

VOLUME I

**HEALTHY SIBLINGS' ADJUSTMENT
TO CHRONIC ILLNESS
IN A BROTHER OR SISTER**

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1998

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ABSTRACT

The present study aimed to assess (1) well siblings' adjustment (positive and negative aspects), (2) level of parental awareness of well siblings' cognitions about the illness and its impact on their everyday lives, and (3) families' views of the appropriateness of Clinical Psychology services for healthy siblings. The participants were a well sibling and mother from 62 families of children with a range of chronic illnesses. The study's design was cross-sectional, and questionnaire-based within a structured interview format. Qualitative data was collected to illustrate quantitative findings.

Well siblings appeared to be at low risk of overall maladjustment, compared to children in the normal population. The majority of well siblings were displaying a high level of pro-social behaviours. The factors most strongly associated with well siblings' adjustment were parental awareness of well siblings' cognitive responses to the illness and the burden of care demanded by the illness. There was no evidence of parental awareness being a mediating factor between well siblings' adjustment and parental adjustment. Mothers rated well siblings as having significantly more negative cognitions about the illness than reported by well siblings themselves. These results supported McCubbin and Patterson's (1983) systemic model of family adjustment. Implications of the findings are considered for families, researchers and clinicians.

Most mothers and well siblings reported that Clinical Psychology services were not appropriate to the siblings' needs at the present time. Many parents commented that they would like to be supported in helping their healthy children themselves.

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INTRODUCTION

1.1 Overview of the Topic and Chapter One: Introduction

There has been minimal research to facilitate the development of specific and evidence-based programmes of care for healthy siblings with a chronically ill brother or sister. The present study aimed to investigate well siblings' adjustment, and also parents' awareness of healthy siblings' cognitions about the illness and its impact on the siblings' lives. Parents and well siblings were also asked about the appropriateness of specific Clinical Psychology services which are slowly being introduced into paediatric hospital settings.

1.1.2 Why Study Families with a Child with a Chronic Illness ?

Meeting the medical and psycho-social needs of families with a child with a chronic illness is a new challenge for the health care system (Bradford, 1997; Sloper & While, 1996). Recent advances in medical technology have meant that the survival rates of children with a wide range of chronic conditions have improved significantly (Eiser, 1998). Such conditions are described as "protracted and can result in a number of diverse and adverse outcomes, ranging from normal life expectancy to death" (Bradford 1997, p.6). Eiser (1996) estimates that as many as 10 to 15 per cent of the total childhood population has a chronic illness, with 1 to 2 per cent of the total population having a severe condition. Increasingly clinicians are recognising the tremendous emotional impact chronic illness can have on family life. Families must adapt to a range of practical demands, such as regular hospital admissions and daily routines of care.

1.1.3 Why Study Well Siblings' Response to Chronic Illness ?

Many parents identify concern about their healthy children as an additional source of distress over and above that related to the disease (Eiser, 1993). However, research into the impact of chronic disease on healthy siblings has been a neglected area (Bradford, 1997; Eiser, 1998), and the available studies have produced variable results. For example, cited rates of well sibling disturbance vary widely: 66 per cent of siblings of children with spina bifida (Tew & Lawrence, 1973), 25 per cent of siblings of children with cancer (Sloper & While, 1996) and 57 per cent of siblings of children with cancer (Carpenter & Sahler, 1991). There are also occasional reports of siblings being less well adjusted than the ill children themselves (Tew & Lawrence, 1973).

The literature suggests that well siblings experience a range of psychological problems, and there has been little research into positive aspects of their adaptation. Carpenter and Sahler (1991) found that within a sample of 107 well siblings, difficulties included emotional lability (31 per cent), negative attention-seeking behaviours (25 per cent), changes in academic performance (22 per cent) and withdrawal (16 per cent). Other changes in behaviour (such as somatic complaints), disturbed sleeping and eating patterns, and bed-wetting or other regressive behaviours were also reported. However, there is some evidence of well siblings developing pro-social skills and an increased capacity for sensitivity and understanding (Ferrari, 1984,1987; Stallard, Mastroyannopoulou, Lewis & Lenton, 1997).

This range of well siblings' responses has initiated an exploration of mediating factors which may influence their adaptation. Bradford (1997) proposes that parental

availability (both emotional and physical) affects siblings' adjustment. This hypothesis is supported by findings which suggest mediating roles of maternal level of distress (Tew & Lawrence, 1973), marital disharmony and maternal dissatisfaction with external social support (Ferrari, 1984; Sloper & While, 1996), and level of disruption to day-to-day family life as a result of the illness (Sloper & While, 1996).

This study additionally aimed to explore whether the accuracy of parental awareness of their healthy children's cognitions of the illness experience was associated with their healthy children's adjustment. Cognitions are defined as thoughts, feelings and attitudes about an event or episode (Carpenter & Sahler, 1991). Furthermore, it was hypothesised that accuracy of parental awareness would also be associated with parents' psycho-social resources (parents' emotional adjustment, parents' satisfaction with social support and the burden of care demanded by the illness).

1.1.4 Clinical and Methodological Importance of the Present Study

The implications of the study's findings are relevant to therapeutic interventions and assessments in research contexts. First, the identification of determinants or modifiers of risk to family members is vital to develop protocols for early intervention. This requires that clinicians and parents are aware of common healthy siblings' cognitions and the likely emotional, behavioural and social sequelae. Second, the service-related questions provide direct information for service planning and provision. Third, an assessment of parents' awareness will indicate whether parents' responses can be used as a valid proxy assessment of their healthy children's cognitions. In research studies primary reporters of well siblings' adjustment have been parents, mostly mothers. This

is less of an issue in clinical practice where it is standard to interview parents and children, and the degree to which the assessment relies upon parents' report reasonably depends upon the child's age (Klein, 1991).

1.1.5 Outline of Chapter One

This chapter reviews the literature relevant to well siblings' adjustment to paediatric chronic conditions. It begins with a historical overview of the methodology used and an outline of family systems theory. Psycho-social factors at the well sibling, parent and family levels, which have been empirically associated with well siblings' responses are then examined. Particular emphasis is paid to well siblings' cognitions of both chronic illness and its impact on their interpersonal functioning. There have been few studies of parents' awareness of their children's cognitions and reference is therefore made to non-paediatric research. The implications of the literature review are discussed in the context of the rationale of the present study, before the chapter concludes with the research questions.

1.2 Summary of Research on Well Siblings' Adjustment to Chronic Illness

Over the last thirty years, a growing body of research has examined the difficulties that healthy siblings may develop (Carpenter & Sahler, 1991; Eiser, 1993; Sloper & While, 1996; Tew & Lawrence, 1973). This section begins by reviewing the concept of adjustment and discusses the range of cited well sibling responses to chronic illness. Methodological limitations in comparing research findings are also discussed.

The literature is striking in its absence of definition of adjustment. The Dictionary of Psychology (Reber, 1985) defines adjustment as “the relationship that any organism establishes with respect to its environment” (Reber, 1985, p.13). Reber (1985) further states that the term ‘good adjustment’ implies “that the individual is involved in an ongoing process of developing his /her potential, reacting to and in turn changing the environment in a healthy effective manner” (Reber, 1985, p.13).

The conceptualisation of well siblings’ adjustment as a process, rather than a state, is supported by Bluebond-Langner’s (1996) work with siblings of children with cystic fibrosis. She found evidence of well siblings’ responses changing over the life cycle of the illness, for example initial shock at diagnosis followed by acceptance. For the purpose of this study, ‘good adjustment’ is succinctly defined as a current absence of emotional, behavioural or social difficulties. However, emphasis is also paid to identifying positive well sibling behaviours in the study’s methodology.

Due to the lack of a universal operational definition of adjustment, quantitative research designs have used many different measures. This has resulted in a difficulty comparing reported rates and types of siblings’ maladjustment. For example, Tew and Lawrence (1973) reported on teachers’ accounts of difficult behaviours at school, and Stallard, Mastroiannopoulou, Lewis and Lenton (1997) reported on parents’ accounts of emotional and behavioural symptoms. A few studies have attempted to examine siblings’ behavioural responses more systematically, by examining their use of coping strategies. This has produced inconclusive results. For example, Sloper and White

(1996) found no pattern in the use of problem-solving and emotion-focused coping strategies (Folkman & Lazarus, 1984) in a group of siblings of children with cancer.

More recently quantitative research designs have been influenced by the methodology and findings of qualitative studies (Carpenter & Sahler, 1991; Sloper & While, 1996) and cognitive theories of stress and coping (Folkman & Lazarus, 1984). These have emphasised the importance of eliciting the perspective of healthy siblings as well as other family members. By using an exploratory approach, qualitative findings indicate a range of well siblings' responses. Kiburz (1994) described well siblings of children with spina bifida as concerned and empathic towards their siblings. Ferrari (1984, 1987) reported that healthy siblings of children with diabetes and developmental disorders were more sensitive and socially mature than a control group of siblings. However, they also expressed concerns about their own intellectual and school achievements, personal happiness and life satisfaction. Cleveland and Miller (1977) reported an elevation in pro-social behaviours over time, finding that well siblings of children with chronic illnesses tended to choose careers in the caring professions. They hypothesised that this was the result of heightened awareness of others' suffering as well as a desire to be involved in help and care.

Horowitz and Kazak (1990) reported quantitative findings that a sample of 25 pre-school children with a sibling suffering from cancer were more considerate and helpful compared with age-matched controls. Their method was exemplary in eliciting information from siblings and parents. The children completed a pictorial scale of perceived competence and parents completed a child behaviour scale. The

experimental group showed no evidence of maladjustment, and scored higher on the pro-social scale in terms of willingness to help, giving gifts, praising, sharing and showing affection.

In summary, there have been two major shifts in quantitative research concerning siblings' adjustment; first towards a hypothesis that chronic illness can result in both negative and positive effects on siblings, and second away from a sole reliance upon observed reports of siblings' adjustment (by parents or teachers). The focus on individual well siblings rather than the system they live within has largely remained (Carpenter & Sahler, 1991). As a result of methodological inconsistencies between studies, there are mixed impressions about siblings' responses to chronic illness. However, the variance suggests that many factors interact to determine siblings' adjustment.

1.3 Systemic Approach to Exploring Well Siblings' adjustment

A number of theoretical perspectives suggest the importance of relationships for adjustment. Attachment theory, for example, proposes that experiences in early intimate relationships and internal representations of relationships are associated with mental health (Bowlby, 1979). Similarly object relations theory is based on the premise that adjustment difficulties in adulthood stem from problems in early mother-child relationships (Winnicott, 1957) and psychoanalytic theory sees the resolution of early oedipal relationship as a basis for later adjustment (Freud, 1958). The present study used systemic theory to conceptualise how the well sibling-parent relationship impacts on the healthy sibling whilst the illness is progressing.

The basic concept of family systems theory is that individuals live in a social context, and are therefore best understood by examining their relationships and the environment they interact within (Hayes, 1991). Families are seen as groups of individuals with relationships based upon structures or rules. The latter regulate the different roles of family members (e.g. physically ill person, disciplinarian) and their individual belief and communication patterns.

The family is recognised as an organised and self-regulating system, which functions along a continuum of adaptation (Bateson, 1972). It is proposed that families re-negotiate their structure when faced with a change, for example the ill child relapsing and being admitted into hospital. The re-negotiation is conceptualised as a circular process, in which every family member influences the others and is in turn influenced by them. Psycho-social symptoms may develop in any or all family members, and are seen as resulting from the ineffective adjustment of families' relationships, behaviours and beliefs.

