has been used widely, and the published norms are based on a large sample. The 36 item short form has been less extensively researched, although Abidin (1995) reported that the same three-factor solution adequately describes both the Short Form and the Long Form and norms are available based on a sample of 800 families. Global coefficient alpha of the short form is reported at 0.95 by Abidin (1995) and 0.91 by Sheeran et al., (1997).

Strengths and Difficulties Questionnaire (SDQ: Goodman, 1997). This is a 25 item questionnaire which covers the main areas of psychological difficulties in children. It consists of five sub-scales: emotional symptoms, hyperactivity, conduct problems, peer relationship difficulties and prosocial behaviour. For each item, the parent reads a brief statement (e.g. "Constantly fidgeting and squirming", "Many worries, often seems worried") and then chooses from three options: "not true" of my child, "somewhat true" or "certainly true". The four symptom sub-scales can be summed to give an overall score of symptoms. The prosocial behaviour score is not included in this total score.

There is an impact supplement to the SDQ that has been shown to discriminate well between community and clinic samples (Goodman, 1999). The impact supplement is useful in determining the impact of the child's symptoms on every day situations, the level of distress experienced by the child and the burden on the family as a whole. This is included in order to improve discrimination between children who have a high level of symptomatology but no associated social impairment, from those who have significant impairment as a consequence of their symptoms.

Several reliability and validity studies have now been conducted (Goodman, 1999; Goodman & Scott 1999; Meltzer, Gatward, Goodman & Ford, 2000) and the measure has been shown to be a reliable and valid scale for screening for psychological difficulties in childhood. A score above 16 on the total score for the parent report version is used as threshold for psychological difficulties. A score of 14-16 is categorised as "borderline". This scale is easy to complete and is acceptable as a screening measure because it includes positive as well as negative items.

<u>The Children's Dermatology Life Quality Index</u> (CDLQI: Lewis-Jones & Finlay, 1995). Children aged 6 or older completed the CDLQI themselves, with help, if necessary, with reading from the researcher. This is a ten item questionnaire which was designed to assess the impact of skin conditions on the quality of life of children (Appendix 13). It consists of items describing symptoms of eczema (e.g. "Over the last week, how itchy, scratchy, sore or painful has your skin been?") with possible responses of "not at all", "only a little", "quite a lot" or "very much". This questionnaire was included in order to ensure that the child's view of the impact of his or her eczema was taken into account if

possible. It is not possible to use the CDLQI reliably with children under the age of 6. Initial reliability and validity data have been published but the measure has not been widely used yet (Lewis-Jones & Finlay, 1995).

Six Area, Six Sign Atopic Dermatitis severity score (SASSAD: Berth-Jones, 1996). It has proved very difficult to develop a reliable measure of eczema severity that can be easily used in a clinic setting (Finlay, 1996). This is partly due to the intrinsic variability in the appearance of eczema depending on temperature and other conditions. However, it is also very difficult to estimate reliably the amount of surface area of the body affected by eczema because the margins between affected and unaffected areas are not clearly delineated (Charman,Venn & Williams, 1999). This measure was developed to help provide an accurate rating of severity of eczema without relying on body surface area estimates (Appendix 14). It consists of the assessment of six signs of eczema; erythema, exudation, excoriation, dryness, cracking and lichenification on six different areas of the body; head and neck, hands, arms, trunk, feet and legs. The scores are then summed to give an overall severity score ranging from 0 to 108. This measure was completed by one of two trained raters: the Research Nurse attached to the Skin Unit or one a Consultant Dermatologist at Lewisham Hospital.

Comparison Group

The parents in the comparison group completed a brief, anonymous questionnaire giving information about the child's age, reason for attending the clinic and basic demographic information, such as ethnic background, family composition and employment status (Appendix 15). They also completed the GHQ, the SDQ and the PSI.

CHAPTER ELEVEN

THE PSYCHOLOGICAL IMPACT OF ECZEMA

RESULTS

The descriptive findings from the study for all the measures, and the relationship between the medical, demographic and psychological measures are given first. Then each of the research questions is addressed in turn. The raw scores for all the measures were plotted out and reviewed to determine whether it could be assumed that there was an approximately normal distribution. Unless stated otherwise for a specific measure, the data was normally distributed.

Description of Samples

There was strong evidence of a family history of eczema in the families of children with eczema, as would be expected. 22 (55 %) of the children with eczema had a mother or father with eczema, and of the 33 children who had a sibling, 13 (39%) had a sibling with eczema. 13 (33 %) of the children with eczema had had an admission to hospital because of their eczema. 76% of the children had developed eczema by six months of age, the modal age of onset being 3 months. 14 % developed eczema between seven months and one year of age, and the remaining 10 % developed eczema between one and two years of age. On average, the children had had eczema for 6 years.

Eczema severity varied considerably and this is reflected in the wide variation in the SASSAD scores, which ranged from 7 to 75, with a mean of 35.9 (SD 17.1). 24

children were aged 6 or over and completed the CDLQI. The scores ranged from 0 to 20, with the mean 9.5 (SD 5.5).

The comparison group were attending the paediatric clinic for a wide variety of reasons. The most common reasons were recurrent urinary tract infections (8), stomach aches (5), concerns about weight or growth (5), enuresis (4), constipation (2) and a variety of other reasons, e.g. headaches, feeling sick, asthma, difficulties with balance.

Relationship Between Demographic and Psychological Measures

Socio-economic status was positively associated with SDQ score for both the eczema and the comparison groups ($r = 0.34$, $p < 0.05$ and $r = 0.30$, $p = 0.06$ respectively). There were significant differences between the mean SDQ scores for single parents compared to parents who were married or cohabiting ($t (78) = 2.09$, $p < 0.05$).

Relationship Between Medical Measures and Psychological Measures

The mean SDQ and GHQ scores were not significantly different for children who had had an admission to hospital for eczema or a family history of eczema compared to those who had not. However, the PSI scores for parents of children who had had a hospital admission for eczema were significantly higher than those for children who had never had a hospital admission for their eczema ($t (38) = 3.04$, $p < 0.01$).

Differences Between Referral Sites

The scores for all the demographic, medical and psychological measures were compared for the two different referral centres. There were no differences between the two sites in terms of the severity of the child's eczema as measured by the SASSAD. However, the children referred from Lewisham rated their eczema as interfering more with their quality of life on the CDLQI, than children from St Thomas' (t(22) = 2.04, p = 0.05). The children recruited from St Thomas' were slightly older than children referred from Lewisham (t(38) = 2.15, p = 0.04). The scores on the PSI parent/child dysfunctional interaction scale were higher at St Thomas' compared to Lewisham (t(38) = 2.05, p = 0.05).

Correlations Between Psychological Measures

Table 11.1 shows the correlation coefficients between the psychological measures for the eczema group and Table 11.2 shows the correlation coefficients for the comparison group.

The three different sub-scales of the PSI were strongly associated with each other, and, as would be expected, with the total PSI score. There was a significant correlation between the PSI and the GHQ. There was also a strong correlation between two of the three sub-scales of the PSI ("Parent-Child Dysfunctional Interaction" and "Difficult Child") and the total PSI score with the SDQ for the eczema group, and for all the sub-scales for the comparison group. The GHQ and the SDQ were not significantly correlated for the eczema group although they were for the comparison group.

Table 11.1 Correlations Between the Psychological Measures for Eczema Group (N=40)

	PSI – Parental Distress	PSI – Parent/Child Dysfunction	PSI - Difficult Child	PSI – Total	GHQ	SDQ Total
PSI – Parental Distress	1.0	0.61 **	0.52 **	0.83 **	0.45 **	0.29
PSI – Parent/Child Dysfunction		1.0	0.71 **	0.89 **	0.34 *	0.51 **
PSI – Difficult Child			1.0	0.87 **	0.44 **	0.59 **
PSI – Total				1.0	0.48 **	0.53 **
GHQ					1.0	0.29

* $p < 0.05$, **$p<0.01$

Table 11.2 Correlations Between the Psychological Measures for Comparison Group (N = 40)

	PSI – Parental Distress	PSI – Parent/Child Dysfunction	PSI - Difficult Child	PSI – Total	GHQ	SDQ Total
PSI – Parental Distress	1.0	0.72 **	0.68**	0.88**	0.67 **	0.43 **
PSI – Parent/Child Dysfunction		1.0	0.79 **	0.91**	0.56 **	0.54 **
PSI – Difficult Child			1.0	0.92 **	0.43 **	0.71 **
PSI – Total				1.0	0.61 **	0.63 **
GHQ					1.0	0.38 *

* $p < 0.05$, **$p<0.01$

Research Question One: Do children with eczema and their mothers experience different levels of psychological distress than children with other chronic, but non-life threatening, physical illnesses?

Table 11.3 shows the mean scores on the Strengths and Difficulties Questionnaire (SDQ) for the two groups, separately for boys and girls, together with the norms for the measure based on a large sample from the general population. These means were compared using ANOVA, and the results are shown in Table 11.4.

Table 11.3 Mean Scores for the Strengths and Difficulties Questionnaire

SDQ sub-scale		Eczema Group		Comparison Group		Norms	
		M	SD	M	SD	M	SD
Conduct Problems	Girls	2.3	(1.8)	1.7	(1.9)	1.5	(1.5)
	Boys	2.4	(1.8)	3.0	(3.1)	1.8	(1.8)
Emotional Problems	Girls	3.7	(2.1)	3.4	(2.5)	2.0	(1.9)
	Boys	3.2	(2.2)	3.3	(2.8)	1.8	(2.0)
Hyperactivity	Girls	4.6	(2.9)	3.7	(1.9)	3.1	(2.5)
	Boys	4.9	(2.1)	4.7	(3.6)	4.1	(2.8)
Peer Problems	Girls	2.6	(1.9)	1.9	(1.6)	1.3	(1.6)
	Boys	2.4	(1.5)	2.7	(2.2)	1.5	(1.7)
Pro-social	Girls	8.0	(2.1)	9.0	(1.2)	8.9	(1.4)
	Boys	7.4	(1.8)	7.9	(1.7)	8.4	(1.7)
Total Problem Score	Girls	13.2	(6.4)	10.7	(6.2)	7.9	(5.4)
	Boys	12.4	(4.9)	13.7	(9.0)	9.3	(6.0)

<u>Sample sizes</u> Eczema group: girls n = 27, boys n = 13
Comparison Group: girls n = 25, boys n = 15
Norms: girls n = 2954, boys n = 2901

Table 11.4 Results of ANOVA on the Strengths and Difficulties Questionnaire

Sub-scale	Effect	df	MS	F	
Conduct	Sex	1	133.54	48.21	***
	Group	2	14.89	5.38	**
	Interaction	2	8.89	3.21	*
	Error	5929	2.77		
Emotional	Sex	1	71.81	18.75	***
	Group	2	95.13	24.84	***
	Interaction	2	0.75	0.20	
	Error	5929	3.83		
Hyperactivity	Sex	1	1367.29	193.94	***
	Group	2	27.85	3.95	*
	Interaction	2	4.98	0.71	
	Error	5929	7.05		
Peer Problems	Sex	1	53.64	19.65	***
	Group	2	38.31	14.03	***
	Interaction	2	2.58	0.95	
	Error	5929	2.73		
Prosocial	Sex	1	371.15	152.74	***
	Group	2	14.18	5.84	**
	Interaction	2	2.22	0.91	
	Error	5929	2.43		
Total Problem Score	Sex	1	2743.89	83.78	***
	Group	2	576.83	17.61	***
	Interaction	2	35.77	1.09	
	Error	5929	32.75		

* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$

The Newman-Keuls test with the Tukey Kramer adjustment for unequal sample sizes was used to identify which of the means were significantly different (Howell, 1997). The results of these will be discussed for each sub-scale in turn.

Conduct sub-scale: Boys had higher scores than girls within both the chronic illness comparison group, and the general population group on which the norms are based. In addition, the score for boys in the chronic illness comparison group, was higher than the norms. There was an interaction between gender and group. Boys in the chronic illness comparison group had higher scores than boys in either the eczema or general population groups, whereas girls in the chronic illness comparison group had lower scores than girls in the eczema group.