McCubbin and co-workers' systemic model is used as the theoretical framework of this thesis (McCubbin, Joy, Cauble, Comeau, Patterson & Needle, 1980; McCubbin & Patterson, 1983). They suggest that successful adaptation of family members to a crisis is a function of two factors: first, the manner in which the family uses new and existing resources (psychological, interpersonal, community and material) to deal with the added physical and emotional demands imposed by the change, and second the manner in which the family handles the differing cognitions of each family member of the

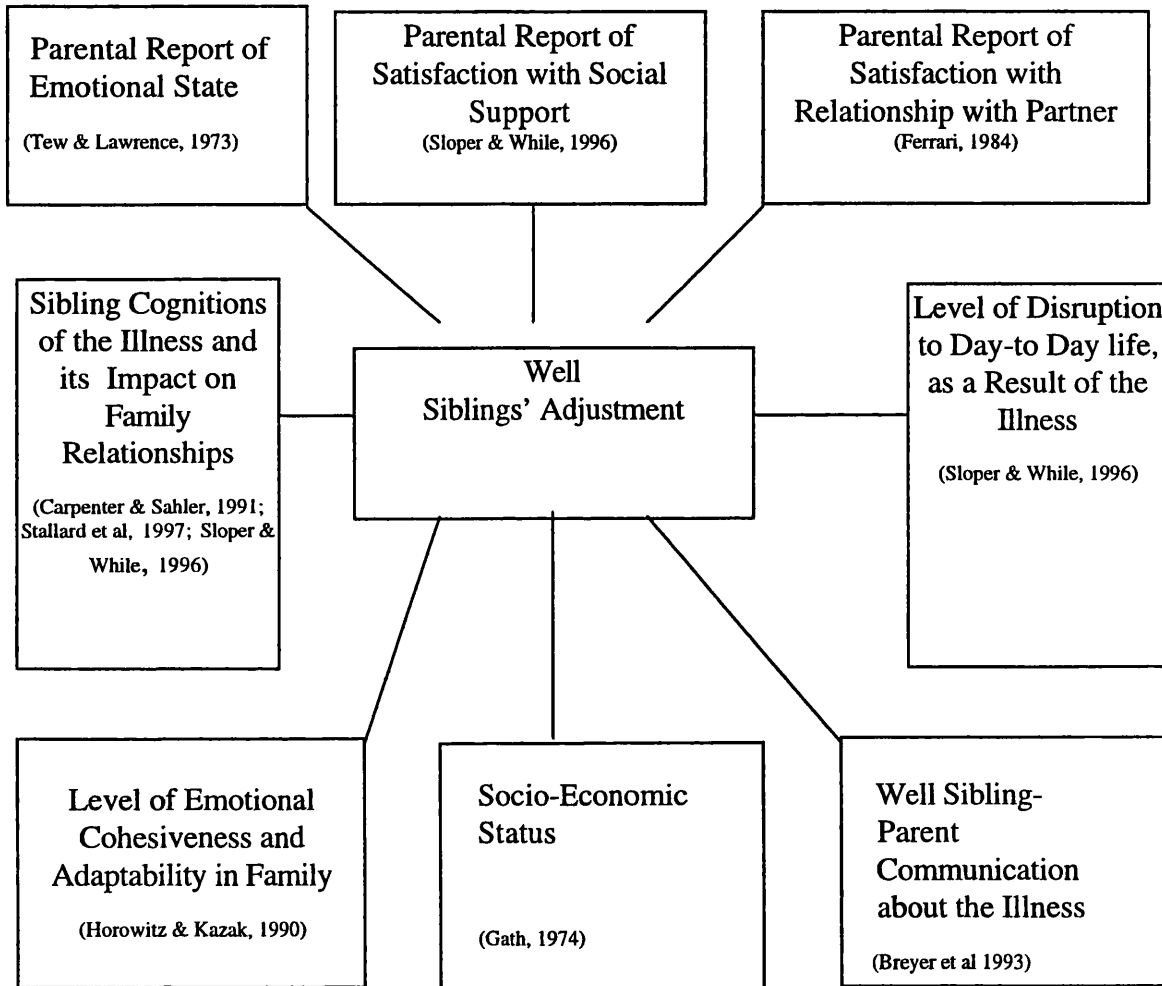
impact of the illness. The model conceptualises children as having dependent social roles in the family system.

The central tenet of this thesis was that the systemic model provides a useful framework for identifying well sibling and family factors that, alone or in combination, are likely to influence siblings' adjustment to chronic illness. Here, the focus is primarily on the potential role of parental awareness as one of the theoretically important variables in predicting well siblings' adjustment.

1.4 Psycho-Social Factors Hypothesised to Underlie Siblings' Adjustment

Systemic theory hypothesises that well siblings' adjustment depends upon their families' use of a range of resources, individual members' cognitions of the illness experience and members' awareness of each others' cognitions. Figure 1 summarises the psycho-social factors associated with poorer well siblings' adjustment: parental distress, parental satisfaction with relationships (intimate and external social supports), level of day-to-day disruption caused by the illness, socio-economic class, well sibling-parent communication about the illness, well siblings' cognitions of the dynamics of family relationships and also measurements of families' adaptability and cohesiveness. Parents' awareness of siblings' cognitions has been a relatively neglected area in paediatric psychology, and so is discussed with research data from non-paediatric samples.

Figure 1:
Psycho-Social Factors Associated with Well Siblings' Adjustment



1.4.1 Well Sibling Variables

An increasing number of studies have investigated well siblings' adjustment using cognitive theories of stress and coping (Folkman & Lazarus, 1984). The studies suggest that well siblings' cognitive appraisals of the illness experience are more strongly associated with their overall adjustment, than level of self-esteem, knowledge about the illness, use of coping strategies or extra-familial social support (Ferrari, 1984, 1987; Sloper & While, 1996). No studies were identified that have specifically investigated the influence of personality characteristics or intelligence in well siblings' adjustment.

One of the first studies investigating well siblings' cognitions was carried out by Menke (1987). She interviewed healthy siblings of children with chronic conditions about concerns related to the illness and to changes in the family. More than half the children expressed concerns, which largely centred on the prognosis, medication, and the sick child's feelings. Many also worried about themselves and their parents. Many healthy siblings felt protective towards their ill sibling, and this was especially true of those who were older. Although 71 per cent of the healthy children reported that difficulties arose because of the extra attention that parents gave the ill children, 59 per cent reported they felt able to emotionally accept the situation. Sloper and While (1996) hypothesise that this ability to self-reflect is an important aspect of resilience. They suggest that well siblings who do not display adjustment difficulties are able to reflect upon the loss of attention and compare their own needs to the ill child's needs.

This self-reflection hypothesis has been supported by three independent studies (Carpenter & Sahler, 1991; Stallard et al, 1997; Sloper & While, 1996), with samples of healthy children with siblings with chronic life-threatening diseases. The results indicated that healthy siblings who showed adjustment problems (according to parents), also rated items concerned with interpersonal adjustment (especially family adjustment) in a significantly more negative fashion than those who were reported to be well adjusted. The items inquired as to whether the healthy siblings thought the illness disrupted patterns of family functioning, whether they felt a lack of resources for coping with their feelings, and whether they reported not wanting to bother parents with worries. These three investigations used the same tool: The Sibling Perception Questionnaire (Carpenter & Sahler, 1991), which is described further in the Method chapter (page 42).

In summary, it is apparent that well siblings experience a range of concerns and anxieties, which focus on the sick child and aspects of their own lives. Siblings' cognitions about the illness' impact on family functioning have been repeatedly associated with the success of their adjustment. It appears that perceived loss of attention for siblings is not a critical factor per se in their adjustment, but their interpretation of this loss is important. Sloper and While (1996) suggest that well siblings who are able to reflect on the ill child's needs in comparison to their own are psychologically more resilient.

1.4.2 Parental and Illness Variables

Most psychological research on well siblings' adjustment has attempted to identify specific parental factors associated with well siblings' adjustment. Three factors have been repeatedly investigated: maternal distress, maternal satisfaction with social support, and socio-economic status. More recently, the disruption the illness causes to everyday life has been proposed as a mediating factor.

Both qualitative and quantitative studies have demonstrated that siblings' adjustment is associated with parental emotional states. Ferrari (1984) and Sloper and While (1996) reported that maternal dissatisfaction with both social and intimate support, respectively, were associated with well siblings' adjustment. Stocker (1991) reported that within a non-paediatric sample of seven year olds, maternal warmth was positively associated with fewer feelings of loneliness and fewer behaviour conduct problems. Similarly, Tew and Lawrence (1973) found that maternal distress was associated with school behavioural problems in well siblings of children with spina bifida. Eiser (1993) reported that well siblings of children with renal conditions were less well adjusted when their mothers deemed the condition to be more serious.

There have been conflicting findings about the influence of socio-economic status on well siblings' adjustment. Gath (1974) (cited in Eiser, 1993) reported that psychiatric disturbance in well siblings was higher in middle-class families than in working class families, where there was a child with long-term physical disabilities. She hypothesised that the adaptability of families was affected by the expectations associated with their social class. She suggested poorer families had lowered (or more realistic) expectations

of life in general, and therefore perceived the disease to add only marginally to their other problems. In contrast high achieving families perceived the impact of the disease more negatively, seeing their ambitions and opportunities thwarted, which considerably increased their psychological distress.

However, at six months post diagnosis of cancer, Sloper and While (1996) reported that well siblings from lower socio-economic groups were less well adjusted than siblings from higher groups. They hypothesised that parents from lower classes were less mobile to visit well siblings at home. A related finding was that the number of nights the ill child spent in hospital in the last six months was negatively associated with siblings' adjustment. Sloper and While suggested that their findings reflected an association between well siblings' adjustment and the degree of disruption to day-to-day family life.

A related issue has been the recent academic discussion as to the relative merits of taking a categorical or non-categorical approach in sampling when investigating psychological outcomes. A non-categorical approach means participants are selected by general characteristics of disease (e.g. chronicity, burden of care) rather than the disease label per se. Several authors argue that the identification of common stresses and challenges posed by paediatric conditions is more appropriate for the development of health services for local communities which present a range of disorders (Eiser, 1993; Stein and Jessop, 1982).

In their comprehensive review of investigations of sampling methods and psychological outcomes in families with a chronic disorder, Pless and Perrin (1991) conclude that little if anything is gained in understanding family adjustment by using a categorical approach:

“there are a limited number of difficulties frequently experienced by many, if not most, families who have a child with a chronic disorder. The difficulties vary only slightly from disorder to disorder or from family to family. If anything, the nature of the family, more than the nature of the disorder, is likely to determine the frequency with which certain problems are experienced”

(Pless & Perrin, 1991, p.49)

In summary, the common theme in these findings is the importance of parental availability at both emotional and physical levels to well siblings' adjustment. Parents may be emotionally unavailable due to personal distress about the illness or its impact, or due to their dissatisfaction with social relationships. They may be physically unavailable due to the demands of the illness. Indeed, Gribble, Emory, Wyman, Work, Wannan and Eaoof (1993) found that a sample of children (n=656) living in a US inner city were better adjusted (as rated by their teachers) if their parents were involved in joint activities with them and showed interest in other areas of their lives (e.g. school).

1.4.3 Family Variables

The importance of a sound parent-child relationship for positive child adjustment is non-controversial. Two family factors are reviewed: the general quality of the families' inter-relationships and well sibling-parent communication about illness facts per se.

A frequently quoted study was carried out by Horowitz and Kazak (1990). Using standardised questionnaires, they found that family adaptability and cohesion were negatively related to behaviour problems in pre-school siblings (n=25) of children with cancer. They defined adaptability as "the family system's flexibility and ability to change" (Horowitz & Kazak, 1990, p.222) and cohesion as "the degree of emotional separateness or emotional connection in a family" (Horowitz & Kazak, 1990, p.222). They suggested that siblings' needs were better met when families found a balance between extreme flexibility (or chaos) and rigidity to cope with the unpredictable course of the illness. They also proposed that siblings in more emotionally cohesive families were more likely to have their needs met because their parents were more likely to be aware of them.

There have been contradictory findings about associations between well sibling-parent communication about illness facts and siblings' adjustment. Breyer, Kunin, Kalish, and Patenaude (1993) interviewed 69 well sibling-parent pairs of families with a child with cancer. They commented that between two sub-samples of the seven best adjusted siblings and six least well adjusted siblings, there were significantly different quantities of sibling-parent communication about illness facts. They suggested that parents may

attempt to 'protect' siblings from the illness by not talking to them about it, which inadvertently leaves siblings feeling isolated from family activity.

In summary, research indicates siblings adjust better if the family unit is flexible to change, emotionally cohesive and supportive to siblings. Parents' continued availability in siblings' day-to-day lives also appears important in siblings' adjustment, over and above the amount of explicit communication about the illness per se.

1.4.4 Paediatric Studies of Parents' Awareness of Well Siblings' Cognitions

A comprehensive literature review found only three paediatric psychology studies which assess parents' awareness of their healthy children's cognitions. Factors associated with parent-child agreement are reviewed with reference to non-paediatric investigations in this area.

Breyer et al (1993) interviewed 69 well sibling-parent pairs in families with a child with cancer, using both open-ended and closed questions. The age range of the healthy siblings was five to eighteen years. They asked open questions about siblings' current concerns. Raters then coded the answers into content categories as to whether the answer focused on the ill child or the well sibling. The investigators reported that 66 per cent of parents said that the major impact of the illness was loss of parental attention, however 66 per cent of siblings reported that the major impact was the child being ill itself. Eiser (1993) hypothesises that parents' response to this question may have reflected their own anxieties about not giving siblings as much attention, rather

than their prediction of siblings' concerns. The finding may also reflect well siblings giving what they perceive to be a socially desirable reply.