Emotional sub-scale: Girls had higher scores than boys within the general population group. Girls and boys within both the eczema and the chronic illness comparison group had higher scores then the norms.

Hyperactivity sub-scale: The main effect due to sex was again due to boys having higher scores than girls within the general population group. Girls with eczema had higher scores than the norms for the general population.

Peer Problems: Again, within the general population group boys had higher scores than the girls. Girls with eczema had higher scores than girls in the general population group. Boys in the chronic illness comparison group had higher scores than the norms.

Prosocial sub-scale: (N.B. High scores represent good prosocial skills on this scale). Girls had higher scores than boys in both the general population group and the chronic illness comparison group. Girls with eczema had lower scores than either the norms or the chronic illness comparison group.

Total Problem Score: Within the general population group, boys had higher scores than girls. Girls and boys in both the eczema and the chronic illness comparison groups had higher scores than the norms.

Summary of Differences Between Eczema and Comparison Group

There were no significant differences between the eczema and comparison groups on the total problems score on the SDQ or on the conduct, emotional, hyperactivity or peer problems sub-scales. However, the girls with eczema were rated as significantly lower on the prosocial scale of the SDQ than girls in the comparison group. This scale measures the mother's rating of positive, helpful behaviour by her child, for example, being kind to younger children, volunteering to help others, being considerate of others' feelings. A high score on this scale reflects helpful behaviour and the maximum score is 10. (This score does not contribute to the total SDQ score, which is based on the 4 problem behaviour scales). Only 2 girls (7%) scored below the threshold for this score, but the distribution of the scores on the prosocial scale of the SDQ was skewed towards lower scores when compared to the distribution for the comparison group.

Summary of Differences Between Eczema Group and Norms

The total scores for both boys and girls in the eczema group were significantly higher than the norms for the SDQ. The effect sizes for these differences were 0.83 for girls, and 0.52 for boys.

The mean scores for the girls with eczema were significantly different from the norms for all of the sub-scales except conduct problems. The mean scores for the boys with eczema were significantly different from the norms for the emotional scale, but not for the conduct, hyperactivity, peer difficulties or prosocial scales.

12 (30 %) of the children with eczema, and 11 (27.5%) of the comparison group children scored above the threshold of 16 (which is used for screening for children who are likely to have psychological problems) compared to 10 % of the children in the general population. However, a slightly higher rate of problems on the SDQ would be expected in this sample compared to a national sample because of the relatively low SES of families in both the groups. As a more direct comparison, a needs assessment carried out in the local area found that 17.9 % of a sample of children in Lewisham scored above the threshold on the SDQ (Attride-Stirling, Davis, Day & Sclare, 2000). When compared with the children with eczema using χ^2, the difference in the rates for the community sample compared to the eczema sample, approached significance ($\chi^2 = 3.46$, $p \approx 0.07$).

Gender Differences

There were no significant differences between scores on any of the sub-scales for boys and girls in the eczema group. This is in contrast to the norms where there are significant differences in scores for boys and girls for each of the sub-scales and the total score. However, the effect sizes of these differences are small.

Impact Supplement of SDQ

The SDQ includes an impact supplement that allows the respondent to indicate whether they feel that their child has any emotional, behavioural or concentration difficulties, and the impact of these difficulties on the child, the family and school life. There was a significant difference between the eczema and comparison groups on this impact supplement. Table 11.5 shows the frequency of responses to the first question of the impact supplement which asks the parent whether they consider the child to have any emotional, behavioural or concentration difficulties. The difference between the comparison group and the eczema group scores on this question was significant (Mann Whitney U = 610, z = 1.99, $p < 0.05$).

More parents in the eczema group than the comparison group identified their child as having difficulties, despite the fact that there was no significant difference between the total symptom scores for these two groups. Most of the problems identified in the eczema group were described as "minor" but there was a much higher rate of "minor" problems than in the comparison group. The proportion of parents rating their child's problem as definite or severe (17.5%) was about the same as those in the Glazebrook et al. (2001) study of children attending paediatric out-patient clinics.

Table 11.5 Response to Difficulties Question of SDQ Impact Supplement

	Eczema Group		Comparison Group	
	N	%	N	%
None	14	(35.0)	25	(62.5)
Minor	19	(47.5)	9	(22.5)
Definite	5	(12.5)	3	(7.5)
Severe	2	(5.0)	3	(7.5)

GHQ-28

There were no significant differences between the levels of maternal distress, as measured by the GHQ between the comparison group and the eczema group. The mean score for the eczema group was 5.1 (sd 6.2) and the mean score for the comparison group was 5.6 (sd 6.9). 45 % of the mothers in the eczema group scored 5 or above, the threshold for "caseness" for this measure. In the comparison group, 40% of mothers scored above the threshold. Although these rates are higher than the rate of 33% reported by women in a large community study (Cox et al., 1987) there was no significant difference between these rates on a χ^2 test.

In summary, this study found that the levels of psychological symptoms in the child and the levels of psychological distress in the mothers did not differ significantly between the eczema and the comparison groups. However, the children in both the eczema and comparison groups had high levels of psychological symptoms compared to the general population. The usual gender differences in the types of difficulties experienced by the children were not apparent in the eczema group.

The girls with eczema were rated as significantly less prosocial compared to the comparison group and the norms. Despite the fact that there were no overall differences in the levels of psychological symptoms for the two groups, significantly more of the mothers of children with eczema reported concerns about their child's mental health compared to the comparison group.

Research Question Two: Do mothers of children with eczema experience different levels of parenting stress than mothers of children with other chronic, but non-life threatening, physical illnesses?

The mean scores on the PSI for the eczema group and the comparison groups are compared in Table 11.6. One way ANOVA was used to compare the mean scores on the total score and the sub-scales. Norms for boys and girls separately are not available for the PSI.

The scores on the PSI for the eczema group were high compared to the norms for this measure on all three sub-scales and the total score. For the eczema group, the mean PSI total score corresponds to the 85th percentile of the published norms, as does the score on the difficult child sub-scale. For both the parental distress and the parent-child dysfunctional interaction sub-scales, the mean scores correspond to the 70th percentile.

Table 11.6 Mean scores and ANOVA for Parenting Stress Index

PSI Sub-scale	Eczema (n = 40)		Comparison (n = 40)		Norms (n = 800)		F (2, 877)
	M	SD	M	SD	M	SD	
Parental Distress	29.5	(9.5)	27.8	(9.7)	25.0	(8.0)	7.69 **
Parent-Child Dysfunction	23.1	(7.9)	20.4	(8.4)	19.0	(7.0)	6.85 **
Difficult Child	33.4	(10.1)	27.2	(10.9)	25.0	(8.0)	20.54 ***
PSI TOTAL	86.1	(23.7)	75.3	(26.2)	69.0	(17.0)	19.17 ***

** $p < 0.01$ ***$p < 0.001$

The Newman-Keuls test with the Tukey Kramer adjustment for unequal sample sizes was used to identify which of the means were significantly different (Howell, 1997).

The scores for the chronic illness comparison group were not significantly higher than the norms for any of the sub-scales or the total score.

The scores for the eczema group were significantly higher than the norms for all the sub-scales and the total score. In addition, the scores for the eczema group were significantly higher than the chronic illness comparison group for the parent–child dysfunctional interaction sub-scale, the difficult child sub-scale and the total score.

Therefore, the study has shown that the mothers of children with eczema reported higher rates of parenting stress than the mothers in the comparison group of children with other chronic illnesses. This difference was significant for the total score and the difficult child and parent-child dysfunctional interaction sub-scales of the PSI.

Research Question Three: Is the severity of eczema related to the child's psychological adjustment or the mother's psychological distress ?

The two measures of eczema severity, the "objective" rating by the research nurse or dermatologist (SASSAD), and the child's own rating of the impact of their eczema on their quality of life (CDLQI) were weakly associated ($r = 0.38$, $p = 0.07$). Although this is only a trend towards significance, it was based on the small number of children (24) who were old enough to complete the CDLQI.

Table 11.7 shows the correlations between these two measures of the severity of the child's eczema and the measures of psychological adjustment in the child and his or her mother and the measure of parenting stress.

Table 11.7 Correlations between Measures of Severity and Psychological Adjustment

	PSI – Parental Distress	PSI – Parent-Child Dysfunction	PSI – Difficult Child	PSI – Total	GHQ	SDQ
SASSAD (n = 40)	.13	.34 *	.23	.27	.17	.13
CDLQI (n = 24)	.23	.13	.34	.29	.21	.45 *

* $p < 0.05$

The severity of the child's eczema (SASSAD) was correlated with the parent-child dysfunctional interaction sub-scale of the PSI. However, the severity of the child's eczema (SASSAD), was not significantly associated with the mother's distress (GHQ) or with the child's psychological functioning (SDQ).

The child's rating of the impact of the eczema on their quality of life (CDLQI) was related to the child's psychological functioning (SDQ). However, it was not significantly related to maternal distress (GHQ), nor to parenting stress (PSI).

In order to clarify the relationship between the severity of eczema and psychological functioning, the SASSAD scores were split about the median and the means for the upper and lower halves compared using a t-test for the SDQ and PSI and a Mann Whitney U for the GHQ. The results are shown in Table 11.8.

This suggests that whilst the severity of the child's eczema was not significantly related to the mother's general distress or to the child's general psychological functioning, it was related to the mother's level of parenting stress, but the effect size was small to moderate.

Table 11.8 Differences between Psychological Measures for Median Split of SASSAD

	SASSAD < 33 N = 20		SASSAD > 33 N = 20		Test for Significance
	M	SD	M	SD	
GHQ	4.2	5.5	5.9	6.8	NS
PSI	77.4	14.6	94.8	27.9	t(38) = 2.5 *
SDQ	12.3	6.0	13.6	6.0	NS

* $p < 0.05$

This study found no relationship between psychological symptoms in the child or psychological distress in the mother with the severity of the child's eczema as rated by an expert rater. However, the child's own rating of the impact of the eczema on his or her quality of life was associated with child psychological symptoms.

Research Question Four: Do mothers who are resolved report lower levels of psychological symptoms than those who are unresolved?

The transcripts of the interviews with the mother were coded by the author and by one independent rater, who was also a clinical child psychologist. A 6 point scale ranging from 1 = "Clearly Unresolved", to 6 = "Clearly Resolved" was used (see Appendix 12 for Coding Instructions). The coding instructions were adapted form those given by Pianta and Marvin (1992), with some alterations due to the adapted form of the interview used in this study. Transcripts for four interviews could not be used because the tape recorder failed to record part, or all, of the interview. Therefore, these calculations are based on 36 interviews. Ratings of 1-3 inclusive were categorised as "unresolved" and 4-6 as "resolved".

Good inter-rater reliability was achieved for the main category "resolved" or "unresolved" (85% exact agreement, Kappa = 0.7). For the 6 point scale, the exact agreement rate was 44% and agreement within one point of the six point scale was 74%. There was a high correlation between the scores of the two raters (r = 0.73). The transcripts which were originally rated as in different main categories were discussed by both raters and a consensus rating agreed on. This increased the inter-rater reliability (r = 0.87).

The following quotations give some indication of the range of this dimension.

Extracts from interviews coded as 'resolved':

> He is quite happy so, obviously I'd like to see him with clear skin but as long as he is confident and happy we can deal with it.
>
> I mean obviously it's not nice but it can be controlled to a certain extent. It could be like a permanent disability that she's not going to grow out of. And obviously its just the same as whether you have got blue eyes or whatever. You've got no control over it so there is no point worrying about it really.
>
> Some get asthma, some get...I think I'm lucky it's just eczema. There are so many other things out there that children have got.
>
> Well sometimes when she's really bad, my friends see her and they get all upset, 'Oh look at her little hands' and that, but I don't. I do, but I cut off a bit now. I'm used to it.

Extracts from interviews coded as 'unresolved':

> Its just everything, I don't know. It's like you're in there on your own, there's you...all right you've got your family with you but there's nothing...You know if you broke your arm you'd take the cast off then your arms mended so to speak, but there's no light sometimes at the end of the tunnel with him.
>
> I can't bear the sound of him scratching, I can't bear it.
>
> I just want my doctor to say the magic word and then he gets better. He has never slept the whole night you know. Even me, I have never had a...Every single night I wake up, it's every single night.

> We feel that we are getting nowhere. It just seems like whatever you do it doesn't make it any better... It just seems like whatever you do it's not enough, it's never enough.
>
> I hate it, it's horrible. It's horrible. I wouldn't wish it on anybody. It marks your child, it makes your child look like they are not perfect. It's not fair you know.

18 transcripts were rated as "resolved" and 18 as "unresolved". The differences between these two groups on the main measures are shown in Table 11.9. Independent t-tests were used for all the measures except the GHQ where the Mann Whitney U was used.

The unresolved group had significantly higher scores than the resolved group on the total PSI score, as well as the Difficult Child and Parent-Child Dysfunctional Interaction sub-scales. There was no statistically significant difference between the two groups on the total SDQ score or on any of the individual problem sub-scale scores. However, the resolved group was higher on the SDQ prosocial scale. The unresolved group had significantly higher GHQ scores than the resolved group. It is interesting to note that the scores for the resolved group on all of these measures on which there is a significant difference between the resolved and unresolved groups, are very similar to the scores for the comparison group, shown in Table 11.5.

There were no statistically significant differences between the unresolved and resolved groups on the eczema severity rating (SASSAD) or the child's rating of the impact of their eczema on their quality of life (CDLQI). There were no significant differences between the two groups in terms of family history of eczema, age, length of time the child had had eczema, marital status of the mother, referral centre or ethnic background. There were equal numbers of boys and girls in both the resolved and the unresolved groups.

Table 11.9 Differences between Resolved and Unresolved groups on Psychological Measures

	Resolved		Unresolved		Test of Significance
	M	SD	M	SD	
Strengths and Difficulties Questionnaire:					
Conduct Problems	1.9	(1.8)	2.9	(1.8)	NS
Emotional Problems	3.5	(2.1)	4.0	(2.2)	NS
Hyperactivity	4.4	(3.2)	5.4	(2.0)	NS
Peer Relationship Problems	2.2	(1.3)	2.9	(2.2)	NS
Prosocial	8.5	(1.3)	6.7	(2.1)	t (34) = 3.1 **
SDQ – Total problems	11.9	(5.6)	14.9	(5.9)	NS
GHQ	3.2	(4.8)	7.9	(6.9)	U = 4.5, z = 2.2*
Parenting Stress Index:					
Parental Distress	26.9	(6.2)	31.4	(11.6)	NS
Parent/ Child Dysfunction	20.2	(4.7)	25.9	(8.7)	t (34) = 2.5 *
Difficult Child	29.3	(8.7)	38.3	(9.8)	t (34) = 2.9 **
PSI Total	76.4	(14.9)	95.7	(26.4)	t (34) = 2.7 *

* $p < 0.05$ ** $p < 0.01$

It would have been interesting to be able to determine to what extent the measure of resolution independently predicted parenting stress over and above the general measure of distress (GHQ), which would in theory be possible using multiple regression techniques. However, this study was cross sectional in design and as shown in Table 11.1 the measures were strongly correlated with each other. In addition, it was not possible to determine the direction of causality from the study. For example, it is likely that a mother's general level of distress (as measured by the GHQ) will influence her rating on the RDI, however, it is also likely that a mother's resolution will influence her overall feeling of distress. The direction of causality could only be determined using a predictive or experimental design and it is highly likely that these are reciprocal influences. Therefore it was not possible to use multiple regression techniques to determine the relative importance of resolution independently from GHQ (Allison, 1999). However, it would be interesting to determine this in further research, using an appropriate research design.

Therefore this study did show that there were higher levels of psychological symptoms in the mothers and children in the unresolved group compared to the resolved group. This difference was significant for both the GHQ and the PSI, but not the SDQ. There were no significant differences between the resolved group and the unresolved group on variables related to the eczema itself (length of time child had had eczema, severity of eczema or family history of eczema) or demographic variables (marital status of mother or ethnic background).

Research Question Five: Does the mother's resolution of her child's illness act as a moderating variable on the relationship between severity of the illness and psychological adjustment?

In order to test if resolution acted as a moderating variable on the relationship between severity of illness and parenting stress, a multiple regression was carried out following the recommendations of Holmbeck (1997). The main effects were entered first into the multiple regression, followed by the interaction between severity and resolution. No significant moderating effects were found for the relationship between severity of eczema and either maternal distress or psychological symptoms in the child.

This study found no evidence of resolution acting as a moderating variable between severity and psychological symptoms.

CHAPTER TWELVE

THE PSYCHOLOGICAL IMPACT OF ECZEMA

DISCUSSION

Summary of Results

This study found that children with eczema had high levels of psychological symptoms compared to the norms for the measures used. There were no significant differences between the levels of psychological distress expressed by the mothers of children with eczema compared to the norms for a community sample. In addition, there were no significant differences between the levels of psychological symptoms in the children with eczema and their mothers compared to a group of children attending paediatric out-patient clinics for other non life-threatening chronic conditions.

However, the mothers of children with eczema expressed a higher level of concern about their child's mental health than the mothers of the children attending general clinics and the levels of parenting stress reported by mothers of children with eczema were higher than for the comparison group.

Psychological distress in the child or mother was not significantly related to the severity of the child's eczema. However, severity of eczema was weakly associated with levels of parenting stress reported by the mother.

Half of the mothers were found to be resolved about their child's eczema, and half were found to be unresolved. The unresolved mothers reported higher levels of psychological distress and higher levels of parenting difficulties than the resolved group. The length of time the child had had eczema and the severity of eczema did not differ between the resolved and unresolved groups. The prediction that resolution would act as a moderating variable on the relationship between severity of eczema and psychological distress was not supported.

Evaluation of Results

The findings from this study are consistent with those of previous studies that children with chronic eczema have higher rates of psychological symptoms than healthy children. However, the levels of psychological symptoms in both the children with eczema and their mothers were not significantly different from those reported by mothers of children with other chronic, but non life-threatening physical conditions attending general paediatric clinics. These findings have to be seen within the context of the small sample size. The effect size of the difference between the children with eczema and the norms was large (0.89 for girls, 0.57 for boys) but the effect size for the difference between the children with eczema and the comparison group was moderate (0.40 for girls) and small (– 0.19 for boys).

However, some interesting differences emerged between the children with eczema and the comparison group of children with other chronic conditions. Firstly, the mothers of children with eczema reported higher levels of concern about their child's emotions or behaviour, as reflected in significantly higher ratings on the impact supplement of the SDQ. In addition, girls with eczema were rated as significantly lower on the prosocial scale of the Strengths and Difficulties Questionnaire (SDQ) than girls in the comparison group. Therefore, although the children with eczema did not have significantly higher levels of psychological symptoms than the children in the comparison group, the mothers did have a higher level of concern about their emotions or behaviour, and rated the girls as less helpful and co-operative. There was also a significant difference between the levels of parenting stress reported by the mothers of children with eczema, compared to the comparison group and to the norms for this measure.

The high levels of psychological symptoms in the children in both groups in this study may be partly accounted for by the relatively low overall socio-economic status of the two groups. SES has been shown to be related to child psychological symptoms (Meltzer et al., 2000). However, this sample also represents a narrow spectrum of the population of children with eczema or with on-going medical problems. The sample is based on children attending paediatric clinics and is therefore not representative of the community as a whole. In other studies of children attending dermatology clinics, similarly high rates of psychological distress have been reported (Absolon et al., 1997; Daud et al., 1993). However, in a large community study, which would have included children across the whole range of severity, much lower rates of psychological distress were reported (Meltzer et al., 2000). Therefore, the high rates of psychological symptoms found in this study support the findings from other studies of the out-patient population but cannot be generalised to all children with eczema.

In the eczema group, the gender differences in the types of psychological difficulties presented that are usually found in community samples, were not apparent. The girls had similar levels of conduct problems and symptoms of hyperactivity as the boys. However, because of the small sample size in this study, these results have to be seen as tentative. Nonetheless these results are consistent with the study by Glazebrook et al. (2001) that found that the odds ratio for girls scoring above the threshold on the SDQ compared to the norms was 3.1 for girls, and 1.99 for boys, in a similar study of children attending paediatric out-patient clinics.

This study also showed high levels of emotional symptoms for both sexes in the eczema group. This is consistent with the study carried out by Absolon et al. (1997) on children with eczema and also with the larger study by Glazebrook et al. (2001) on children with a range of chronic illnesses. It is also consistent with the results of the meta-analysis carried out by Lavigne and Faier-Routman (1992).

It is important to note that all these results are based on the mother's report and no external verification of the child's difficulties was possible in this study. It is possible that all of these reported difficulties are due to differences in the mother's experience of her child, and her perception of the child's difficulties, rather than differences in the child's behaviour per se. However, these results do suggest that the mothers of children with eczema did consider their child to be more difficult to parent than do mothers of children with other illnesses.

Many other studies of children with a chronic illness have shown that such children are at increased risk of developing psychological difficulties compared to healthy children (Lavigne & Routman, 1992; Wallander & Varni, 1998). However, there has been considerable debate in the literature about whether it is preferable to study the effects of chronic illness in general (the noncategorical approach, Stein & Jessop, 1982) or whether each illness has its own particular demands that warrant studying each illness separately (Quittner et al., 1992). The differences that emerged in this study suggest that eczema may have effects that are different from other forms of chronic non-life threatening conditions and that these differences would be obscured if a noncategorical approach were used.

The differences may partly be explained by the nature of the group of children with eczema in this study, compared to those in the comparison group. As noted in the introduction, the majority of children with eczema are managed in the community, usually by their GP. Only a very small proportion of children with eczema are referred to dermatology services and these children are likely to be those who have severe, chronic eczema and/or whose parents are very concerned about the eczema and motivated to seek further help. They are very likely to be experiencing management problems with their child's eczema, and likely to show a high level of concern about the impact of the eczema on the children. This is also true to a certain extent for the comparison group in this study, and the comparison mothers did report similarly high levels of psychological distress in both themselves and their child. However, the children attending the general out-patient clinics did have a mixture of different physical illnesses, which may not have been so long-standing and their parents may not have experienced the same level of ongoing difficulty managing the condition as the children with eczema.

Even allowing for these factors, the question arises as to why it might be that children with eczema are experienced as more "difficult" to parent, cause a higher level of concern to their parents and the girls are rated as less prosocial than girls with other medical conditions. At least some of the explanation for this may be the nature of eczema itself. Children with eczema frequently have very disturbed sleep patterns (Stores, Burrows & Crawford, 1998) and the scratching caused by itching is in itself very difficult to manage. In addition, the "burden of care" that the parents report is considerable. Many children dislike the treatment routine of creams and bandages and this obviously adds to the parent's perception of difficulties (Lawson, Lewis-Jones, Finlay, Reid & Owens, 1998). All these factors make it highly likely that parents would report higher levels of management difficulties because these burdens are additional to normal parenting tasks.

The PSI is a widely used instrument to measure parenting stress. However, it has been criticised because of the inclusion of items which reflect general psychological difficulties rather than difficulties related to parenting and because of some inconsistency in the robustness of the three factor structure (Deater-Deckard & Scarr, 1996). For example, the "Parental Distress" scale is both a general measure of distress and a measure of distress related to parenting difficulties. This results in a substantial degree of overlap between this measure and measures such as the GHQ used in this study. It is important to be aware of this when interpreting the results of the measure, because it is not strictly speaking a measure of only parenting difficulties.

Relationship Between Severity and Psychological Adjustment

As has been found in many other studies of children with a chronic illness, there was no simple association between the severity of the child's eczema and his or her psychological functioning. The correlations between the objective measure of severity of eczema and the mothers' levels of distress, or the child's psychological functioning, were low and non-significant.