This study also attempted to look at an association between well sibling-parent agreement and well siblings' adjustment. Healthy siblings and parents were asked about any changes in their relationship since illness onset: 73 per cent of siblings said there had been none and 80 per cent of parents said none. However, a different picture emerged when they went on to compare the responses of siblings and parents of seven siblings who had developed pro-social skills, and six siblings who had developed behavioural difficulties. Their impression of these small samples was that siblings' adjustment was associated with level of sibling-parent agreement on reports of changes in their relationship. Parents and well adjusted siblings were more likely to agree that their relationship had not changed, whereas parents and poorly adjusted siblings were more likely to be discrepant in their reports of whether their relationship had changed. This result was also supported by Gribble et al's (1993) study cited earlier, which interviewed families living in stressful conditions (i.e. poor US inner-cities). They found that a higher level of agreement between parents' and children's views on their relationship was associated with resilient outcomes in children.

Breyer et al's study is limited in several ways. First, 18 parents were represented twice in the study and therefore the data points were not independent. Second, sampling took place over five years due to difficulties in recruiting sample numbers. No consideration was paid to the changes in medical and psycho-social care different families may have received over the time period. Third, some participants were as

young as five years old. They may have answered questions in a manner which they deemed more socially acceptable, or may not have had the intellectual ability to understand, reflect on and answer interview questions. The authors did not report on any associations between gender or age and parent-child agreement. Fourth, there was likely to be a sample bias due to recruitment solely taking place from a Sibling Support Group.

In a qualitative study, Menke (1987) interviewed 72 parents and healthy siblings (aged between six and twelve years) of children with cystic fibrosis, congenital heart disease, spina bifida or severe burns. Her findings indicated that parents tended to vastly underestimate concerns of the healthy siblings. She found that 35 per cent (n=25) of parents reported that the sibling had no concerns, compared with 11 per cent (n=8) of siblings who reported they had no concerns. Chi-squared analysis indicated that this difference was significant. She also reported that only 8 per cent (n=6) of parents reported that the sibling had concerns unrelated to the ill child (e.g. own health and school progress), whereas 21 per cent (n=15) of siblings reported they had concerns unrelated to the ill child. The level of agreement did not vary with age or gender of the sibling, or chronic condition.

The final study by Craft and Craft (1989) was exemplary in several ways. First, it was the first systematic comparison of sibling and parent responses on a single measure. Additionally, the researchers emphasised to parents the importance of thinking from their healthy child's perspective and trying to separate this from their own point of view. They analysed 123 sibling-parent pairs (age range of siblings was five to

seventeen years), from families where a child had a range of acute, chronic and progressive illnesses. Siblings and parents separately completed a twelve item categorical scale to ascertain perceived changes in siblings' functioning from the perspective of the healthy sibling. Changes investigated included: physical functioning (trouble sleeping, amount of food eaten, feeling physically healthy) emotional functioning (becoming angry, difficulties concentrating at school, feeling nervous), behavioural functioning (fighting with other children, having night-mares, bed-wetting, nail-biting), and interpersonal functioning (wanting to be alone, wanting to be closer to parents).

The highest agreement was on behavioural changes that occurred at home; bed-wetting (87 per cent agreement), nail-biting (76.4 per cent agreement) and experience of nightmares (71.5 per cent agreement). There was much less agreement about changes in siblings' emotional responses ('getting angry' (51.3 per cent agreement), wanting to spend time with parents (49.6 per cent agreement)), or difficulty concentrating at school (54.3 per cent agreement). The reader should note that the use of percentage agreements to compare sibling and parent responses on each item does not control for chance agreement.

Overall, well sibling-parent agreement on the number of changes reported varied with the threat of the illness, siblings' age and involvement in family activity. Parents and healthy siblings agreed most when the hospitalised child had a one-off acute illness, compared with when the child had a chronic disease or a progressive illness. Craft and Craft also reported that there was a lower level of agreement in the younger age group

(age range five to nine years) than in the older age group (ten to seventeen years). Moreover, there was a higher level of agreement in the older age group when the siblings were able to visit the sick child in hospital. Similarly, other studies have reported that level of agreement varies with amount of family discussion around problematic issues (Csikszentmihalyi & Graef, 1975).

In a non-paediatric review of factors that are likely to be associated with the accuracy of parent-child agreement, Klein (1991) examines child age and gender, parental psycho-pathology and child adjustment. She concludes that there have been contradictory reports about associations between age and accuracy of parent-child agreement. Similarly to Craft and Craft (1989), Herjanic and Reich (1982) report better agreement at later (twelve to sixteen years) than earlier ages (six to nine years) on a range of objective and subjective items, but no statistical contrasts were conducted. However, this finding was not replicated in two other studies (Breslau, Davis & Prabucki (1988); Weissman, Wickramaratne, Warner, John, Prusoff, Merikangas & Gammon (1987)). Three studies which contrasted parent-child agreement between girls and boys, found no significant effect of gender (Breslau et al, 1988; Edelbrock, Costello, Dulcan, Conover & Kalas, 1986; Reich, Herjanic, Welner, & Gandhi, 1982).

There is also contradictory evidence about associations between parents' and children's emotional states on agreement level. Breslau et al (1988) found that the presence of a major depressive disorder in a mother significantly raised the reported number of child adjustment symptoms, but agreement per se on this measure was not

affected. Watchel, Rodrigue, Geffken et al (1994) reported that parent-child agreement varied with parental anxiety. Edelbrock et al (1986) found no evidence of length of child's depressive disorder being associated with agreement. These latter findings are contrary to the findings by Breyer et al (1993) and Gribble et al (1993) discussed earlier.

In summary, little paediatric research has investigated the discrepancies between well siblings' and parents' reports on children's cognitions and therefore research is still at an initial exploratory phase. Parents appear most accurate in reporting factual information (e.g. bed-wetting incidents), than their children's internal states (e.g. cognitions, emotions). Parents also tend to underestimate the number of well siblings' concerns, and report unlike well siblings that they are concerned about loss of parental attention. However, this finding may be due to a lack of validity and reliability of the question as discussed.

It appears that the congruence of parent-child agreement varies with level of involvement in joint activities. These parents are likely to be both more physically and emotionally available to their healthy children, and the present study would hypothesise that they will be more aware of these children's cognitions about the illness experience. There is no evidence of agreement varying with the gender of the child, and inconsistent evidence with the adjustment status of parent and child and the child's age. Associations between agreement and parental satisfaction with social support (external and marital (if applicable)) and day-to-day disruption caused by the illness have not yet been explored.

1.5 Limitations of Previous Studies

Paediatric research into the psycho-social effects of childhood illness on healthy siblings has been limited in several ways. Most significantly until the last ten years, studies have concentrated on outcome measures (i.e. the degree of mal-adjustment compared to a control group). This approach is contrary to the systemic conceptualisation of illness and also does not take into account the changing process of adjustment over time. It also does not help to identify specific factors which place a healthy sibling at risk (or conversely facilitate adaptation) and therefore design preventative interventions (Gribble et al, 1993).

Additionally, many different adjustment measures have been used which has made comparison between findings (rates and nature of adjustment) difficult. The emphasis of these measures has been on overt signs of psychological disturbance, which have ignored the centrality of individuals' appraisals of events in the process of stress response and coping suggested by cognitive and systemic theories (Folkman & Lazarus, 1984, and McCubbin & Patterson, 1983, respectively). Further, the assessment of healthy siblings' cognitions has been problematic because of a lack of standardised instruments. This has been recently addressed by the development of the Sibling Perception Questionnaire (Carpenter & Sahler, 1991).

1.6 Clinical Psychology Services for Well Siblings

Systemic theory has been widely applied in the psychological treatment of families within paediatric settings (Altschuler, 1997). The therapy aims to help families re-organise their relationships and roles (and their beliefs about these) to meet new

demands imposed by the illness and also coinciding factors in normal family development (e.g. an adolescent asserting independence). Some hospitals are now beginning to offer services tailored specifically for well siblings' needs, but a comprehensive literature review found no investigations of families' opinions of the appropriateness of these services.

Specific services offered to well siblings and their families are mostly individual and group interventions. Individual services are recommended for well siblings who are likely to be seriously affected by the illness process (e.g. sick child's death) (Sloper & While, 1996). Such services are most frequently offered at specialist hospitals for conditions such as cancer. The aims and content of well sibling groups appear to vary enormously from informal support groups, to psycho-education about the illness (Cunningham, Betsa & Gross, 1981; Williams, Hanson, Karlin, Ridder, Liebergen, Olson, Barnard & Tobin-Rommelhart, 1997) to group therapy (Heiney, Goon-Johnson, Ettinger & Ettinger, 1982). Evaluations of both types of intervention indicate that they provide well siblings with an opportunity to decrease their sense of isolation, to ventilate negative feelings, to learn facts about the illness and, with the group interventions, to learn from one another.

1.7 Present Study

The study aimed to collect well siblings' and parents' opinions of two Clinical Psychology service provisions, in addition to exploring the process of well siblings' adjustment. The latter aims were to investigate positive and negative aspects of well siblings' adjustment, to assess levels of parental awareness of well siblings' cognitions

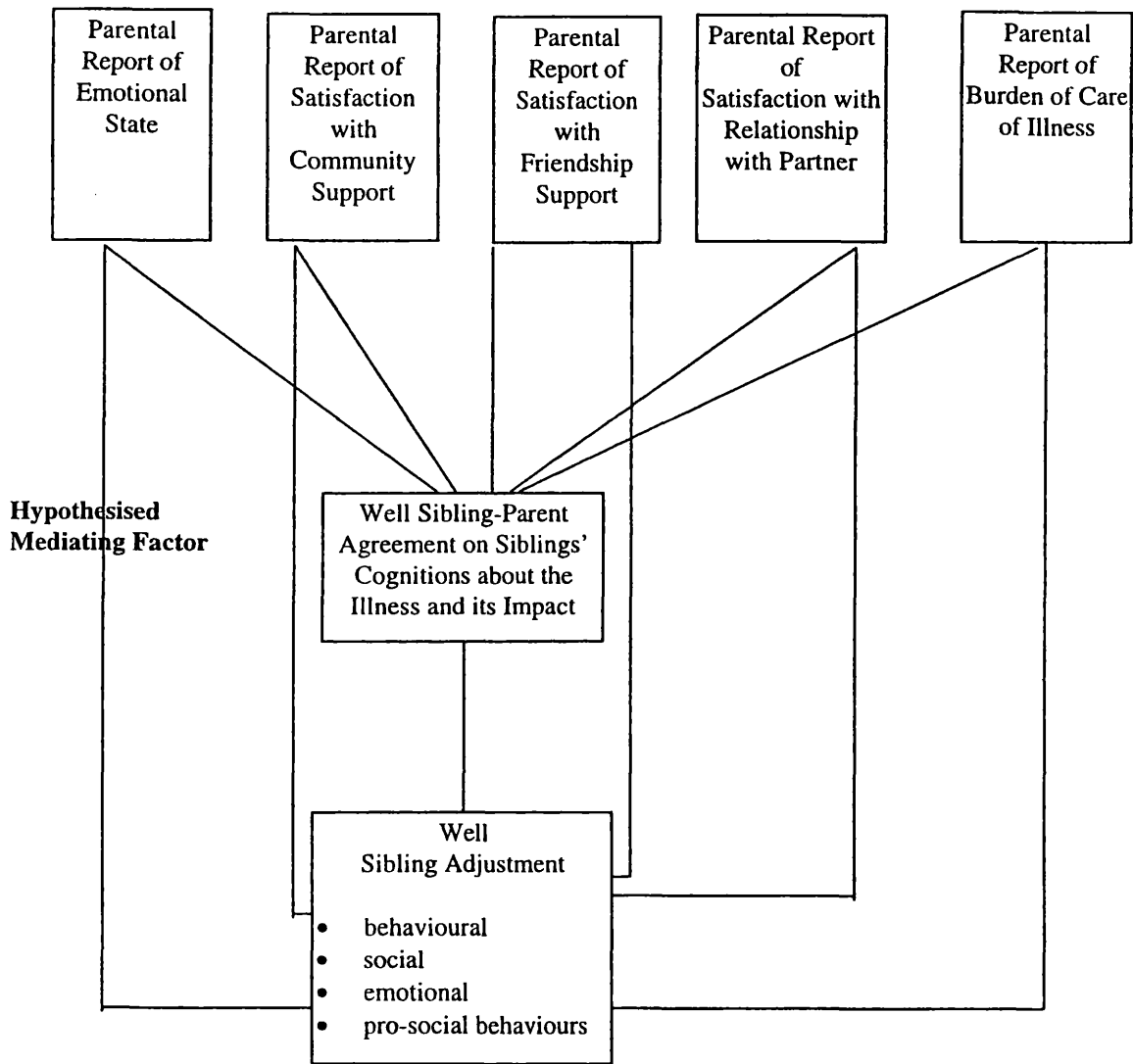
about the illness and its impact, and also to investigate whether parental awareness was a mediating factor between well siblings' adjustment and parental psycho-social adjustment. This latter hypothetical model is presented pictorially in Figure 2.

As proposed by McCubbin et al's (1980, 1983) systemic model, well siblings' adjustment is hypothesised to be dependent upon the family system. The psycho-social variables relate to parents' adjustment: parents' emotional state, parents' satisfaction with social support (community, friendship and intimate relationships) and burden of care of the ill child. Contrary to previous paediatric research, well siblings' adjustment was studied in terms of difficulties (behavioural, emotional and social) and the presence of skills, here pro-social behaviours.

The research questions were:

1. What is the adjustment status of well siblings, in comparison to standardised norms?
2. What are the percentage agreements between parents and well siblings about well siblings' cognitions about the illness and its impact, across the whole sample?
3. Do parental psycho-social variables affect well siblings' adjustment directly, but also indirectly through well sibling-parent dyad agreement ?
4. Do parents and well siblings report that it would be appropriate for the well sibling to receive psychological support from either (a) participating in a group for brothers and sisters who have chronically ill siblings and /or (b) individual sessions with a member of the hospital staff ?

Figure 2
Hypothesised Model of Well Siblings' adjustment



METHOD

2.1 Overview of Method

Separate interviews were carried out with 62 well siblings and their mothers from families of children with a chronic disease. Both interviews were structured and based upon questionnaires. The parent interviews elicited information about the mothers' emotional state, satisfaction with available social support and burden of care demanded by the illness. A parent version of the Sibling Perception Questionnaire (SPQ) was developed to elicit mothers' awareness of their healthy children's cognitions about the illness and its effects on their lives. Mothers and well siblings were administered respective versions of the SPQ. They were also asked whether Clinical Psychology help, in individual or group formats, would be appropriate for the well sibling. Comments made both by mothers and well siblings were recorded.

2.2 Design

The design was cross-sectional and questionnaire-based, involving a one group sample. A control group was not used because the research questions referred to families' specific adjustment to illness, and most importantly the meanings well siblings gave to the illness and its disruption to family life.

2.3 Procedure

The majority of interviews were conducted in families' homes. The interviews were structured. Separate interviews were conducted with the mother and one well sibling of the ill child. In order to avoid confounding the data analysis, only one well sibling in

each family was included. The target sibling was identified as the one nearest in age to the ill child who fell within the age limits of the study.

The parent interview lasted approximately one hour and included five questionnaires (Malaise Inventory, Inventory of Parent Experiences, Burden of Care Index, SPQ (parent version) and the Strengths and Difficulties Questionnaire). The well sibling interview lasted approximately 20 minutes and included the SPQ (well sibling version). Older children (10+ years) often chose to read questions aloud themselves from the questionnaires before giving their answer. Both mothers and well siblings were asked only to reflect on the last six months, when responding to the questions asked. When the parent version of the SPQ was administered, mothers were asked to try to put themselves in the position of the well sibling and separate themselves as much as possible from their own experience of the illness.

A pilot study of four families with different illnesses (cancer, spina bifida, diabetes) was carried out prior to contacting families. The content and procedure of the interview schedule appeared acceptable and relevant. As explained later, amendments were made to the basic response format of the SPQ.

2.4 The Sample

The sample was made up of families with a child with a serious and chronic disease of more than three months duration (i.e. a non-categorical approach to sampling). Other inclusion criteria were that well siblings were aged between eight and sixteen years and did not have physical health problems themselves, the ill child was aged eighteen years

or less, the family spoke English and that the ill child's condition required demanding daily routines of care and /or vigilance (as judged by a Consultant Paediatrician).

Table 1 shows the demographic and illness characteristics of the sample. There were no families where the mother was not present. For the purpose of this study, the family's socio-economic group was defined by the profession of the male parent if there were two parents present, and by the mother if she was a single parent. Compared to the 1981 Population Census for England and Wales (Central Statistics Office, 1995), this sample was biased towards the 'non-manual' socio-economic group.

The participants were recruited from five London hospitals. The researcher approached Consultant Paediatricians with details of the study and their agreement was obtained to contact eligible families from their patient lists. Three hospitals provided a full hospital list, and two hospitals identified eligible families themselves. At one hospital, the Clinical Psychology department additionally provided a patient list.

One hundred and four families were identified as eligible for the study. They were initially contacted by post using an introductory letter and information sheets (parent and well sibling versions) to explain the study (see Appendix 1). This was followed by a telephone call, when further study details were given and any questions answered. On the bottom of the introductory letter there was a reply slip which families could choose to send back indicating their interest in participating or not.

Table 1
Demographic and Illness Characteristics of the Sample

Characteristics	Per cent (Number)	
<i>Families (n=62)</i>		
Ethnic Origin		
English	80.6%	(n=53)
Irish	9.7%	(n=6)
Jewish	1.6 %	(n=1)
Persian	1.6 %	(n=1)
Brazilian	1.6 %	(n=1)
Number of Children in Family		
2	41.9%	(n=26)
3	35.5%	(n=22)
4+	22.6%	(n=14)
Socio-Economic Class		
	This Sample	National Sample (1981 Census)
Non-Manual	64.5% (n=40)	46.9%
Manual	27.4% (n=17)	48.2 %
Other (retired, disabled, unemployed)	8.1% (n=5)	4.7%
Marital Status of Mother		
Married	87.1% (n=54)	
Single Parent	12.9% (n=8)	
Recruited from:		
Hospital 1:	38.7% (n=24)	
Hospital 2:	8.1 % (n=5)	
Hospital 3:	14.5 % (n=9)	
Hospital 4:	12.9% (n=8)	
Hospital 5:	25.8 % (n=16)	
<i>Well Sibling Participants (n=62)</i>		
Gender		
Male	53.2 %	(n=33)
Female	46.8 %	(n=29)
Age of Well Sibling compared to Ill Child		
Older	59.7%	(n=37)
Younger	38.7%	(n=24)
Twin	1.6 %	(n=1)
Received Clinical Psychology Input?		
Yes	14.5 %	(n=9)
No	85.5 %	(n=53)
Age (years)		
Mean (Standard deviation), Range 12.29 (2.55), 8-16		

Table 1 continued
Demographic and Illness Characteristics

Characteristics		Per cent (Number)	
<i>Ill Children (n=62)</i>			
Gender			
	Male	67.8 %	(n=42)
	Female	32.2 %	(n=20)
Age (years)			
	Mean (Standard deviation), Range	11.13 (3.99),	2-18
Diagnosis			
	Cancer	35.5%	(n=22)
	Spina Bifida	25.8%	(n=16)
	Other Condition	38.7%	(n=24)
	Muscular Dystrophy	8.0 %	(n=5)
	Cystic Fibrosis	6.5 %	(n=4)
	Velo-Cardio-Facial Syndrome	6.5 %	(n=4)
	Severe Diabetes	4.8 %	(n=3)
	Severe Congenital Abnormalities	3.3 %	(n=2)
	Severe Epilepsy	1.6 %	(n=1)
	Retts Syndrome	1.6 %	(n=1)
	Severe Crohn's Disease	1.6 %	(n=1)
	Severe Brittle Bone Disorder	1.6 %	(n=1)
	Severe Multiple Extosis	1.6 %	(n=1)
	Non-malignant Brain Tumour	1.6 %	(n=1)
Prognosis			
	Cancer	Unknown	35.3 % (n=22)
		Fatal	0% (n=0)
		No Change Expected	0% (n=0)
		Degenerative, Not Fatal	0% (n=0)
		Improvement Expected	0% (n=0)
	Spina Bifida	Unknown	6.5 % (n=4)
		Fatal	1.6% (n=1)
		No Change Expected	8.1% (n=5)
		Degenerative, not Fatal	8.1% (n=5)
		Improvement Expected	1.6% (n=1)
	Other Condition	Unknown	8.1% (n=5)
		Fatal	16.1% (n=10)
		No Change Expected	6.5 % (n=4)
		Degenerative, not Fatal	1.6% (n=1)
		Improvement Expected	6.5 % (n=4)
Acutely ill in last six months			
	Cancer	Yes	33.9% (n=21)
		No	1.6% (n=1)
	Spina Bifida	Yes	8.1 % (n=5)
		No	17.7 (n=11)
	Other Condition	Yes	25.8 % (n=16)
		No	12.9% (n=8)
Duration of Illness (years)			
		Mean	(Standard deviation)
	Cancer	1.30	(1.65).
	Spina Bifida	14.80	(4.18)
	Other Condition	6.97	(6.07)
			Range
	Cancer		0.25 - 7
	Spina Bifida		9 - 18
	Other Condition		0.6 - 15

Sixty-two families (59.6 per cent of the sample) agreed to participate in the study. Twenty-five families (24 per cent) could not be contacted by telephone and did not return the reply slip. Table 2 shows that a further 17 families (16.1 per cent) declined to participate for various reasons.

Table 2
Reasons for Non-Participation

Reasons for Non-Participation	Per cent	(Number)
Parents did not wish to participate	5.8	(6)
Well sibling did not wish to participate	3.8	(4)
Sick child became critically ill	3.8	(4)
Parent became seriously unwell	0.9	(1)
Family already involved in similar research	0.9	(1)
Recent family bereavement	0.9	(1)

2.5 Ethical Considerations

Ethical approval for the study was obtained from each hospital (see Appendix 2 for example letter of approval). Throughout recruitment and interviewing, the researcher endeavoured to provide full information about the purpose and content of the study to all participants. Families were informed that an overview of the main results of the study would be available to them, if they so wished.

Written consent was obtained from mothers and well siblings (see Appendix 3). The parent consent forms additionally asked them to consent for their healthy children. Both forms asked for participants to verify that the study had been explained to them. The completed consent forms were filed in the ill child's medical notes.

The information sheets and consent forms emphasised that the families' decision to participate, not to participate or to terminate the interviews would not affect the ill child's current or future treatment. They also stated that all information given was confidential and would only be available for the purpose of the study. However, families were informed prior to the interview that if the researcher felt that parents should be made aware of issues discussed by the well siblings they would be informed. An example issue might be the well sibling feeling responsible for causing the illness.

Prior to beginning and after the interviews, the researcher met with the whole family to provide an opportunity for any member to ask questions, express concerns about participating and finally to discuss how the interviews went. Parents were also given the opportunity to review the one questionnaire intended for well siblings. One hospital's ethics committee advised that all well siblings should be interviewed to avoid any feelings of exclusion, although data was then only analysed from the target sibling.

The name and telephone number of a staff member in the ill child's hospital clinical team was available to families. This gave families the opportunity to request advice or further support on any issues that arose during the interview. One ethical problem was that the study was not intended to directly help participating families, but families treated in the future.

2.6 Measures

The variables in the proposed well sibling adjustment model (refer to Figure 2, Chapter 1, p.30) and their associated questionnaires are summarised in Table 3. This section will review these variables and their associated measures and the demographic and illness variables.

Table 3
Variables in Well Sibling Adjustment Model and Associated Measures

Variable Type	Variable Name	Measure
Independent	Parental Emotional Symptoms	Malaise Inventory (Rutter, Tizard & Whitmore, 1970)
Independent	Parental Satisfaction with Social Support (Community, Friendship, Intimate Relationship)	Inventory of Parent Experience (Crnic & Greenberg, 1983)
Independent	Parental Report of Burden of Care of the Illness	Clinician's Overall Burden Index (Stein & Jessop, 1982)
Mediating	Well Sibling-Parent Dyad Agreement on Siblings' Cognitions of the Illness	Level of Statistical Agreement between Well Sibling and Parent Responses on Sibling Perception Questionnaire (Carpenter & Sahler, 1991).
Dependent	Sibling Adjustment	The Strengths and Difficulties Questionnaire (Goodman, 1997)

2.6.1 Demographic and Illness Variables

In accordance with previous literature (Carpenter & Sahler, 1991; Sloper & While, 1996; Stallard et al, 1997), information on a number of demographic and illness variables was collected from parents.

The demographic variables were socio-economic class classified according to the Standard Occupational Classifications (Office of Population Censuses and Surveys, 1991); ethnic origin; family composition (single versus two parent family; number of children in family); well siblings' gender, age, position in family relative to the ill child, Clinical Psychology input; ill children's gender and age; and hospital recruited from.

The illness variables were diagnosis; prognosis; whether the children had been acutely ill in the six months preceding the interview; and duration of the condition.