However, there was a significant relationship between the child's rating of the impact of the eczema on his or her quality of life, as measured by the CDLQI, and the child's psychological functioning. This is particularly interesting because there are no other studies of the importance of the child's own report of the impact of eczema on themselves. Unfortunately, the number of children old enough to complete the CDLQI in this study was quite small and larger studies will be needed to confirm this finding. However, symptom severity scores, such as the SASSAD that was used in this study, do not accurately reflect the impact of the condition on the individual and it is more likely that the perceived impact of the condition will be related to psychological distress, rather than symptom severity.

There was some evidence of a relationship between the severity of the child's eczema and the parenting stress experienced by his or her mother. When the severity scores were split about the median, the upper and lower halves of the scores on the PSI were significantly different. However, the correlation between the two measures was only moderate and hence non-significant for this sample size.

Both Absolon et al. (1997) and Daud et al. (1993) found a relationship between severity of eczema and the child's psychological functioning. However, this was done by dividing the groups into "severe", "moderate" or "mild" based on the symptom severity score or the amount of surface area affected by eczema. Hence it is not surprising that this has not been replicated in this study. Both of those studies and also this one, only included children from a very restricted sample of children with eczema, because all three studies were carried out in out-patient clinics. It is highly likely that if a broader range of affected children were included that some relationship between severity and psychological adjustment would emerge. This is supported by the differences in rates of psychological difficulties reported in the out-patient clinic studies compared to the rates reported in a large community sample (Meltzer et al. 2000).

Severity of eczema is difficult to measure accurately (Finlay, 1996). In order to minimise this problem, a standardised measure of eczema severity was used in this study and only two expert raters, who were not involved in the psychological assessments, were used. Nonetheless, it was not possible to measure inter-rater reliability in this study due to practical constraints and it is possible that measurement error may have contributed to the results investigating the link between severity of eczema and psychological factors.

Resolution of the Diagnosis

An adapted form of the Reaction to Diagnosis Interview was used to rate the level of "resolution" shown by each mother. Mothers coded as "unresolved" differed from those rated as "resolved" on the measures of maternal psychological distress and parenting difficulties. This interview focused on the mother's reaction to the specific stress of having a child with eczema and showed that the mothers who were able to overcome this difficulty did not experience the parenting difficulties that those who were still struggling with accepting the condition still experienced. In addition, although the differences between the two groups on the measure of child psychological symptoms were not significant, a moderate effect size (0.5) was shown for the difference in total score. It is possible therefore that a larger sample would have shown a significant difference in SDQ scores as well.

The same numbers of mothers were rated as "resolved" as "unresolved". This high number of "unresolved" mothers is perhaps rather surprising although it is consistent with results of Sheeran, Marvin and Pianta (1997) and Marvin and Pianta (1996). These other studies were carried out with younger children with cerebral palsy or epilepsy, which are known to have marked psychological impact on children. However, it was apparent during the interviews carried out for this study that there are several characteristics of eczema that make it a difficult condition to become resolved about. Firstly, all parents are told initially that their child will grow out of the condition, and this is true for the vast majority of children with eczema and intended to be reassuring for parents. However, for this particular group, it is a considerable source of frustration because of the sense that the condition is, or should be, temporary. All these families do therefore have a sense that their child may at some point be free of eczema, and hence may never accept that they do have to become resigned to the condition. This makes it harder for them to become resolved.

In addition, many families believed that there was an appropriate treatment for their child, but they had not yet managed to find this treatment. Many mothers commented on trying to find triggers for the flares in the severity of the eczema which are a common feature of the condition. The most commonly suspected causes for the flares were allergies, and yet very few parents felt that their suspicions were taken seriously by conventional medical practitioners. Many had turned to alternative practitioners and some had been given advice about management of the child's "allergy". This conflict between the conventional wisdom and alternative or lay explanations for eczema also made it harder for the mother to become resolved about her child's eczema, because of the uncertainty aroused about whether or not her child was receiving the most appropriate treatment.

Eczema is very variable and some mothers commented that if they had been interviewed at a different time, their replies would have been very different. There are no published data about the test-retest reliability of the RDI and yet it is likely that resolution may vary over time especially in a condition like eczema that is so variable. This should be addressed in further work.

There was no evidence that resolution was related to aspects of the illness itself e.g. severity, time since diagnosis, family history of eczema or history of admission for eczema. This was also demonstrated in the study by Pianta et al. (1996), which also showed that type of illness, whether cerebral palsy or epilepsy did not affect rates of resolution.

An adapted form of the RDI was used in this study, and this may make it inappropriate to generalise to other studies using the RDI. An adapted form was used because of the nature of eczema, and the relative unimportance of the actual diagnosis for most families. The emphasis in the interviews was much more on the mothers' beliefs about current difficulties and ongoing management problems and current acceptance of the condition, rather than on the shift between the traumatic period of diagnosis to acceptance of the condition, which is the usual emphasis in the RDI.

It was not possible to measure attachment status in this study. However, Marvin and Pianta (1996) demonstrated a very strong relationship between resolution and attachment status. In their study, 82 % of the mothers classified as Resolved had securely attached children, compared to only 19 % of the mothers classified as Unresolved. This is a very striking finding and needs to be replicated in another sample. The children they studied had cerebral palsy, and some were not able to move independently so an adapted form of the Strange Situation had to be designed which makes it even more important that their finding is replicated.

Daud, Garralda and David's (1993) study of attachment difficulties showed no evidence of higher rates of attachment difficulties amongst mother-child dyads where the child had eczema, compared to a healthy children. 86% of the children in the eczema group were securely attached to their mother, compared to 70% in the comparison group. However, in Marvin and Pianta's (1996) study only about half the children were rated as securely attached, which is slightly lower than the normal population. This difference is striking, and it would be interesting to assess the relationship between resolution and attachment status in children with eczema in a future study.

There was no support from this study for the hypothesis that resolution acts as a moderating variable on the relationship between severity and psychological adjustment. As noted above, no direct relationship between severity and psychological adjustment was found either. There was a lot more variation in the scores of the unresolved groups compared to the resolved groups on the psychological measures, and more variation in the severely affected compared to the mildly affected on the psychological measures. However, these results do not fit in with a moderator model.

Clinical Implications

This study was cross sectional in design and resolution cannot be interpreted as a "causal" factor in determining psychological distress from this study alone. It is extremely likely that the relationship between resolution and psychological difficulties is in fact reciprocal or bi-directional. For example, it is highly likely that resolution is influenced by general levels of distress and by parenting difficulties, rather than just being a "cause" of parenting difficulties or psychological distress. It is also possible that resolution is itself a consequence of other pre-existing differences between the two groups e.g. in relation to attachment status, and that resolution is therefore an epiphenomenon rather than a causal mechanism. Also, many people would argue that eczema symptoms may be influenced by levels of distress in both the child and parent, as well as causing such distress. It would only be possible to test these further by using a longitudinal design, such as the study by Kovacs et al. (1989), or an experimental design in order to determine whether or not resolution is a causal mechanism or simply a measure of psychological distress.

However, Marvin and Pianta (1996) do suggest that the concept of resolution may have important clinical implications. For example, it may be possible to identify mother-child dyads who are at risk of developing difficulties because of the mother's lack of resolution. If it were possible to help a mother become more resolved about her child's illness, this may reduce the parenting difficulties they experience, as well as help reduce overall levels of distress. This could be evaluated further using the RDI before and after a clinical intervention.

Studies with adults with chronic illnesses have demonstrated the importance of the person's illness representations and have demonstrated links between their illness representation and symptoms of psychological distress (Moss-Morris, Petrie & Weinman, 1996). The mother's beliefs about her child's illness probably influence how she copes with managing the illness, and hence the child's psychological adjustment, and this is a feature of both the Wallander and Varni (1998) and Thompson et al. (1994) models. However few empirical studies have studied mother's beliefs about her child's illness and there are no intervention studies which aim to intervene at the level of the mother's cognitive and emotional response to her child's illness. Eiser (1990) and Wallander and Varni (1998) concluded their reviews of studies on the impact of chronic illness in children by highlighting the need for research to study processes underlying adjustment because of the need to develop clinical interventions in this field. This would be an important area of future research.

An alternative clinical approach would be to offer clinical intervention to those mothers who express high levels of parenting stress and by offering help with parenting skills. In a longitudinal study, Goldberg et al. (1997) showed that high scores on the PSI in young childhood were important predictors of later psychological difficulties amongst children with cystic fibrosis and congenital heart disease. Reducing parenting stress would be a valuable intervention whether or not the mother's resolution changed as well.

Limitations of the Study

There are important limitations to this study and the results should be considered in the light of these.

The study was based on a very specific group of children with eczema, and the results cannot be generalised to all children with eczema. Eczema is an extremely common condition in childhood and in most cases it is self-limiting and minor. This study has confirmed that very different levels of psychological distress are found in out-patient samples, compared to those in the community.

This study used brief screening measures of psychological distress for both the children and mothers. These are no substitute for either more comprehensive assessments of psychological well being or full diagnostic interviews. The presence or absence of psychological symptoms is a very crude measure of adjustment and it is likely that more subtle difficulties within the normal range of behaviour will have been missed by using these measures. Unfortunately the range of useful measures of psychological well being is rather limited and these too have their drawbacks (Harris, Canning & Kelleher, 1996). Given the practical constraints involved in carrying out this study, it was not possible to use more time consuming interview methods to assess psychological functioning.

This study did not evaluate the impact of eczema on the whole family and concentrated instead on the impact on the mother alone. This was due to practical restrictions that made it difficult to recruit fathers or siblings to the study. The majority of children attending out-patient clinics are brought by the mother alone and the mother does provide the physical care for her child in most families. However, it is now well documented that there is a significant impact on fathers of children with a chronic illness that may be very different from the impact on mothers and this should be explored further (Mastroyannopoulou, Stallard, Lewis and Lenton, 1997; Sloper, 2000).

The comparisons used in this study were with a heterogeneous group of children attending paediatric out-patient clinics because of long term but non life-threatening conditions. This comparison group was chosen partly to control for the peculiarities of the sample due to the location of the study in South East London (high economic deprivation and multi racial community). However, it was also felt important to compare with another out-patient group to determine if there was something more specific about eczema that caused such apparently high levels of distress. The findings would not necessarily generalise to other illness groups and it would be interesting to compare the levels of parenting stress in children with other chronic illnesses. Most studies in this field have used a noncategorical approach and have argued that there are not significant differences between disease groups. A noncategorical approach may be appropriate for looking at very global measures but cannot detect the specific difficulties experienced with each illness, and may obscure important features of a particular illness.

CHAPTER THIRTEEN

DISCUSSION AND CONCLUSIONS

This thesis investigated the psychological impact on children and families of two different skin disorders, Epidermolysis Bullosa (EB) and eczema. The results of the studies show that these two disorders do have a marked impact on children and their families. However, there was a lot of variation in the psychological response to having a child with a skin disorder. The thesis examined three factors that may account for the variation in psychological adjustment: the severity of the illness, family functioning and the mother's resolution of the diagnosis. The results provide some support for the hypothesis that family functioning and resolution are correlated with psychological outcome, but the findings for severity were more mixed. Each of these three factors will be reviewed in turn and the implications for the theoretical models developed by Wallander and Varni (1998) and Thompson et al. (1994) will be considered.

In addition, the qualitative study of the experiences of mothers of children with EB described four themes that were important for understanding the mother's adjustment to caring for her child with EB. The findings from the qualitative study complemented the findings from the quantitative study, by providing a more detailed exploration of the adjustment process. This study illustrated the individual variation in the psychological response to having a child with a severe skin disorder, and although the results are limited because they are based on only one interview, they provide some information about psychological processes that were important for these mothers.

The study of the psychological impact of EB did show an association between family functioning and psychological adjustment in the child. Family cohesion was positively related to psychological adjustment in the child whereas conflict was negatively associated with child adjustment for the mother's ratings. These findings are consistent with many other similar studies (Drotar, 1997; Thompson et al., 1999). This direct association between family adjustment and child psychological adjustment is consistent with both of the models by Wallander and Varni (1998) and Thompson et al. (1994). This association has now been shown across several different disease groups and yet there are few studies that are prospective in design that make it possible to determine the direction of cause and effect. This finding could potentially be clinically useful because it may be possible to offer an intervention to address difficulties in family functioning and hence improve psychological adjustment in the child. As Thompson et al. (1999) conclude:

> "Experimental level research is needed to confirm theories and correlational evidence of the role of family functioning in adaptation to chronic illness" (p. 497).

Therefore, there is now clear evidence from many studies that family functioning is associated with psychological adjustment to chronic illness in childhood, and further research is needed to clarify the direction of causality and whether this association has useful clinical implications.

The evidence from this thesis linking the severity of the skin disorder to psychological adjustment was more mixed. In the study of the psychological impact of EB, there was no evidence of a relationship between the severity of the child's EB and child adjustment, or maternal distress. However, there was a relationship between severity and paternal distress. In the study of the psychological impact of eczema, there was no evidence of a relationship between severity as measured by an expert rater and child psychological adjustment or maternal distress. However, there was a small to moderate relationship between severity of eczema and parenting stress. In addition, the child's rating of the impact of their eczema on their quality of life was strongly associated with child psychological adjustment. Finally, there was no evidence that resolution acted as a moderating variable on the relationship between severity and adjustment. These rather mixed results are also found throughout the literature and it is still not clear how much influence severity has on psychological adjustment to an illness (Hommeyer, Holmbeck, Wills & Coers, 1999).

The two main theoretical models propose different pathways for the impact of severity of illness on psychological adjustment. In the Wallander and Varni (1998) model, there is both a direct link between severity and adjustment and an indirect link, via functional independence and psychosocial stressors. In the Thompson et al. (1994) model, severity is indirectly related to adjustment via maternal adaptational processes. However, Thompson et al.'s (1998) description of the type of relationship between the maternal adaptational relationships and psychological outcome is ambiguous: "maternal adaptational processes are hypothesized to mediate/moderate the illness-adjustment relationship" (p.122).

The studies in this thesis provide only a partial test of either of the above models, but the results do not clearly support either one of the models. In fact, the results suggest that there may be gender differences between mothers' and fathers' responses to the severity of the child's illness. In addition, severity of illness can be measured in many different ways. Even within this thesis, several different types of measures of severity were used: objective severity as rated by an expert rater, a parental report of severity, functional disability of the child as rated by a parent and the child's own report of the impact on their quality of life. The use of different types of measures of severity may account for some of the differences in findings. Therefore, there is no clear answer as to whether the severity of illness is an important predictor of psychological adjustment to chronic illness at present. Future research in this area needs to take into account the differences between the various types of measures of severity and to specify much more clearly the hypothesised link(s) between severity and adjustment (Hommeyer et al., 1999).

The results from the study on the psychological impact of eczema suggest that the concept of resolution is a helpful one in describing mother's attitudes towards her child's illness. The qualitative study of the experiences of mothers of children with EB identified resolution as a key theme, and the study of the psychological impact of eczema found that there were significant differences in psychological symptoms in the resolved group compared to the unresolved group. However, considerable work still needs to be done in establishing the credibility of this concept. Firstly, the relative importance of the concept of resolution as distinct from general psychological distress in predicting psychological adjustment has to be examined, because there is some overlap between the concepts of resolution and general psychological distress. In addition, the test-retest reliability of the Reaction to Diagnosis Interview (Pianta & Marvin, 1992) needs to be established.

The concept of resolution appears to reflect the mother's beliefs about the significance of her child's illness and the emotional impact of the illness on herself and her child. It is intended as a specific measure of the impact of illness rather than a generic measure of psychological distress, although as noted above further research needs to be done to assess to what extent these concepts can be separated. As such, it is consistent with Thompson et al.'s (1994) model that identifies the importance of "maternal adaptational processes". However, whereas Thompson et al. (1994) use the Lazarus and Folkman (1984) model of coping as the theoretical framework underlying maternal adaptational processes, the concept of resolution was developed from a different theoretical basis, namely attachment theory.

Lazarus and Folkman's (1984) theory of coping has been the main theoretical framework that has influenced research on the impact of chronic illness on children, as well as with adults. As discussed in Chapter One, this theory is central to both the Wallander and Varni (1998) and the Thompson et al. (1994) models of the impact of chronic illness on children and families. Thompson et al. (1999) stress the importance of identifying cognitive processes that underlie adaptational processes because they offer possibilities for intervention, whereas it is harder to intervene at the level of illness severity or demographic parameters. As a consequence, several of their studies have focused on cognitive processes, such as stress appraisal, health locus of control and expectations of efficacy, that predict adjustment for both children and mothers.

The coping model has been criticised however, both because of the conceptual limitations of the model and because of the gulf between theoretical research and the clinical application of this research (de Ridder, 1997). Coyne and Racioppo (2000) go so far as to say,

> "This literature is in crisis because of its failure to yield substantive findings concerning the role of coping in adaptation that cannot be dismissed as truisms, trivia, or the product of a confounding of stress, coping and distress." (p 655)

They argue that on a theoretical level, the definition of coping is too narrow. Coping is defined as the reaction to a stressful situation and is assumed to be directed at minimising distress, but other factors that may also influence the coping response, such as an individual's goals or personal commitments, are not included in the model. As Bradford (1997) argues, it would be an oversimplification to expect that adjustment would be "solely the function of an individual's stress processing abilities" (p 167). In addition, there is still no consensus on taxonomies of coping styles and as a consequence different measures use different numbers and types of dimensions, which makes it hard to compare between studies. Coyne and Racioppo (2000) also criticise the model because of the circularity of many findings that confound cause and effect:

> "The issue of temporal and causal priority can be seen in the robust finding that emotion-focused coping is positively related to distress, perhaps the most consistent finding in the coping literature. Undoubtedly, much of this reported emotion-focused coping follows upon, rather than contributes to, psychological distress" (p 657).

In response to these criticisms, Lazarus (2000) argues that the theoretical model has become more sophisticated over time. For example, Folkman and Moskowitz (2000) stress the importance of including "appraised situational meaning" (i.e. the evaluation of the personal significance of a stressful situation) as an influence on coping processes. More recent studies have addressed some of the concerns expressed by Lazarus (1993) about the misapplication of the model. For example, some studies have used prospective designs and within-person, process-oriented methods rather than relying on trait measures based on group differences (Tennen et al., 2000). In addition, qualitative methodology has been used in some studies to overcome the criticism that coping processes have only been measured in a superficial way (Folkman and Moskowitz, 2000). Finally, the gulf between theoretical research and the application of such research to clinical interventions exists in most areas of psychological research and cannot be seen as specific to the coping model (Lazarus, 2000).

This criticism does however raise the question of whether the coping model is the most theoretically valid model to explain individual variation in response to a child's illness. Other models of the response to illness have suggested that beliefs about the illness, or the meaning of the illness for an individual, might help explain individual differences in response to illness, and that it is these beliefs that might themselves determine the coping response used.

One such model is the self regulatory model of illness behaviour which emphasises the role of the individual's representations of their illness both at a cognitive and an emotional level (Leventhal & Nerenz, 1985; Leventhal et al., 1997). These representations are based on previous experiences or beliefs about an illness and the threat it poses to that individual. The processes are said to be "self-regulatory" because the different processes operate in order to maintain the status quo. Weinman and Petrie (1997) claim that:

> "Whereas most current research has focused on coping as a way of explaining illness adjustment and outcome, we believe that illness perceptions may not only explain the variety of coping responses to the same illness but also be more directly related to such outcomes as adherence, emotional distress, and illness-related disability" (p 114).

Weinman et al. (1996) developed a questionnaire to assess illness perceptions based on Leventhal and Nerenz (1985) self regulatory model of illness beliefs. This questionnaire assesses beliefs about illness along five different dimensions: identity, perceived cause, timeline, consequences and curability. Illness perceptions, as measured by this questionnaire, were significantly related to psychological adjustment in Moss-Morris et al.'s (1996) study of adults with chronic fatigue syndrome and in Heijmans (1997) study of patients with Addison's disease. More importantly, these studies all found that illness representations were better predictors of outcome than coping measures.

Whilst this model is now being researched within the field of health psychology, the research has so far focused on adults with chronic illness. However, both the main theoretical models within the paediatric psychology literature emphasise the importance of the mother's beliefs about her child's illness and hence this model might be applicable to understanding adjustment to chronic illness in a child. There is some evidence that the five dimensions identified by Leventhal and Nerenz. (1985) are also found in children's representations of illness (Goldman, Whitney-Saltiel, Granger & Rodin, 1991). Future research should take into account the criticisms of the coping model and consider alternative theoretical frameworks, such as the Weinman and Petrie (1997) model of illness representations, or the Pianta et al. (1996) model. Whilst the coping model developed by Lazarus and Folkman (1984) was originally adopted with enthusiasm, it has not been useful in predicting psychological response to illness as originally hoped, and this model needs to be reviewed alongside other theoretical models.

Future Research Areas

There are some aspects of the impact of skin disorders on children and families which were not covered in this thesis but which are of particular importance for future research. These will be discussed in turn below.

This thesis did not examine peer relationships and social functioning in detail, yet these are likely to be affected by skin conditions. Jowett and Ryan's (1985) study on the impact of skin disorders on adults demonstrated the importance of social factors and difficulties in social relationships experienced by adults with skin disorders. Social context is very important in childhood too, and it is likely that the skin disorders included in these studies had some impact on social relationships. Many of the children and mothers did talk about difficulties in relationships with other children, mostly because of episodes of teasing or bullying, and other people's fear of contamination. It is quite difficult to measure peer relationships adequately using self report questionnaires (Cramer, 2000) and it is a sensitive subject to interview children about, particularly when the child does have concerns about his or her social functioning or appearance. However, by adapting the interview context to make it more appropriate to the child's developmental stage, interview methods can provide reliable information (Ross & Ross, 1984). This is an area that should be addressed in future work because if is likely to be particularly relevant for children with skin disorders.

The studies in this thesis were cross sectional in design and hence cannot reflect the dynamic nature of the processes that are of interest. Prospective studies would enable changes over time to be measured, and although these studies are very time consuming and difficult to carry out, this has now been done for some disorders, such as cystic fibrosis and sickle cell disease (Thompson et al., 1999). More prospective studies are needed to understand the psychological impact of skin disorders on children and families.

Future research should take a broad view of the psychological impact of skin conditions and should take into account the methodological difficulties identified here. This thesis has focused mostly on mothers but research should ideally include the impact on siblings and fathers, as well as mothers (Seagull, 2000). As well as measuring psychological distress or adjustment, measures of social functioning and peer relationships should be included. In addition, multiple informants should be used, including the affected child, him or herself and his or her school.

The impact of the skin disorder on the mother-child relationship was not assessed specifically in these studies. However, there are several reasons why skin disorders would be expected to have an impact on the mother-child relationship (Howlett, 1998; Koblenzer, 1990). Firstly, skin disorders affect the appearance of the baby or child, and this may have both direct and indirect effects on the mother. Some skin disorders, such as severe EB, are very disfiguring and for some mothers this will affect their reaction to their child. In addition, all mothers of children who appear different also have to cope with the reactions of other people to her child and this can lead to a heightened sense of protectiveness towards the child. Both the skin disorders in this thesis also caused a considerable amount of discomfort for the child. This too, will have an impact on the mother-child relationship because the mother has the difficult task of trying to offer comfort to an irritable and uncomfortable child. In addition, both EB and eczema are genetic conditions and the way in which these conditions are inherited may also have some impact on the mother-child relationship. Finally, the substantial burden of care placed on many of these mothers is an additional source of both physical and emotional stress.

Research evidence so far suggests that there is no difference in attachment status between mother-child dyads of children with eczema, or other types of physical illness compared to controls, (Daud et al., 1993; van IJzendoorn et al., 1992). However, there are many other ways in which a skin disorder may influence the mother-child relationship which have not yet been studied. For example, Hodes, Garralda, Rose and Schwartz (1999) demonstrated higher levels of EE (Vaughn & Leff, 1976) amongst mother-child dyads with a child with epilepsy compared to healthy sibling controls. In particular, they found higher levels of "emotional overinvolvement" and hostility in the mothers of children with epilepsy. Although it is possible that higher levels of emotional involvement are appropriate in this group because of the demands of the illness, this study did demonstrate a significant difference in the relationship between a mother with a healthy child compared to a child with a chronic illness, which may have important implications for psychological adjustment.