2.6.2 Parental Psycho-social Resources

2.6.2.1 Parental Emotional Symptoms

Parental emotional symptoms were assessed using the widely used Malaise Inventory (Rutter, Tizard & Whitmore, 1970) (see Appendix 4). This is a 24-item scale with a Yes-No response format. Example items are "Are you easily upset or irritated?" and "Do you often feel miserable or depressed?". The inventory was adapted from the Cornell Medical Index and was designed to assess psycho-somatic symptoms associated with emotional disorders. A higher score indicates greater distress, and scores of eight or more have been widely used to identify those at high risk of depression (Cheung & Buchanan, 1997).

The Malaise Inventory is a well established scale with extensive reliability and validity data (Grant, Nolan & Ellis, 1990; McGee, Williams & Silva, 1986). Rutter and colleagues found the test-retest reliability of the scale to be good (correlation coefficient=0.91) with a sample of 35 mothers of children with physical disabilities.

They reported that the scale had moderate criterion validity in differentiating between mothers with and without a psychiatric diagnosis made by a clinician.

2.6.2.2 Parents' Satisfaction with Social Support

Parents' satisfaction with social support was measured using the Social Support Scale from the Inventory of Parent Experiences (Crnic & Greenberg, 1983) (see Appendix 4). This scale consists of 16 items, and assesses quantity of and satisfaction with support from three areas: community, friendships and an intimate relationship (if applicable). Each satisfaction item has a four point Likert scale, marked as "very dissatisfied (I wish things were very different)", "somewhat dissatisfied (I would like some changes)", "somewhat satisfied (OK for now, pretty good)", and "very satisfied (I'm really pleased)". The total satisfaction scores with community and friendship support are the sum of the satisfaction sub-scale items (scores ranges 0 to 8 and 0 to 16, respectively); the total satisfaction score with an intimate relationship includes indices of presence and satisfaction with the support source (score range 0 to 16). A higher score on each sub-scale indicates greater satisfaction.

The Social Support Scale has been used in a recently published study focusing on the adjustment of family members caring for children with serious illness or disability (Sloper & While, 1996). It has good face validity, and unlike many scales it conceptualises social support as a multi-dimensional concept (Bowling, 1997). Crnic and Greenberg report that the satisfaction sub-scales' test-retest reliabilities over 18 months are as follows: community (correlation coefficient = 0.53), friendship (correlation coefficient = 0.64) and intimate relationship (correlation coefficient =

0.82). They report internal consistency values of: community (Cronbach's alpha coefficient = 0.50), friendship (Cronbach's alpha coefficient = 0.65) and intimate relationship (Cronbach's alpha coefficient = 0.69).

2.6.2.3 Parental report of burden of care of the illness

Burden of care was assessed using the Clinician's Overall Burden of Care Index (COBI) (Stein & Jessop, 1982) (see Appendix 4). The scale is based upon an assumption that caring for an ill child is burdensome, irrespective of individual families' coping abilities. The authors propose that the scale reflects five generic features of caring for an ill child: disruption to the family routine (e.g. hospital visits), nursing tasks that families need to perform (e.g. stethoscope use), fixed deficits in a child requiring compensatory parental behaviour (e.g. limited mobility and wheel-chair use), dependency of a child who cannot perform age-appropriate activities of daily living (e.g. toileting), and the psychological burden inherent in the child's prognosis.

The COBI consists of 44 weighted items, compiled through a literature review and interviews with clinicians providing care for chronically ill children. The items were weighted by 104 health care professionals assessing the relative burden imposed by each item. The results indicated that items concerning major deficits (e.g. deafness) and those entailing considerable parental inconvenience (e.g. frequently leaving work for hospital appointments) were rated as most burdensome. Nursing tasks were seen as less burdensome.

An example item, with its associated weights, is as follows: “Do clinic visits or treatments require: a) rearrangement of parents’ work schedule ? (12), b) special baby-sitting ? (9) and /or c) frequent absences from work or school ? (17)”

The scale is completed by clinicians or researchers on the basis of parental responses, and the response format for each item is dichotomous (yes/no). Total scores are obtained by summing the derived weights of individual items (score range 0 to 879), with a higher score indicating a higher burden of care on the family. For example, Stein and Jessop report that in their test sample of 219 chronically ill children, a child with quadriplegia and a tracheostomy received a score of 276 in contrast to a child with stable diabetes who received a score of 114.

The internal consistency of the scale is high (Cronbach’s alpha coefficient = 0.70) (Stein & Jessop, 1982). Its concurrent validity, when correlated with parents’ subjective overall rating of the burden of care of the illness, was low (correlation coefficient = 0.52).

2.6.3 Well Sibling-Parent Dyad Agreement on Well Siblings’ Cognitions of the Illness and its Impact

The level of agreement between well siblings and parents was operationally defined as the statistical agreement between siblings’ and mothers’ responses on the SPQ (Carpenter & Sahler, 1991) (see Appendix 4 (parent version of SPQ) and Appendix 5 (well sibling version of SPQ)). Please refer to section 2.7 (Plan of Data Analysis) for a more comprehensive definition of well sibling-parent dyad agreement.

The SPQ was originally designed to assess specific attitudinal issues relevant to well siblings of children with cancer, aged between six and eighteen years of age. It has 23 items which address several dimensions of the illness experience, including interpersonal interactions (e.g. "I wish my parents would spend less time with my brother /sister"); how siblings perceive the illness to affect themselves (e.g. "I feel sad about my brother's /sister's cancer"); fear of the condition (e.g. "I worry that I can catch cancer from my brother /sister"); and communication about the illness (e.g. "I can talk to my parents about my brother's /sister's cancer"). The items were derived from themes suggested by the empirical literature, and in consultation with the siblings of children with cancer and paediatric clinicians. The scale has a five-point Likert-type response format, ranging from "never" to "always". Higher scores on each item and the total scale indicate more negative perceptions.

Several changes were made to the basic format of the questionnaire (both the well sibling and parent version). First, the response format was changed to a categorical yes /no choice, as a result of participants in the pilot study reporting it was easier to make a simple distinction as to whether siblings had had that cognition or not, rather than grade how often the sibling had had it. Second, the word "cancer" on the original questionnaire was changed to "illness" as the sample included well siblings of children with a range of conditions. This latter change was also made in two recently published papers (Sloper & While, 1996; Stallard et al, 1997). Stallard et al (1997) also used the questionnaire to assess well siblings of children with longer term conditions, such as muscular dystrophy.

For the purpose of this study, a parent version of the SPQ was developed. This aimed to enable parents to reflect on whether their healthy child had experienced any of the negative cognitions listed on the SPQ. The parent version of the scale was exactly the same as the sibling version, with the exception of differences in pronouns.

The psychometric properties of the new scales were assessed by their internal consistency and their test-retest reliability. The internal consistency was good for both scales: well sibling scale (Cronbach's alpha coefficient = 0.76) and parent scale (Cronbach's alpha coefficient = 0.74). Sixteen families were re-contacted by telephone a week after the interview to re-administer the well sibling and parent versions of the SPQ. Five of these families had a child with cancer, five had a child with spina bifida, and six had a child with another condition. The test-retest reliabilities of the scales' total scores was high: well sibling scale (correlation coefficient = 0.88) and parent scale (correlation coefficient = 0.89). This compared favourably with Rende and Plomin (1991) report of test-retest reliability over one week ($r=0.81$) on children's perceptions of the stressfulness of a range of trivial and life changing events.

2.6.4. Well Sibling Adjustment

Well sibling adjustment was assessed using the Strengths and Difficulties Questionnaire (SDQ) (Goodman, 1997) (see Appendix 4). This scale is applicable for children aged four to sixteen years, and is completed by a parent and /or a teacher. In this study, it was administered to parents only. Informants are asked to answer on the basis of the child's behaviour over the last six months.

The SDQ has 25 items, based upon the items included in Rutter's (1967) adjustment scale, the Children's Behaviour Questionnaire. However, the SDQ additionally includes items which would generally be thought of as strengths, for example "considerate of other people's feelings". It also includes items of more contemporary interest, such as feeling bullied within peer relationships and hyperactivity symptoms (e.g. inattention and impulsivity) (Goodman, 1997). Each item is graded on a three point Likert scale, marked "not true", "somewhat true" and "certainly true".

The SDQ has a Total Deviance scale (score range 0 to 40) and five sub-scales (score range 0 to 10) assessing different aspects of adjustment: emotional symptoms, conduct problems, peer problems, pro-social behaviour and hyperactivity. The Total Deviance score is operationally defined as the sum of the following four sub-scale scores: Peer Problems, Hyperactivity, Emotional Symptoms and Conduct Problems. A higher score indicates greater disturbance, with the exception of the Pro-Social Behaviours sub-scale which is scored in the opposite direction.

The data collected are continuous, but can be categorised into normal, borderline and abnormal bands using standardised norms (see Appendix 4). These were developed using UK psychiatric and dental samples of children; 80 per cent of children in the community fall in the normal range of scores, 10 per cent in the borderline range and another 10 per cent in the abnormal range (Goodman, 1997).

The author assessed the psychometric properties of the SDQ by investigating its concurrent validity with the Rutter questionnaire. The latter has well established

validity and reliability (Elander & Rutter, 1996). Goodman presents high values of inter-measure correlations between the two scales: Total Deviance scale (0.88), Conduct Problems sub-scale (0.88), emotional symptoms sub-scale (0.78) and Hyperactivity sub-scale (0.82). The concurrent validity of the Peer Problems and Pro-Social sub-scales could not be assessed due to them having no Rutter counterparts.

2.7 Plan of Data Analysis

The Results chapter starts with descriptive and exploratory data analyses of the variables in the well sibling adjustment model (refer to Figure 2), and a computation of well sibling-parent dyad agreement. The research questions are then addressed using percentage agreement calculations and multiple regression techniques. The service-related questions were assessed using simple percentages.

The descriptive data analysis section provides a summary of the mean scores, standard deviations and possible range of scores for each of the variables. The well sibling adjustment data were compared with standardised norms. There was also an exploration of statistical associations between the model's variables with the demographic and illness variables (refer to Table 1). Relationships between variables were explored using chi-squared analyses, independent sample t-tests or analysis of variance and product-moment correlations. This exploration did not control for inflated Type 1 error rates, but retained higher statistical power.

Two types of agreement were calculated: agreement on specific SPQ items across the whole sample and agreement within well sibling-parent dyads across all items on the SPQ.

Agreement on specific SPQ items was assessed using percentage agreements across the sample. These have previously been reported in similar research, and therefore can be compared across studies. However, the reader is cautioned that percentage agreements do not control for agreement by chance, which was high with the SPQ's binary response format.

Agreement within well sibling-parent dyads across all SPQ items was assessed using correlation coefficients, which can be entered as data in further statistical analyses (Howell, 1992). This was necessary to identify correlates of well sibling-parent dyad agreement. Phi correlation coefficients were chosen because the SPQ has a binary response format. Each coefficient was transformed into a Fisher Z coefficient, to ensure homogeneity of variance (Howell, 1992).

In order to investigate whether any potential relationships between well sibling-parent agreement scores and siblings' adjustment scores were confounded by each being dependent upon parental report and the two scales sharing some similar items, a hierarchical multiple regression analysis was carried out.

A path diagram of the hypothesised relationship between parental psycho-social variables, well sibling-parent dyad agreement and well sibling adjustment was derived

(Kenny, 1979). Multiple regression analyses were carried out to identify variables which predicted variance in the sibling adjustment scores (Total Deviance scale and sub-scales) and well sibling-parent agreement scores. A revised well sibling adjustment model was proposed, and a discriminant analysis used to assess the predictive ability of this new model.

RESULTS

3.1 Description and Exploration of Variables in Sibling Adjustment Model

This section describes the dependent variable (well sibling adjustment), the independent variables (parental psycho-social variables), and finally computes the mediating variable (well sibling-parent dyad agreement) from the hypothesised Well Sibling Adjustment Model (Figure 2, Introduction chapter, p.30). Bi-variate relationships between these variables and with illness and demographic variables are also explored.

3.1.1 Well Sibling Adjustment

Well sibling adjustment was assessed using the Strengths and Difficulties Questionnaire (SDQ) Total Deviance scale, four sub-scales measuring different problem areas and one sub-scale assessing level of pro-social behaviours. This latter sub-scale was scored in the opposite direction to the other sub-scales.

The distributions of all the SDQ scores were normal. From Table 4, it can be seen that the mean Total Deviance score was 9.60 (standard deviation (SD)=6.32). The sub-scales assessing problem behaviours which yielded the highest mean score was Hyperactivity (mean 3.40 (SD=2.80)), and the lowest mean score was Peer Problems (mean 1.40 (SD=1.69)).