Many of the descriptions of the impact of skin disorders on the mother-child relationship have been very critical of the mother's role, largely because of the assumption by many writers that the child's skin condition is to some extent caused by the difficulties in this relationship (Van Moffaert, 1992). Too little emphasis has been placed in the past on understanding the difficulties experienced by mothers and fathers, and their efforts to overcome these. The focus in future research should be on trying to understand the difficulties experienced by parents in order to improve support to children with skin disorders, both on a very practical level and on an emotional level. Qualitative methodology has been shown to be very useful in providing not only more detailed and in-depth descriptions of psychological processes, but also in allowing participants to have some influence on what processes are seen as important.

As has been identified in many previous reviews of this field (Eiser, 1990; Pless & Nolan, 1991) there are very few studies of clinical interventions based on the descriptive studies of the impact of chronic illness on children. At present there is no consensus about whether it is possible to identify families who are the ones at risk of developing psychological difficulties and offer them preventative interventions. Only very specific, problem focused interventions have been shown to be effective (Kibby, Tyc & Mulhern, 1998). More research is needed on the most effective ways of providing psychological support to children and families affected by skin disorders.

In summary, there is still a considerable amount that needs to be done to improve our understanding of the psychological impact of skin disorders on children and their families. Many studies in this field rely on methodology that cannot capture the complexities of the psychological impact of a chronic illness in childhood. In particular, the reliance on self-report measures and cross sectional studies limit the results to findings which may accurately reflect groups differences but which do not increase our understanding of the psychological processes involved in adjustment. As a consequence, the results from these studies do not have clinical relevance, and theoretical developments have not resulted in interventions that might either prevent or reduce the psychological consequences of chronic illness in childhood. Some recent studies have tried to address these difficulties, for example, by using qualitative methodology and more sophisticated, interview based measures in order to study psychological processes, but many of the methodological difficulties identified in successive reviews of the field have not been addressed. In addition, there are very few good quality studies of the impact of skin disorders. Therefore, there is considerable scope for future research in this area to improve our understanding of the psychological impact of skin disorders on children and their families.

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LIST OF APPENDICES

APPENDIX 1

COPING WITH EPIDERMOLYSIS BULLOSA – RESEARCH PROJECT

Information Sheet

We are currently carrying out a project on behalf of DEBRA (Dystrophic Epidermolysis Bullosa Research Association).

The project aims to look at the ways in which families and children adjust to EB. We hope that this research will help us identify ways in which children and families cope with the illness, and ways in which further support could be provided. Ultimately, we hope this may enable us to prevent serious problems occurring by identifying them at an early stage.

If you decide to participate in the project this will involve filling in some questionnaires and being interviewed by the researcher. In addition, your child will be interviewed by the researcher. If you consent to the project at this stage you can decide to withdraw at any time in the future. The interview can be arranged at a time to suit you, in your own home.

The information you give us will be completely confidential. The project will be entirely separate from you regular medical treatment and you decision about whether or not to take part will not affect your medical treatment in any way.

APPENDIX 1

COPING WITH EPIDERMOLYSIS BULLOSA – RESEARCH PROJECT

Consent Form

I/we agree to participate in the research, as outlined in the information sheet.

I/we understand that this eill require the completion of questionnaires and an interview with myself/ourselves and an interview with our child named below.

I/we have the right to withdraw form the research at any point.

Name of Child:..

Signed:....................................	Signed:...................................
Print Name:..............................	Print Name:.............................
Relationship to Child:..................	Relationship to Child:................

APPENDIX 2

COPING WITH EPIDERMOLYSIS BULLOSA

CHILD INTERVIEW

INTRODUCTION:

I want to talk to you to find out what it's like to have the sort of illness that you have. You're the one who knows the most about this, because only you know what it is really like for you having this illness.
I'd like to know what you would say to someone who wanted to know about your illness to help them understand. I have some questions here that this person would want to ask you, but you can also add anything of your own that you think is important.
This is not like a test and there are no right or wrong answers to these questions. What I'd like is if you could tell me how you would explain your illness to someone.

APPENDIX 2

SECTION ONE : KNOWLEDGE

1 If someone wanted to know what the full **name** of your illness was, what would you tell them ?

If they wanted to know whether there were different **types** of this illness, what would you tell them? If they asked whether you have a particular type, what would you say?

(PROBE : What type of Epidermolysis Bullosa / EB do you have ?)

2 Now suppose the other person wanted to know how this illness **affects** you. What parts of your body would you tell them it affects ?

Are there any other parts of the body you would tell them it affects?

(PROBE : Does it affect your skin ? What about inside your body?

3 If the other person wanted to know in what way the illness affected your skin, what would you tell them? **What does EB do to your skin** ?

How would you try and explain what it is that is wrong with the skin that **makes it more fragile** ?

4 What would you want to tell the other person about the **treatment or care** of your skin? What would you tell them is the best way of looking after your skin?

What would you tell them is the best way of making your skin feel better? And are there things that make it feel worse?

Would you tell them you have to do anything else to take care of yourself? (PROBE : Do you have to eat special foods ? Or take any type of medicine or tablets for it ?)

5 Now suppose the person wanted to know **how** it is some people get EB. What would you tell them about that ?
How would you explain to them how someone gets EB?

6 Is there anything else that you would want to say about EB that is important ?

APPENDIX 2

SECTION TWO : DESCRIPTION OF ILLNESS

1 I expect the other person would also want to know what it feels like to have EB. It's very hard for someone else to understand how an illness feels. Its also hard for you to know what its like not to have EB since you've had it all your life. How would you try and describe to them what your body feels like if you have EB ?

Prompt: Would you tell them it hurts ?
Would you tell them it itches ?

2 Would you tell them it affects the sorts of things you can do compared to children who don't have EB? What sort of things does it affect ?

Prompts: Would you tell them it affects joining in games at school?
Would you tell them it affects the sorts of food you can eat?
Would you tell them it affects making friends?

3 Now suppose the other person wanted to know some of the bad things about having EB and how this makes you feel inside. (By this I mean the sort of feelings you have like being happy or sad and not the feelings in your body that we just talked about.) Can you tell me some of the bad things about having EB and how that makes you feel ? (Probe to get full list and prompt for feelings associated)

You've told me several things that are bad about having EB. If the other person asked you what the worst thing was about having EB out of the things you've told me about, what would you tell them it was? (When that happens) How does that make you feel?

What would you tell them was the next worse thing ?
(When that happens) How does that make you feel ?

And the next worse ?
(When that happens) How does that make you feel ?

4 If the other person asked you it there were any good things about having EB what would you tell them?

APPENDIX 2

SECTION THREE : STRATEGIES FOR COPING WITH DIFFICULTIES

Now I want to ask you about some difficult situations to get some ideas about what sort of things you find helpful. Again there are no right or wrong answers to these questions and this is not a test. I am interested in what you think **you** would find helpful.

1 Suppose you had a bad tummy ache. Do you think there is anything you could do to make yourself feel better ?
What could you do to make yourself feel better?
How well would that work ? - would it make you feel any better?
Why do you think that works ?

2 Now suppose you were expecting someone you liked very much to come and visit you and they just telephoned you to say they were not able to come so you feel disappointed they can't come.
Do you think there is anything you could do to make yourself feel better and feel less disappointed?
What could you do to make yourself feel less disappointed?
How well would that work - would it make you feel any better?
Why do you think that works ?

3 Now suppose you were being teased by one of the older girls/boys at school. So you were feeling upset at being teased.
Do you think there is anything you could do to make yourself feel better and feel less upset?
What could she do to make yourself feel less upset?
How well would that work - would it make you feel any better?
Why do you think that works ?

APPENDIX 2

SECTION FOUR : STRATEGIES FOR COPING WITH EB

1. Earlier you told me the worst thing for you about having EB was .....
You said that when that happens to you, it makes you feel ....

Do you think there is anything you can do to help yourself feel better about that ?

What could you do ?
Does that work - how well?
Does it make you feel any better ?
Why do you think it does/ doesn't work?

2 You told me the second worst thing for you about having EB was .....
You said that when that happens to you, it makes you feel ....

Do you think there is anything you can do to help yourself feel better about that ?
What could you do ?
Does that work - how well?
Does it make you feel any better?
Why do you think it does/ doesn't work?

3 You told me the next worst thing for you about having EB was .....
You said that when that happens to you, it makes you feel ....

Do you think there is anything you can do to help yourself feel better about that ?
What could you do ?
Does that work - how well?
Does it make you feel any better?
Why do you think it does/ doesn't work?

APPENDIX 3

CODING INSTRUCTIONS

For each situation, read the child's response and code the **main** coping strategy according to the codes on the following sheet. If more than one strategy is described, code the main strategy and then indicate any further strategies in brackets after.

A list of examples for each category is given to provide more information about each type of strategy.

APPENDIX 3

CODING CATEGORIES

AA	Aggressive Activities	Verbal or motor activities that may be hurtful
BA	Behavioural Avoidance	Behaviour other than isolating that is a deliberate attempt to keep oneself away from a stressor
BD	Behavioural Distraction	Behaviour other than isolating or avoidant that delays or reduces the need to deal with a stressor
CA	Cognitive Avoidance	Deliberate cognitive attempts to avoid acknowledging the existence or a stressor
CD	Cognitive Distraction	Deliberate cognitive attempts to keep thoughts away from a stressor
CPS	Cognitive Problem Solving	Thoughts focused on ways to modify, prevent or eliminate the stressor
CR	Cognitive Restructuring	Thoughts that alter one's perceptions of the characteristics of the stressor
EE	Emotional Expression	Behaviour other than aggressive that expresses feelings or emotions
E	Endurance	Behaviour that causes one to face the stressor and accept its consequences
IS	Information Seeking	Behaviour that involves obtaining information about the stressor
IA	Isolating Activities	Behaviour that serves to separate the individual from the presence of others
SCA	Self Controlling Activities	Behaviour or cognitions that serve to reduce the tension or control one's behaviour or emotions
SS	Social Support	Non-aggressive behaviour that involves seeking the presence of an individual
SpS	Spiritual Support	Behaviour that suggests an appeal to a higher being
SM	Stressor Modification	Non-cognitive behaviour that eliminates the stressor or modifies the stressor

APPENDIX 3

		EXAMPLES
AA	Aggressive Activities	Aggression, heavy handed persuasion, physical Attack, destroy, verbal abuse, yell, argue
BA	Behavioural Avoidance	Escape, avoidance, go somewhere else, sleep, leave the situation, change the topic, try to get out
BD	Behavioural Distraction	Do something else, watch TV, play, play music, strenuous activity, group activity
CA	Cognitive Avoidance	Deny situation exists, don't think about it, ignore it, forget about it, thought stopping
CD	Cognitive Distraction	Diversionary thinking, visual distraction, think about something else, read, fantasy, humour
CPS	Cognitive Problem Solving	Focus on situation, analyze, learn, reason, decision making, processing information, stress recognition
CR	Cognitive Restructuring	Positive restructuring, emphasize the positive, wishful thinking, tell self it's ok, convince self, defensive reappraisal
EE	Emotional Expression	Cry, express feelings, act out feelings
E	Endurance	Expose self to fear, peaceful acquiescence, submit, endure, comply
IS	Information Seeking	Questioning, clarify reasons, media, explore, investigate
IA	Isolating Activities	Time out, go to special place, exclusion, isolate self, solitary
SCA	Self Controlling Activity	Self-soothing activities, think about relaxing, Eating, relax, behaviour or emotion regulating cognitions
SS	Social Support	Physical contact, talk to parents, talk to teacher, talk to peers, seek support
SpS	Spiritual Support	Pray, Spiritual support
SM	Stressor Modification	Alter situation, change physical characteristics of stressor, pain relief, propose a compromise

APPENDIX 4

SEVERITY OF ILLNESS QUESTIONNAIRE

This questionnaire asks you to rate the severity of the child's EB in four ways: extent of body affected, impact on mobility, visibility of EB and impact on lifestyle.
Please indicate which statement best describes the child by circling the letter of the appropriate statement.

1. Overall : When you consider your child's EB, how severely affected physically do you think s/he is? (Please circle one letter)

a. Very mildly
b. Mildly
c. Moderately
d. Severely
e. Very Severely

2. Extent of EB: Please indicate which areas of your child's body are affected by EB. Please use a number from the following scale to indicate how much each body part listed is affected.

KEY:
1 - Not affected
2 - Very mildly affected
3 - Mildly affected
4 - Moderately affected
5 - Severely affected
6 - Very severely affected

Hands ____
Arms (including elbows) ____
Face and Neck ____
Chest and Back ____
Buttocks ____
Legs (including knees) ____
Feet ____
Mouth ____
Oesophagus/gut ____
Bowels ____

APPENDIX 4

3. How much impact does your child's EB have on his/her mobility ? (Please circle one letter)

a. None or Very minor. No restriction on mobility or negligible effect.
b. Minor. Restriction only on boisterous activity or activity that is unusual.
c. Moderate. Some restrictions on usual activities including games.
d. Severe. Many activities restricted.
e. Very severe. Little independent mobility or very restricted eg walks less than 100 metres.

4. How visible is your child's EB ? (Please circle one letter)

a. No affected areas visible in normal clothing.
b. Visible only when partially clothed eg on knees and elbows only.
c. Visible but mild. Small areas of hands and/or face affected or affected very mildly.
d. Face and/or hands markedly affected.
e. Hands and/or face very severely affected.

5. When you compare your child with a healthy child of a similar age, what impact does his/her EB have on her lifestyle ? (Please circle one letter)

a. No impact. Usual developmental process and tasks can be accomplished.
b. Slight impact. Little effect on normal age appropriate activities.
c. Moderate impact. Limits some age appropriate activities.
d. Severe impact. Unable to do many age appropriate activities and/or significant developmental delay.
e. Very severe impact. Unable to join most age appropriate activities and/or very significant impact on developmental process.

APPENDIX 5

SCHOOL QUESTIONNAIRE

Dear ,

I am carrying out some research on how families and children cope with having Epidermolysis Bullosa. As part of this research, I have interviewed and family, and I am contacting you with their consent.

This study is being funded by the Dystrophic Epidermolysis Bullosa Research Association (DEBRA) and we hope that the results of the study will enable us to provide better services and care for children and families with Epidermolysis Bullosa. I am writing to you because I am also interested in finding out how copes at school. I can assure you that the information you give me will be confidential. I appreciate that you have many pressures on your time already, but I hope you can find the time to complete the enclosed questionnaires and return them to me in the envelope provided. They should only take about 15-20 minutes to complete.

If you have any queries about the study, please feel free to contact me on the above number and I will be happy to discuss them with you.

I look forward to hearing from you in due course.

Yours sincerely,

Penny Titman BA MSc
Lecturer in Psychology

APPENDIX 5

COPING WITH EPIDERMOLYSIS BULLOSA
SCHOOL QUESTIONNAIRE

Name of child:

Name of teacher completing questionnaire:

Number of years you have known child:

Role with respect to child:
Class Teacher
Year Head
Head/Deputy Head
Other (Please Specify)............

Type of School:
State Primary
State Secondary
Independent
Special School (Please specify).......

Is the child in a special class ? Yes / No
If yes, please give further information:

Is the child in the appropriate school year for his/her age ? Yes / No
If not, please specify age group of class:

Does the child have extra help (individual or small group) with schooling ? Yes / No
If yes, please specify what sort of extra help the child receives:

In comparison to other children of his/her age, how well does this child function in the following areas? (Please circle the number that best applies.)

	Very Poorly		Average		Very Well
Achievement	1	2	3	4	5
Self care	1	2	3	4	5
Sports	1	2	3	4	5
Age appropriate independence	1	2	3	4	5
Relationships with teachers	1	2	3	4	5
Relationships with friends	1	2	3	4	5
Ability to work alone	1	2	3	4	5

In comparison with the rest of the class, how well do you feel the child copes with school life ?

Very Poorly		Average		Very Well
1	2	3	4	5

APPENDIX 5

School Absence

How many days in the last full term did the child miss ?

Please list the reasons given for absences:

Restrictions on the child due to Epidermolysis Bullosa

What sorts of restrictions (if any) are placed on the child because of her/his Epidermolysis Bullosa?
(For example in PE or games, going on school trips or journeys, joining in at playtime)

Information about Epidermolysis Bullosa

How much information were you given about Epidermolysis Bullosa when the child came into the class ?

Please list the sources of this information (eg child's parents, school nurse or doctor, leaflets from the Epidermolysis Bullosa Research Association):

How satisfied were you with the amount of information you were given about the condition?
(Please circle one)

Very satisfied / Satisfied / Neutral / Dissatisfied / Very dissatisfied

If applicable, what sort of information would you have liked or would you like now about Epidermolysis Bullosa ?

Please add any points you would like to make overleaf.

APPENDIX 6

COPING WITH EPIDERMOLYSIS BULLOSA

PARENT INTERVIEW

As you know, this research is funded by DEBRA and the aim of the research is to look at the way in which families and children cope with having EB. We hope that this research will help to identify the sorts of difficulties that parents and children with EB experience and the sorts of support or help that they find most useful.

Obviously these are some of the areas that can be most difficult to talk about to other people. I hope that I can help reduce some of those difficulties by reassuring you that what you tell me will be treated confidentially. Although I am going to ask for details of the family, I will make sure that when I produce the report on the findings of the research, no individual child or family will be identified. I will report on the findings in a general way, but without giving personal details such as names.

I also want to remind you that you are free to not answer any questions you find difficult or to stop the interview at any time if you decide you don't want to go any further.

The interview starts with some questions about the type of care you give your child. I will then ask you about your reaction to coping with EB, the sorts of things you have found difficult and the sorts of support you have found helpful.

APPENDIX 6

1. GENERAL

Could we start with some basic information about the family. Can you tell me who's in your family ? (Draw tree)

Brief details of each family member: age, working/looking after children, any significant factors, presence of EB. If appropriate, details of separation/divorce and custody arrangements

Can you tell me a bit about ---'s EB. For example what type of EB does s/he have?

Does anyone else in the family have EB ? What type ?

2. MANAGEMENT OF ILLNESS AND CARE GIVING

I would like to ask you about managing ----'s EB and whatever you have to do to manage the illness. What I am particularly interested in is how much extra care you have to give compared to another child of the same age. Obviously it is difficult to know exactly but I'd like to know what your impression is of how much extra you have to give.
I would also like to know who helps with this, if anyone, and roughly how long you have to spend on this management.

Management of Illness

Bursting or treating blisters

Applying dressings or creams

Physiotherapy or Exercises

Visiting clinics or doctor

Other:

APPENDIX 6

Caregiving

Now I'd like to ask you about the sort of care that you have to give --- because of her/his EB. I will ask you about the sort of care that has to be provided for all children, depending on their age, but what I am particularly interested in is if you can estimate how much extra care you have to give compared to another child of about the same age.

Feeding

Washing and keeping self clean

Dressing

Toiletting

APPENDIX 6

3. IMMEDIATE IMPACT OF EB

Reaction
Thinking back to the time when ----- was born.
Can you tell me about when ---- was born and how you found out he/she had EB ?
How would you describe your reaction when ---- was born ?
Probe for more information.

Sources of support
What sort of things did you find helped you to cope when ---- was still a baby?
What or who was most helpful to you then ?
In what ways?

Information
Did you know anything about EB before --- was born?
How you get information about the condition ?

Retrospective
Looking back on that experience now, what would you feel would be most helpful to a mother or father who have had a child with EB ?

Prompts: Information
    Support from people who know about EB - other families
    specialist nurses
    General support - counsellor
    Support for own family
    DEBRA

(Depending on position of child in family and EB in family):
Did ---'s EB influence your decision about having/not having another child?
How did you come to that decision?
What sort of advice or help did you get?
Would you have liked further advice or help ?

APPENDIX 6

4. CURRENT IMPACT OF EB

Difficulties
You've told me about some of the care that you have to provide for --- now. I'd like to ask you a bit more about some of the difficulties you encounter now and how you cope with them.
What are the most difficult things for you personally about coping with ---'s EB at the moment?
(Prompt for up to three other difficulties)

Now thinking about the more long term of helping your child grow up with EB. Are there times when you are thinking ahead and you feel that it is a struggle?
Or were there times in the past when you felt like that ?
When you find it difficult to cope with things in this way, what sort of things do you find helpful to enable you to cope ?

Supports
What sort of things do you find most helpful as support now ?

Prompts: Information
Support from people who know about EB - other families
specialist nurses
General support - counsellor
Support from own family
DEBRA
Having other things in life (eg work, social activities friends)

Help seeking
Have you ever felt the need for help for youself/ves eg counselling to help with difficulties or feeling low?
Have you ever felt that you would like to get help but didn't know who to go to for help or didn't want to ask ?

Ideal resources
In an ideal world, what sort of resources would you find helpful personally?

Personal gains
Are there things that you feel you have gained personally through having a child with EB?

APPENDIX 6

5. GENERAL PROBLEMS

Thinking about ---- in general rather than just focusing on her/his EB, are there things that you find particularly difficult about his or her behaviour ? These are some common problems that any child may have at times.

Temper tantrums
Moods/depression: feeling miserable, unhappy, tearful
Eating problems
Sleeping problems
Toiletting
Helping around the house with household chores
Doing things he/she is told to do
Difficulties getting on with others: shyness, isolation
Difficulties at school: concentration, achievement, bullying, refusing to go
Irritability
Worries
Separation problems
Somatic symptoms: headaches, stomach aches
Anitsocial behaviour: stealing, lying
Restlessness or hyperactivity
Habits eg thumb sucking, nail biting, tics

(Get descriptions of each thing including what the family do to try and improve the situation, especially how the parents manage this between them)

Help seeking
Have you ever tried to get help with any of these things?
Have you ever felt you'd like to get help but didn't know who to go to for help or didn't want to ask ?

APPENDIX 6

6. FAMILY LIFE

Family activities
What sorts of things does .... do outside school hours ? eg guides, cubs, swimming
Would you like ... to be able to do more, less or do you feel s/he does about the right amount?

What sorts of things do you as a family do eg on the weekends or in the evening ?

Marital relationship
Now thinking just about you two as a couple. In what ways (if any) do you feel that having a child with EB has effected your relationship?

(If no partner) Now thinking about your relationship with .....s mother/ father. In what ways if any do you feel that having a child with EB affected that relationship?
And what about subsequent relationships - has it affected those in any way ?

APPENDIX 7

INFORMATION SHEET

CARING FOR A CHILD WITH ECZEMA

We are currently carrying out a study to look at the impact of caring for children with eczema. If you agree to take part in this study, this will involve a short interview about your experience of caring for you child and managing his or her eczema, and completing questionnaires about your child and yourself. This will probably take approximately 20-30 minutes and we can arrange to do this in the clinic or at home at a time to suit you.

All information you tell us will not be disclosed to anyone else. Your child's treatment will not be affected by whether you decide to take part in this study. In the longer term, we hope that this study will help to offer more effective services to families caring for a child with chronic eczema.

If you do have any queries, please do ask for more details.

Penny Titman
Clinical Psychologist

APPENDIX 7

CONSENT FORM

CARING FOR A CHILD WITH ECZEMA

I consent to take part in the research as described in the Information Sheet I have been given.

Signed:

Please Print Name:

Name of Child:

Date:

APPENDIX 8

INFORMATION SHEET

CARING FOR A CHILD WITH A CHRONIC ILLNESS

We are currently carrying out a study to look at the impact of caring for children with a chronic illness. If you agree to take part in this study, this will involve completing questionnaires about your child and yourself. This will probably take approximately 10 - 15 minutes and can be done while you are waiting in the clinic.