Table 4
Well Sibling Adjustment Data

Strengths and Difficulties Questionnaire	Mean Score	SD	Range of Scores (Maximum Range)
Total Deviance Score	9.60	6.32	0 - 24 (0 - 40)
Sub-Scale Scores:			
Peer Problems	1.40	1.69	0 - 6 (0 - 10)
Conduct Problems	1.77	1.97	0 - 8 (0 - 10)
Hyperactivity	3.40	2.80	0 - 10 (0 - 10)
Emotional Symptoms	2.92	2.52	0 - 10 (0 - 10)
Pro-Social Behaviours	8.48	1.53	5 - 10 (0 - 10)

3.1.2 Parental Psycho-Social Variables

Table 5 illustrates the data collected for each of the parental psycho-social variables. The data distribution for each variable resembled a normal curve, with the exception of data derived from the Satisfaction with Intimate Relationships sub-scale. The distribution of data was skewed towards the higher range of scores, with 42.3 per cent (n=33) of mothers reporting maximum satisfaction with an intimate relationship. This finding was not supported by many mothers who spoke about the strain their relationships with partners had been under. Additionally, 16 mothers (20.5 per cent of sample) either refused to answer this sub-scale's questions or did not have an intimate relationship to appraise.

There was a wide range of Malaise and Burden of Care scores. Thirty-nine per cent of mothers (n=22) scored eight or more on the Malaise Inventory, which indicated that they were at risk of depression. The lowest burden of care was for a child with cancer (COBI score = 94) and the highest for a child with severe spina bifida and hydrocephalus (COBI score = 431).

Many mothers spontaneously talked about the high practical and psychological burdens of care required by the illness and the helpfulness of social support. For example, one mother of a child with cancer, remarked “When I’m not at the hospital, which these days isn’t often, I feel drained of energy and can’t get D.’s [the patient] treatment and checks off my mind. If it wasn’t for having normal conversations with friends about other things, I don’t know what I’d do.

Table 5:
Parental Psycho-social Resources Data

Measure	Mean Score	SD	Range of Scores (Maximum Range)
Malaise Inventory			
Parents’ Emotional Symptoms	6.48	4.66	0 - 18 (0 - 24)
Inventory of Parent Experiences			
Satisfaction with Community Support	6.13	1.47	2 - 8 (0 - 8)
Satisfaction with Friendship Support	13.18	2.45	5 - 16 (0 - 16)
Satisfaction with Intimate Relationship	14.49	2.17	7 - 16 (0 - 16)
Burden of Care Index	230.97	75.39	94 - 431 (0 - 879)

3.1.3 Agreement in Well Sibling-Parent Dyads

Across the sample, mothers’ and well siblings’ total scores on the SPQ were normally distributed. As Table 6 shows, mothers rated well siblings as having significantly more negative cognitions about the illness and its impact, than reported by the siblings themselves ($t(59)=2.90, p<0.01$).

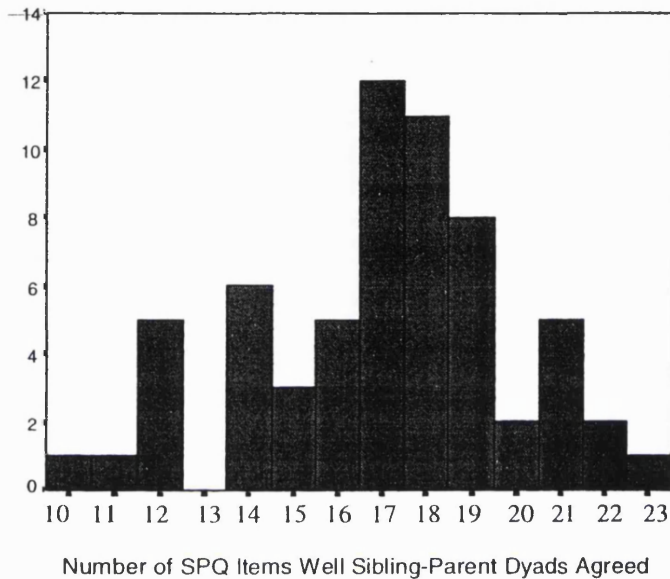
Table 6:
Total Scores on Sibling Perception Questionnaire

Sibling Perception Questionnaire	Mean	SD	Range of Scores (maximum range: 0 to 23)
Parent Total Score	10.15	3.87	1-19
Well Sibling Total Score	8.71	3.65	2-18

Total scores on the SPQ do not represent the number of items well sibling-parent dyad agreed or disagreed on. The mean well sibling-parent dyad percentage agreement was 73.99 (SD=12.51), which was equivalent to a mean agreement on 17 items on the SPQ. As illustrated in Figure 3, percentage agreement ranged between 43.40 (i.e. agreement on 10 SPQ items) and 100.00 per cent (i.e. agreement on 23 SPQ items).

As explained in the Method chapter, a correlation coefficient was calculated for each dyad and converted into a Fisher Z score. The mean value of Fisher Z for this sample was 0.56 (SD=0.31, range -0.04 to 1.54). Unfortunately, the Fisher Z transformation converts correlation coefficients equalling 0 or 1 into infinite values. In practice, this meant that the data from one well sibling-parent dyad who had agreed on every SDQ item, was excluded from further data analysis.

Figure 3
Frequency of Well Sibling-Parent Dyads Agreeing on Sibling Perception Questionnaire
Items



3.1.4 Statistical Associations within Well Sibling Adjustment Model

A series of bi-variate correlations was performed between each independent, mediating and dependent variable, as shown in Table 7. A hierarchical multiple regression analysis was also carried out to assess whether any potential relationship between well sibling-parent agreement scores and the overall adjustment scores was confounded by their dependence upon parental report.

The following associations should be interpreted with caution since the exploration did not control for inflated Type 1 error rates, but retained the advantage of higher statistical power. As was predicted in the well sibling adjustment model, there were significant correlations between:

- overall well sibling adjustment (Total Deviance score) and (1) mothers' report of emotional symptoms ($r=0.33$, $p<0.01$), (2) mothers' satisfaction with friendship support ($r=-0.31$, $p<0.05$), (3) burden of care of the illness ($r=0.26$, $p<0.05$), and (4) well sibling-parent dyad agreement ($r=-0.53$, $p<0.01$). The Total Deviance score was not significantly correlated with maternal satisfaction with support from the community ($r=-0.22$, $p=0.08$), and an intimate relationship ($r=-0.09$, $p=0.50$);
- sibling-parent dyad agreement and (1) mothers' report of emotional symptoms ($r=0.33$, $p<0.01$), (2) mothers' satisfaction with community support ($r=0.30$, $p<0.05$) and satisfaction with friendship support ($r=0.31$, $p<0.05$), (3) overall well sibling adjustment (Total Deviance score) ($r=-0.53$, $p<0.01$) and all sub-scale scores. Sibling-parent dyad agreement was not significantly correlated with maternal satisfaction with support from an intimate relationship ($r=0.04$, $p=0.94$) and the burden of care of the illness ($r=0.02$, $p=0.97$);
- burden of care of the illness and (1) overall well sibling adjustment (Total Deviance score) ($r=0.26$, $p<0.05$) and (2) well sibling emotional problems (SDQ sub-scale) ($r=0.45$, $p<0.01$).

Mothers' report of emotional symptoms was also significantly correlated with (1) maternal satisfaction with community support ($r=0.38$, $p<0.01$) and (2) maternal satisfaction with friendship support ($r=0.28$, $p<0.05$).

Table 7

Correlations between Variables in the Sibling Adjustment Model

Scale	1	2	3	4	5	6	7	8	9	10	11	12
1. Maternal Report of Emotional Symptoms		-0.38**	-0.27*	-0.24	0.20	-0.29*	0.33**	0.14	0.36**	-0.00	0.49**	-0.06
2. Satisfaction with Community Support			0.50**	0.09	-0.12	0.30*	-0.22	-0.04	-0.17	-0.05	-0.42**	0.27*
3. Satisfaction with Friendship Support				0.28*	-0.28*	0.31*	-0.25*	-0.09	-0.23	-0.09	-0.33**	0.12
4. Satisfaction with Intimate Relationship					0.08	0.04	-0.09	0.04	-0.23	0.08	-0.15	0.03
5. Burden of Care Index						0.02	0.26*	0.15	0.45**	0.01	0.07	0.15
6. Well Sibling-Parent Dyad Agreement Score							-0.53**	-0.30*	-0.41**	-0.38**	-0.35**	0.37**
7. SDQ Total Deviance score								0.78**	0.70**	0.77**	0.43**	-0.30*
8. Conduct Difficulties Sub-scale									0.33**	0.64**	0.14	-0.30*
9. Emotional Symptoms Sub-scale										0.27*	0.25*	0.00
10. Hyperactivity Sub-scale											0.04	-0.23
11. Peer Problems Sub-Scale												-0.32*
12. Pro-social Sub-scale												

* p<0.05; **p<0.01

The hierarchical multiple regression analysis showed that the amount of variance predicted in the overall adjustment score (Total Deviance score) increased significantly from 19.3 per cent to 34.8 per cent ($F(1,57)=14.09$, $p<0.001$), when sibling-parent agreement was entered, after total parent and total sibling SPQ scores. This result implied that the agreement index scores were independent of the overall adjustment scores.

The amount of variance predicted in the SDQ sub-scale data also significantly increased when sibling-parent agreement was entered after total parent and sibling SPQ scores at the one per cent or five per cent levels, with the exception of the Conduct Difficulties sub-scale. The overall results of the hierarchical regression, when Conduct Difficulties was the dependent variable, were not statistically significant ($F(3,57)=2.20$, $p=0.10$). The percentage variance increase ranged between 5.3 per cent for the Conduct sub-scale and 11.9 per cent for the Hyperactivity sub-scale.

3.1.5 Statistical Associations with Demographic and Illness Variables

The independent, mediating and dependent variables were largely not statistically associated with demographic or illness variables. No significant associations were found between the demographic or illness variables and well sibling-parent dyad agreement, mothers' emotional state, mothers' satisfaction with intimate and community support and burden of care of the illness. Again, the reader should be cautious in interpreting the associations stated since the exploration did not control for inflated Type 1 error rates.

As the sample covered a wide age range, an initial analysis was undertaken to examine relationships between well sibling age (8 to 11 years classified as 'younger' (n=27), and 12 to 16 years classified as 'older' (n=35)) and variables within the model. It was proposed that these age groups would be more homogenous in terms of developmental stage and associated non-illness life stressors. The older age group scored significantly higher on the Peer Problems sub-scale of the SDQ ($t(60)=2.62$, $p<0.05$), and younger siblings were significantly more likely to have mothers who reported more satisfaction with support given by friends ($t(60)=2.09$, $p<0.05$). There was not a significant difference in mean age between male and female well siblings.

Three other demographic factors showed significant associations with well sibling adjustment: well siblings who scored higher on the Pro-Social scale (i.e. displaying more pro-social behaviours) were more likely to be female ($t(54)=2.89$, $p<0.01$) and younger ($r=-0.31$, $p<0.05$), well siblings who scored higher on the Conduct Difficulties sub-scale (i.e. displaying more conduct difficulties) were more likely to be from manual social class backgrounds ($F(3,58)=5.24$, $df=2$, $p<0.01$) and well siblings who scored higher on the Emotional Symptoms sub-scale (i.e. displaying more emotional symptoms) were more likely to have received Clinical Psychology input ($t(60)=2.66$, $p<0.01$).

The sample included a broad range of diseases. These were categorised into three distinct groups: cancer (n=22; 35.5 per cent of the sample), spina bifida (n=16; 25.84 per cent), and 'other diagnoses' (n=23; 37.1 per cent). An exploratory analysis was

undertaken to examine relationships between illness characteristics and variables within the model.

There were no significant relationships between well siblings' SDQ Total Deviance scale (or sub-scale) scores or well sibling-parent dyad agreement and the diagnosis, prognosis, duration and current acuteness of illness. Therefore the total sample was used for the remaining analysis. However, well siblings in both the spina bifida group and in the 'other diagnoses' group were significantly older than siblings in the cancer group ($\chi^2=10.25$, $df=2$, $p<0.01$). As would be expected, the disease groups varied significantly in terms of prognosis ($\chi^2=47.16$, $p<0.001$, $df=8$), duration ($t(60)=-3.06$, $p<0.01$) and current acuteness of illness ($\chi^2=19.196$, $df=2$, $p<0.001$).

As summarised in Table 1 (Method chapter), the prognosis of cancer was uncertain in every case, the prognosis of spina bifida was no change or degenerative (not fatal) in most cases ($n=10$), and the prognosis of the "other condition" group was most often fatal ($n=10$). The mean duration of the illness in each group varied between 1.3 years for cancer, to 6.97 years for "other conditions" and 14.80 years for the spina bifida group. Most ill children in the cancer and "other condition" groups had been acutely ill in the last six months ($n=21$ and $n=16$, respectively). In contrast, most ill children in the spina bifida group had not been ill in the last six months ($n=11$).