All information you tell us will not be disclosed to anyone else. Your child's treatment will not be affected by whether you decide to take part in this study. In the longer term, we hope that this study will help to offer more effective services to families caring for a child with chronic illnesses.

If you do have any queries, please do ask for more details.

Penny Titman
Clinical Psychologist

APPENDIX 8

CONSENT FORM

CARING FOR A CHILD WITH A CHRONIC ILLNESS

I consent to take part in the research as described in the Information Sheet I have been given.

Signed:

Please Print Name:

Name of Child:

Date:

APPENDIX 9

THE
LEWISHAM HOSPITAL
NHS TRUST

Leading Health Care

Lewisham Hospital, Lewisham High Street, London SE13 6LH

Your Reference CNM/CT
Our Reference

Telephone: 0181 - 333 3000
Ext:
Fax:

THE LEWISHAM HOSPITAL NHS TRUST
RESEARCH ETHICS COMMITTEE

2nd April 1997

Penny Titman
Principal Clinical Psychologist
78 Lewisham Park
London SE13 6LH

Dear Ms Titman

Title of Protocol: **Caring for a child with a chronic illness**

Protocol No: 97/3/7 *(please quote in all correspondence)*

This protocol was approved at a meeting of the Research Ethics Comittee meeting which took place on 25th March 1997.

The members of the committee present agreed that there is no objection on ethical grounds to the proposed study whose title is given at the head of this letter. I am, therefore, happy to give you our approval on the understanding that you will follow the protocol as agreed.

However the approval is subject to the information sheet being slightly reworded as follows: rather than "how parents ADJUST to caring for a child" to "how parents care for a child". The idea of adjustment and maladjustment seemed to strike an unfortunate note in many of us and we felt patients and relatives would not appreciate it.

You also mention the word Mothers almost exclusively rather than parents.

Another point is that you may run into the problem of parents on being faced with this sounding board about their care will become distressed as a result of doing this study, and we would like to know what facilities exist for this eventuality.

It is your responsibility as the researcher who made the application to notify the Local Research Ethics Committee immediately you become aware of any information which could cast doubt upon the conduct, safety or an unintended outcome of the study for which approval was given.

If there are amendments which, in your opinion or opinion of your colleagues, could alter radically the nature of the study for which approval was originally given, a revised protocol should be submitted to the Committee.

Members of the Committee would like to know the outcome of the study and therefore ask that a report or copy of the results is sent to the secretary in due course.

Yours sincerely

Dictated by Dr C N Mallinson
Chairman
Signed by Annette Jeanes
Vice Chair

APPENDIX 10

Interview

I am going to ask you about.....and about how you manage his/her eczema.

Anything you tell me during the interview will be confidential and I will only keep a record of this information anonymously. If you are unsure about any of the questions please do ask me and if you feel that you don't want to answer a particular question, please tell me.

Name of child:
DOB:
Current Age:
Sex:

Treatment

Who is involved with the treatment of your child's eczema at present ?

What medication is your child currently using ?

Has your child ever had to stay in hospital overnight because of his/her eczema ?

Has your child ever had to attend as a day patient because of his/her eczema ?

Social Background

Who else lives at home apart form you and ....?

Do you (or partner) work outside the home ?

What exactly do you do ?

What would you say is your child's ethnic group?

APPENDIX 11

REACTION TO DIAGNOSIS INTERVIEW

Now I'd like to ask you some questions about your child's eczema. I'd like to tape record this, because it can be difficult to write down everything you say. The tapes will be kept confidential and only I will be able to listen to them, it is just much easier than trying to write down everything. Permission yes/no.

1. Can you tell me when you first realised that….had eczema ? Can you tell me a bit more about what happened then ?

2. Can you tell me how you felt about his/her eczema at that time ?

3. How has his/her eczema changed since then ? Has it stayed the same, got better, got worse, gone up and down…?

4. How would you say you feel about your child's eczema now ?

5. To what extent do you think your child's eczema has affected the way you and your child get on? Do you think it would be any different if she/he didn't have eczema?

6. Parents sometimes wonder why it is their child has eczema. Do you have any ideas or theories about why it is …..has eczema ?

APPENDIX 12

Coding Procedure

See Pianta and Marvin manual for more detailed information:
There are four key areas to look at:

1.Current emotional reaction (not necessarily original emotion – may have changed over time). How much concern do they express? How much of an issue is it for them?

2. Description of hardest aspects to manage: how much passion, how many difficulties listed, emphasis on symptoms or on coping or managing?

3. How much has it affected mother's relationship with child? Balanced reflection on difficulties presented by child's eczema?
(Some parents are reluctant to admit to any difficulties, not necessarily denial, but maybe an attempt to show they've done their best for the child – everyone aware of best option being to "treat them as normally as possible").

4. Ongoing questions about cause of eczema or appropriate treatment. (NB because eczema is very variable and there isn't a "perfect" treatment, some level of doubt or questioning is normal). How intensively are they still engaged in finding a "cure"? Are they still hoping for a miracle, or have they accepted the frustration of the condition? How superstitious are their beliefs about the cause or cure for eczema?

For each interview give a score from 1 to 6:

1- Clearly Unresolved
  EG. High levels of current emotion, wants a miracle, long list of distress caused by eczema. Emphasises anger or symptoms.

2 – Unresolved

3 – Mixed Unresolved
  EG Mixture of resolved and unresolved statements or very neutral balance to overall story but more unresolved than resolved

4 – Mixed Resolved
  EG Mixture of resolved and unresolved statements or very neutral balance to overall story but more resolved than unresolved

5 - Resolved

6 – Clearly Resolved
  EG. Eczema is no longer an issue, does not have many concerns (although may have done in past), accepts it totally as part of life.

Many interviews are likely to contain elements of both resolution and non-resolution. Coding is done by looking at both the overall level/balance, as well as individual statements.

Definitions and Examples

Resolution

"Resolution is characterised by elements reflecting integration of the experience of the child's illness into parents representations, which in turn allows for a re-orientation and refocus of attention and problem solving on present reality."

1. Recognition of change since the diagnosis:
   The difficulty of the diagnostic period is acknowledged, some degree of mourning may continue, but baseline emotion has changed.
   eg "I was hoping it would disappear, but now I accept she has got eczema"

2. Assertion of Moving on in life:
   "That's life", "You have to get on with it"

3. Suspending the search for a reason:
   Trying to find out cause does not dominated parents thoughts, although they may still feel no adequate explanation exists.

4. Accurate representation of child's abilities: No unrealistically high or low expectations.

5. Balanced statements regarding benefits of the experience: ie no outright denial of any difficulties or unrealistic claims of how they have all "learned" from the experience.

Examples:

"It wasn't a major problem, you don't think of eczema being life threatening or anything"

"That's life – most kids have eczema or asthma or something"

"Children do get eczema. I've coped with it quite well"

"Now I accept he's got eczema. Then I was hoping there would be this miraculous cure to make it go away."

Lack of Resolution

"Lack of resolution is characterised by elements associated with grieving. These elements reflect an underlying strategy of attempting to change or alter the reality of the experience."

1. Cognitive distortions:
Including unrealistic beliefs, denial, wish for a different diagnosis in the face of evidence to the contrary.

2. Active search for reasons:
Unrealistic search for a cause, refusal to accept medical opinion

3. Stuck in the past:
Unable to move on from anger or sadness, preoccupation with emotional reaction and emotional impact of the illness.

4. Cut off from the experience of the diagnosis: Denial of any difficulties whatsoever, and minimisation of child's experience.

5. Confusion and mental disorganisation:
Losing train of thought, inconsistent, contradicting self. Swinging between different episodes or reactions.

Examples:

"I'm stressed out about it a lot of the time."

"I just want the doctor to say the magic word and he gets better"

"I hate it. It's horrible. It's horrible. It marks your child...Its not fair you know"

"I can't bear the sound of him scratching. I hate it. I hate it."

APPENDIX 14

SIX AREA, SIX SIGN SCORE

Head and neck	
Erythema	
Exudation	
Excoriation	
Dryness	
Cracking	
Lichenification	
Total	

Subject No.

Subject Initials

Visit

Date

Trunk	
Erythema	
Exudation	
Excoriation	
Dryness	
Cracking	
Lichenification	
Total	

Hands	
Erythema	
Exudation	
Excoriation	
Dryness	
Cracking	
Lichenification	
Total	

Score

0 = absent
1 = mild
2 = moderate
3 = severe

Feet	
Erythema	
Exudation	
Excoriation	
Dryness	
Cracking	
Lichenification	
Total	

Arms	
Erythema	
Exudation	
Excoriation	
Dryness	
Cracking	
Lichenification	
Total	

Legs	
Erythema	
Exudation	
Excoriation	
Dryness	
Cracking	
Lichenification	
Total	

Total body score = ☐

APPENDIX 13

CHILDREN'S DERMATOLOGY LIFE QUALITY INDEX

Hospital No:
Name:
Age:
Address:

Diagnosis:
Date:

CDLQI SCORE: ☐

The aim of this questionnaire is to measure how much your skin problem has affected you OVER THE LAST WEEK. Please tick ✓ one box for each question.

1.	Over the last week, how **itchy**, **"scratchy"**, **sore** or **painful** has your skin been?	Very much	☐
		Quite a lot	☐
		Only a little	☐
		Not at all	☐
2.	Over the last week, how **embarrassed** or **self conscious**, **upset** or **sad** have you been because of your skin?	Very much	☐
		Quite a lot	☐
		Only a little	☐
		Not at all	☐
3.	Over the last week, how much has your skin affected your **friendships?**	Very much	☐
		Quite a lot	☐
		Only a little	☐
		Not at all	☐
4.	Over the last week, how much have you changed or worn **different** or **special clothes/shoes** because of your skin?	Very much	☐
		Quite a lot	☐
		Only a little	☐
		Not at all	☐
5.	Over the last week, how much has your skin trouble affected **going out**, **playing**, or **doing hobbies?**	Very much	☐
		Quite a lot	☐
		Only a little	☐
		Not at all	☐
6.	Over the last week, how much have you avoided **swimming** or **other sports** because of your skin trouble?	Very much	☐
		Quite a lot	☐
		Only a little	☐
		Not at all	☐
7.	Last week, was it **school time?** → **If school time**: Over the last week, how much did your skin affect your **school work?**	Prevented school	☐
		Very much	☐
		Quite a lot	☐
		Only a little	☐
		Not at all	☐
	OR		
	was it **holiday time?** → **If holiday time**: How much over the last week, has your skin problem interfered with your enjoyment of the **holiday?**	Very much	☐
		Quite a lot	☐
		Only a little	☐
		Not at all	☐
8.	Over the last week, how much trouble have you had because of your skin with other people **calling you names**, **teasing**, **bullying**, **asking questions** or **avoiding you?**	Very much	☐
		Quite a lot	☐
		Only a little	☐
		Not at all	☐
9.	Over the last week, how much has your **sleep** been affected by your skin problem?	Very much	☐
		Quite a lot	☐
		Only a little	☐
		Not at all	☐
10.	Over the last week, how much of a problem has the **treatment** for your skin been?	Very much	☐
		Quite a lot	☐
		Only a little	☐
		Not at all	☐

Please check that you have answered EVERY question. Thank you.

Figure 1. Children's Dermatology Life Quality Index questionnaire.

APPENDIX 15

Background Information

1. Child's Age........Years........Months

2. Boy / Girl (please circle one)

3. Please give brief details about why your child has been referred here: (eg asthma, stomach aches, constipation etc)

4. Child's Ethnic Group: Please circle one-
   White
   Asian
   Afro-Caribbean
   African
   Mixed (please give more information if possible)
   Other (please give more information if possible)

5. Who else lives at home with you and your child ?
   Brothers – please give ages
   Sisters - please give ages
   Partner / husband
   Child's grandparents
   Other please give more details if possible

6. Do you or your partner (if appropriate) work ?
   Please give brief details (eg part-time secretary, plumber, teacher etc)

Thank you very much for your help.
Please return the questionnaire before leaving the clinic.