3.2 Research Question 1:

What is the adjustment status of well siblings, in comparison to standardised norms?

As can be seen from Table 8, 17 of the 62 well siblings (27.4 per cent) had Total Deviance scores which fell in the borderline or abnormal ranges of the SDQ. This did not differ significantly from the SDQ norms ($\chi^2=2.86$, $df=2$, $p=0.24$). Most of these siblings showed mixed problems: 14 had emotional difficulties in the borderline or abnormal ranges, 12 had conduct difficulties in the borderline or abnormal ranges, 11 had hyperactivity difficulties in the borderline or abnormal ranges, six had peer difficulties in the borderline or abnormal ranges and one was displaying pro-social behaviours in the abnormal range.

The distribution of well siblings' scores on three of the five sub-scales were significantly different to the SDQ norms. Well siblings were more likely to score in the borderline or abnormal ranges of the Emotional Symptoms sub-scale ($\chi^2=17.86$, $df=2$, $p<0.001$) and the Conduct Problems sub-scale ($\chi^2=10.98$, $df=2$, $p<0.01$). Conversely, well siblings were also more likely to score in the normal range of the Pro-Social Behaviours sub-scale ($\chi^2=6.25$, $df=2$, $p<0.05$). The Peer Problems and Hyperactivity sub-scales' distribution of scores did not differ significantly from population norms.

Qualitative information given by mothers and well siblings suggested other aspects of well siblings' adjustment were present that were not assessed by the SDQ. Five mothers reported that the well sibling had developed physical symptoms similar to the

ill child (e.g. lumps in groin) or symptoms which may have been induced by stress (e.g. asthma).

Table 8:
Percentage (and Number) of Well Siblings Scoring in Normal, Borderline, Abnormal Adjustment Categories

Strengths and Difficulties Questionnaire	Percentage (Number)		
	Normal	Borderline	Abnormal
Total Deviance Score	72.6% (45)	11.3% (7)	16.1% (10)
Sub-Scale Scores:			
Peer Problems	77.4% (48)	6.5% (4)	16.1% (10)
Conduct Problems	67.7% (42)	9.7% (6)	22.6% (14)
Hyperactivity	77.4% (48)	4.8% (3)	17.7% (11)
Emotional Symptoms	62.9% (39)	11.3% (7)	25.8% (16)
Pro-Social Behaviours	91.9% (57)	6.5% (4)	1.6% (1)

Many well siblings also reported feeling responsible for their ill brother or sister. For example, R, the 13 year old brother of a two year old boy with bowel and kidney problems said "I don't mind him [the patient] having more attention from Mum, I also want to care for him". Also, A., a 10 year old sister of a 13 year old girl with cancer said, "I stop people from hugging her [the patient], because I'm worried they'll pull the tubes out by mistake". Finally, D., a 12 year old sister of a 16 year old boy with spina bifida said "I try and think of lots of cures to make him normal".

Some well siblings' statements implied a sense of personal responsibility for causing the illness, for example six children reported that kicking or hitting their sick sibling had caused the illness. In three of these cases, this belief was previously unknown to their mothers.

3.3 Research Question 2:

What are the percentage agreements between parents and well siblings about well siblings' cognitions about the illness and its impact, across the whole sample?

As Table 9 illustrates there was considerable item-by-item fluctuation in well sibling-parent dyad percentage agreement across the sample. As discussed in the Method chapter, the reader should bear in mind that these percentages do not control for chance agreement.

The highest percentage agreements between well siblings and mothers were on items which asked directly about the illness or the ill child. For example, 93.5 per cent of sibling-parent pairs agreed that the siblings understood (or did not understand) why mothers had to spend more time with their ill brother or sister; 91.9 per cent of sibling-parent pairs agreed that the siblings worried (or did not worry) that they could catch the illness from their brother or sister; 90.3 per cent of sibling-parent pairs agreed that the siblings wished (or did not wish) there was something they could do about their brother's or sister's illness.

There were high percentage disagreements about well siblings' emotional feelings about the illness. For example, 51.6 per cent of siblings and mothers disagreed about siblings' feelings of anger about the illness, and 34.4 per cent of siblings and mothers disagreed as to whether the sibling was able to forget their brother or sister was ill. However, only 6.5 per cent of siblings and mothers disagreed as to whether the sibling felt sad about his brother or sister being ill.

Table 9

Well Sibling-Parent Percentage Disagreements on Negative Cognitions Experienced by the Well Sibling, Given in Percentage Disagreements per Item on the SPQ

Sibling Perception Questionnaire	Percentage of cases		
	Sibling reports negative cognition and parent does not	Parent reports negative cognition and sibling does not	Sibling and parent disagree
Well sibling can talk to parents about brother's /sister's illness	12.9	12.9	25.8
Well sibling can talk to friends about brother's /sister's illness	14.5	16.1	30.7
Well sibling can talk to parents about school work	9.7	6.5	16.1
Well sibling can talk to other adults about brother's /sister's illness	16.4	13.1	29.5
Well sibling worries about catching illness from brother /sister	4.8	3.1	8.1
Well sibling feels afraid of brother's/ sister's illness	14.5	17.7	32.2
Well sibling feels friends worry that they can catch the illness from brother/ sister	3.1	1.6	4.9
Well sibling wishes parents would spend less time with brother / sister	6.5	25.8	32.2
Well sibling wishes parents would spend more time with him /her	8.1	33.9	42.0
Well sibling feels people are more interested in brother /sister than him- /herself	3.1	21.0	24.2
Because of brother's /sister's illness, well sibling feels people don't care how s/he feels	8.2	26.2	34.5
Because of brother's /sister's illness, well sibling feels parents ignore him /her	0.0	17.7	17.7
Because of brother's /sister's illness, well sibling feels s/he has too much to do in the house	16.1	14.5	30.6
Well sibling doesn't want to bother parents with worries	21.0	25.8	46.8
Well sibling wishes s/he knew someone who understands how s/he feels	16.1	11.3	27.4
Because of brother's /sister's illness, well sibling feels the family does not do as much together as they might have done otherwise	8.1	22.6	30.4
Well sibling wishes there was something s/he could do about his/her brother's /sister's illness	3.1	6.5	9.7
Well sibling feels sad about brother /sister being ill	3.1	3.1	6.5
Well sibling thinks about brother's /sister's illness	3.1	11.3	14.5
Well sibling wonders why brother /sister got sick	14.5	16.1	30.7
Well sibling feels angry about brother /sister being ill	17.7	33.9	51.6
Well sibling understands why parents have to spend more time with ill brother /sister	1.6	4.8	6.5
Well sibling is able to forget that brother /sister is ill	21.3	13.1	34.4

There were also high percentage disagreements on issues associated with family functioning around the sibling. For example, 46.8 per cent of siblings and parent disagreed that the siblings did (or did not) want to bother their parents with their worries. As one mother of a 12 year old healthy girl and a boy (M.) with muscular dystrophy commented: "She writes stories about M. and what's going on at school - things she has stopped talking to us about". These results support the hypothesis of well siblings feeling a high degree of responsibility, here for themselves, and not wanting to bother their parents.

Additionally, 42 per cent of siblings and parent disagreed that the siblings wished (or did not wish) their parents would spend more time with them; 34.5 per cent of siblings and parent disagreed that the siblings felt people did (or did not) care about how they feel.

This level of disagreement on family interpersonal functioning was wholly supported by comments made by most well siblings interviewed, and also occasionally by mothers. For example, a 12 year old sister of a six year old with cancer stated "my parents now spend 80 per cent of the time with K. [the patient] and the other 20 per cent in bad vibes shouting at me". Her mother independently said that she felt her daughter "blames me for K.'s cancer because I smoke ... I wish she could talk to me more, instead she talks to her Grandmother". Additionally, a 15 year old sister of a boy with Crohn's Disease, said "The illness doesn't bother me because I know I can't do anything about it ... it's just how my parents have now started fussing over him that gets me ... when I say something to them about their fussing they don't know what I mean".

3.4 Research Question 3:

Do parent psycho-social variables affect well sibling adjustment directly, but also indirectly through well sibling-parent dyad agreement ?

Figure 4 illustrates a path diagram of the hypothesised relationship between parental psycho-social variables, well sibling-parent dyad agreement and well sibling adjustment. The arrows between variables indicate the theoretically proposed direction of causation, and carry the regression coefficients detailed later. The reader should bear in mind that some parent psycho-social variables were correlated with each other, but for the sake of clarity these relationships are not displayed on the diagram.

The pathway model was derived using regression procedures (Kenny 1979), from the following propositions:

- **Research Question 3a: Are well siblings better adjusted, if their parents report:**
 - less parental emotional disturbance ?**
 - higher satisfaction with community support ?**
 - higher satisfaction with friendship support ?**
 - higher satisfaction with an intimate relationship (if applicable) ?**
 - lower burden of care demanded by the illness?**
 - and there is higher sibling-parent dyad agreement ?**
- **Research Question 3b: Is there higher agreement between well siblings and parents about the siblings' illness cognitions, when parents report:**

less parental emotional disturbance?

higher satisfaction with community support ?

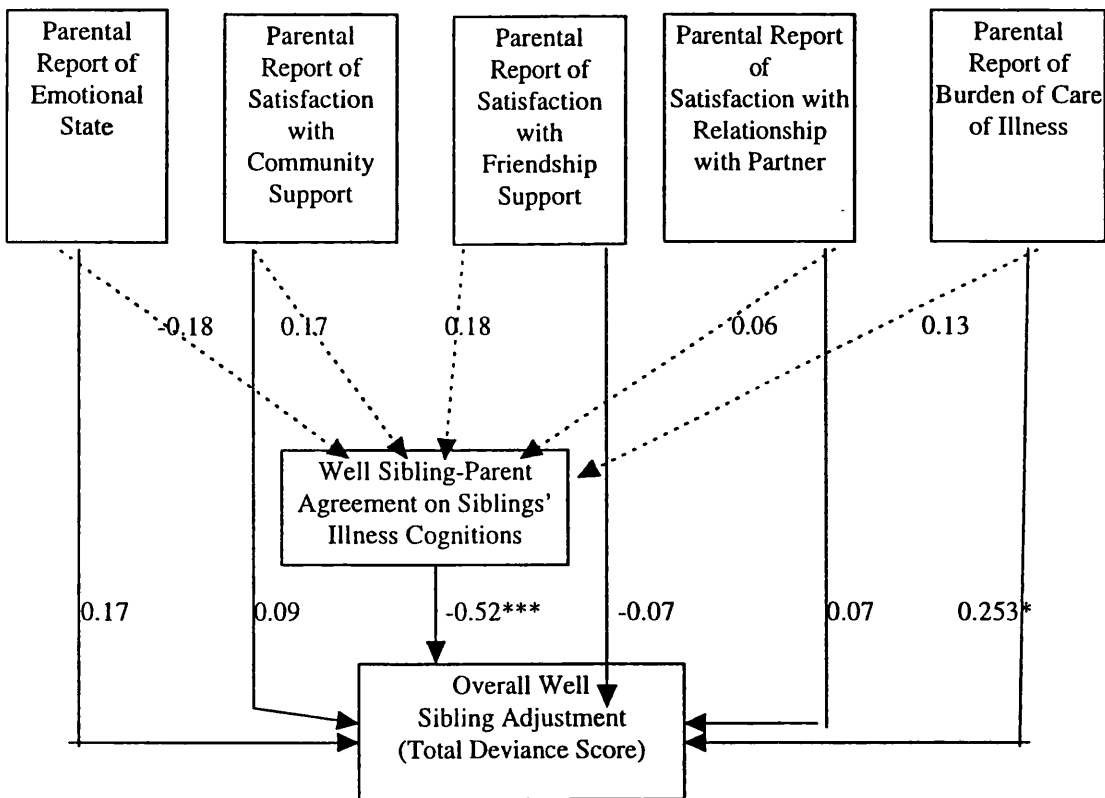
higher satisfaction with friendship support ?

higher satisfaction with an intimate relationship (if applicable) ?

and lower burden of care demanded by the illness ?

Figure 4

Path Diagram Modelling the Effects of Parent Psycho-Social Variables, Mediated by Well-Sibling Agreement, on Well Sibling Adjustment



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

3.4.1 Research Question 3a

To address question 3a, the parent psycho-social variables and well sibling-parent agreement were regressed onto the SDQ Total Deviance score and sub-scale scores, in multiple regression analyses. The standardised residuals in each multiple regression were normally distributed.

As Table 10 shows, the Beta weights indicated that burden of care and well sibling-parent agreement were significantly associated with overall well sibling adjustment (Total Deviance score). The model accounted for 37.8 per cent of the variance in the Total Deviance scores, and the overall regression was significant ($F(6,54)=5.47$, $p<0.001$). (The regression coefficients (or Beta values) of the parent psycho-social variables and well sibling-parent agreement are written on the arrows in the path diagram, illustrated in Figure 4).

To ensure that this finding was not result from multi-collinearity of the maternal variables of emotional state and satisfaction with social support, a hierarchical regression analysis was carried out. The satisfaction and emotional symptoms variables were entered, after well sibling-parent agreement and burden of care. The amount of variance predicted in the Total Deviance score did not increase significantly (from 35.7 per cent to 37.8 per cent ($F(1,57)=0.45$, $p>0.10$)), and it was therefore concluded the multiple regression finding was valid.

Table 10:
Multiple Regression Analysis for Well Siblings' Overall Adjustment Scores

Predictor Variable	Beta	t	p
<u>Parent Report of:</u>			
Emotional State	0.17	1.37	0.18
Satisfaction with Community Support	0.09	0.68	0.49
Satisfaction with Friendship Support	-0.07	-0.51	0.61
Satisfaction with Intimate Relationship	0.07	0.61	0.55
Burden of Care	0.25	2.23	0.03
Agreement Index (Fisher Z)	-0.52	-4.46	0.001

The parental psycho-social and agreement variables were then regressed on the SDQ sub-scale scores. As Table 11 illustrates, the predictor variables accounted for significant proportions of the variance in the Emotional Symptoms and Peer Problems sub-scales.

Table 11:
Multiple Regressions for Well Siblings' Adjustment Sub-Scale Scores

Dependent Variable	F (6,54)	p
Pro-social Behaviours	1.90	0.10
Emotional Symptoms	7.13	0.001
Hyperactivity Difficulties	1.82	0.11
Peer Problems	4.93	0.001
Conduct Problems	1.76	0.13

Burden of care and well sibling-parent agreement were significantly associated with well sibling emotional symptoms ($F(6,54)=7.13$, $p<0.001$). The model accounted for 44.2 per cent of the variance in the Emotional Symptoms sub-scale scores, and is displayed in Table 12.

Table 12:

Multiple Regression Analysis for Well Siblings' Emotional Symptoms Sub-Scale Scores

Predictor Variable	Beta	t	p
Parent Report of:			
Emotional State	0.17	1.49	0.15
Satisfaction with Community Support	0.11	0.87	0.39
Satisfaction with Friendship Support	0.02	0.17	0.86
Satisfaction with Intimate Relationship	-1.12	-1.10	0.28
Burden of Care	0.47	4.38	0.000
Agreement Index (Fisher Z)	-0.40	-3.62	0.001

As can be seen from Table 13, the only significant and independent variable associated with well sibling peer problems was maternal emotional symptoms ($F(6,54)=4.93$, $p<0.001$). The model accounted for 26.5 per cent of the variance in the Peer Problems sub-scale scores.

Table 13:

Multiple Regression for Well Siblings' Peer Problems Sub-scale Scores

Predictor Variable	Beta	t	p
Parent Report of:			
Emotional State	0.38	3.01	0.004
Satisfaction with Community Support	-0.13	-0.92	0.36
Satisfaction with Friendship Support	-0.15	-1.04	0.30
Satisfaction with Intimate Relationship	0.04	0.33	0.74
Burden of Care	-0.04	-0.36	0.72
Agreement Index (Fisher Z)	-0.19	-1.55	0.13

The quantitative findings that burden of care and well sibling-parent agreement were strong predictors of overall adjustment, were supported by many comments. For example, a mother of a well adjusted healthy child of eight years and a twelve year old child with muscular dystrophy, anticipated her child's degenerating health triggering off emotional symptoms in her daughter. She said: "J. [well sibling] and us talk very openly about all kinds of things about the illness ... in the future, we will have to spend so much time looking after B. [the patient] ... I imagine J. will be very sad about the illness and the lack of attention from us".

A similar comment was made by an 11 year old brother of a child with spina bifida and hydrocephalus. He felt that since starting a new school, he had found it difficult to cope with his parents' behaviours toward his ill sibling. He said: "I am angry and I sometime kick my door, when my parents help Z. [the patient] with her homework because she finds it hard...they don't know I find things hard too".

In a number of well adjusted families, mothers and well siblings alike spontaneously talked about the family's philosophy to coping with the illness, and by doing so demonstrated a high level of well sibling-parent agreement in attitudes towards the illness. As one 13 year old sister of a child with leukaemia said "Mum's told us to look after her [the patient] and be grateful that she's here with us". Several other families talked about the helpfulness of their religion in reducing any anxieties about feeling responsible for the illness.

3.4.2 Research Question 3b

To investigate which parental psycho-social variables were significantly associated with sibling-parent dyad agreement, a further multiple regression analysis was carried out. The model did not quite reach statistical significance ($F(5,54)=2.20$, $p=0.07$) and only accounted for 17 per cent of the variance in sibling-parent dyad agreement scores. From Table 14, it can be seen that the independent variables' regression coefficients were not statistically significant. (These regression coefficients are written on the arrows in the path diagram, illustrated in Figure 4). The standardised residuals were normally distributed.

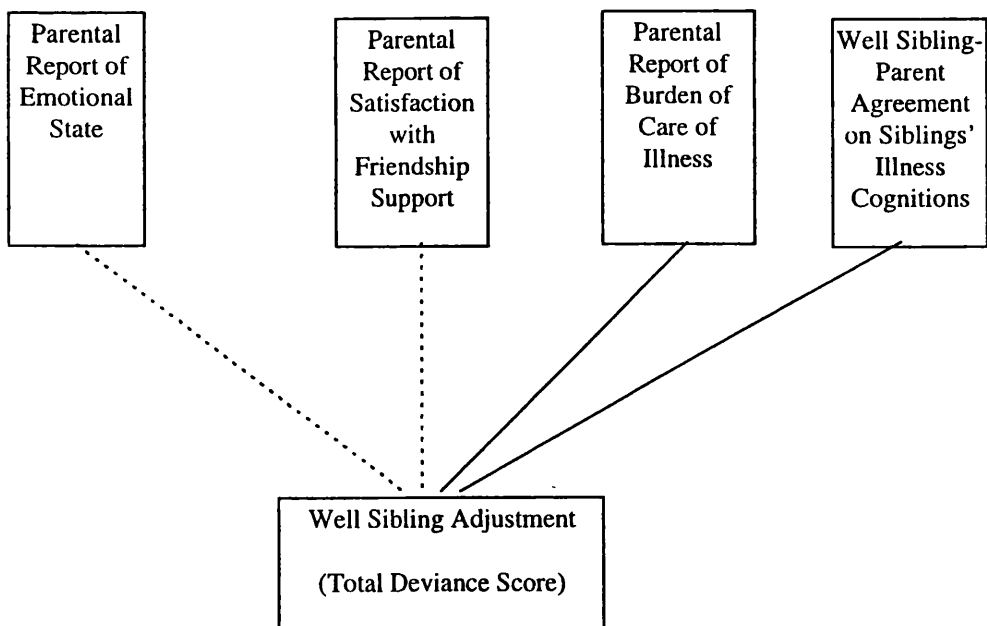
Table 14:
Multiple Regression for Well Sibling-Parent Dyad Agreement Scores

Predictor Variable	Beta	t	p
<u>Parent Report of:</u>			
Emotional State	-0.18	-1.27	0.21
Satisfaction with Community Support	0.16	1.06	0.30
Satisfaction with Friendship Support	0.18	1.13	0.27
Satisfaction with Intimate Relationship	0.06	0.47	0.64
Burden of Care	0.13	1.01	0.32

There was therefore no evidence to support the theoretical suggestion that sibling-parent agreement was a mediating variable between parent psycho-social variables and well sibling adjustment.

A revised well sibling adjustment model is illustrated in Figure 5, in which overall well sibling adjustment (Total Deviance score) is the dependent variable. The variables of maternal satisfaction with community and intimate support were omitted due to their relationship with well sibling adjustment not being supported by either the correlations or multiple regression analyses. The dotted lines represent relationships where there is some evidence of statistical association (from the bi-variate correlations) and the bold lines represent relationships in which there was strong evidence of statistical association (from the bi-variate correlations and multiple regression analyses).

Figure 5
Revised Well Sibling Adjustment Model



3.5 Predictive Ability of Revised Well Sibling Adjustment Model

A discriminant analysis was carried out to estimate the number of cases correctly classified by the revised model (Figure 5). Due to the number of well siblings scoring in

the Total Deviance abnormal and borderline ranges not significantly differing from the normal population, the sample was categorised as a low risk group (Goodman, 1997). Therefore, individuals scoring in the abnormal range formed a 'poor adjustment' group, and individuals scoring in the normal or borderline ranges formed a 'good adjustment' group.

The model correctly classified 78 per cent of cases: 80 per cent of those in the 'good adjustment' group and 60 per cent of those in the 'poor adjustment' group. There were ten false negatives (good adjustment classified as poor adjustment) and four false positives (poor adjustment classified as good adjustment).

3.6 Research Question 4:

Do parents and well siblings report that it would be appropriate for the well sibling to receive psychological support from either (a) participating in a group for brothers and sisters who have chronically ill siblings and /or (b) individual sessions with a member of the hospital staff ?

Most well siblings and mothers felt that group or individual psychological input would not be appropriate. As Table 15 illustrates only thirty-one and thirty-four per cent of well siblings and mothers respectively reported that attending a group would be helpful. Twenty-six and twenty-three per cent of well siblings and mothers respectively reported that attending individual sessions would be helpful.

When mothers reported that their healthy child would benefit from individual psychological input (n=11), either their healthy children had already met with a Clinical

Psychologist (n=7) and /or the ill child was currently acutely ill (n=8). Several mothers who felt that if their healthy child became critically ill, the healthy sibling would benefit from psychological input. Two mothers reported that they would be in favour of individualised work only if it was carried out by someone familiar to the child, such as a teacher supervised by a Psychologist.

Table 15
Participants' Views of Appropriateness of Psychological Input

	Sibling Group		Individual Psychological Sessions	
	Sibling Response n (%)	Parent Response n (%)	Sibling Response n (%)	Parent Response n (%)
Appropriate	19 (30.6)	21 (33.9)	16 (25.8)	14 (22.6)
Not Appropriate	43 (69.4)	41 (66.1)	46 (74.2)	48 (77.4)

The same pattern was not evident for well siblings in favour of individual psychological input. Well siblings in favour of individualised work felt that it should focus on how their sibling's illness was progressing. Several well siblings advised that a special emergency number would be useful to gain information about the illness in times of crisis. This again indicated a heightened sense of responsibility in well siblings.

Most well siblings and mothers who felt group or individual input would be appropriate, suggested that it should focus on practical information (e.g. cause, treatments) or provide an opportunity for well siblings to meet informally together (e.g. visiting theme parks). Several said that they would find returning to the hospital

too distressing and would prefer such input to occur locally in a neutral informal context. Many also stated that groups would work most effectively if they only included children of one age or siblings of children with only one type of condition.

Many mothers and healthy siblings reported that a sibling group would be useful for younger children who were not able to rationalise their mothers' focus on the ill child. As one 13 year old well sibling of a boy with spina bifida said: "A group would be helpful for younger children to help them realise that their parents love them but sometimes need to put the ill person first". Many older well siblings reported that meeting with their own friends and doing their homework was more of a priority than psychology input.

Most mothers discussed wanting to contain the distress of the illness within the family and their close social network. Several mothers commented that they feared another child in their family as being labelled as 'ill', which they felt would be the case if their healthy child received psychological help. However, many felt that they needed help to identify well siblings' needs and felt guilty they spent less time with them than they would wish. It is noteworthy that many mothers commented that the interview questions helped to orient them further to consider the needs of the well siblings. Many mothers recommended that books and videos about chronic illnesses and their impact on family functioning would be usefully developed and given to families to view together and discuss.

Most mothers reported that families would most benefit from NHS resources being allocated to help practically with caring for younger well siblings alongside the ill child. For example, there were several suggestions for hospital family accommodation or the hospital play-groups for well siblings.

DISCUSSION

4.1 Overview of Main Findings

The majority of mothers and healthy siblings were coping well with living with a chronically ill child. The adjustment scale norms indicated that well siblings were more likely to be displaying pro-social behaviours than peers from healthy families. However, they were also significantly more likely to be experiencing clinical levels of emotional and /or conduct difficulties. Qualitative information given by many well siblings indicated feelings of responsibility for the cause of the illness and care for the ill child.

In the whole sample, mothers rated well siblings as having significantly more negative cognitions about the illness and its impact than reported by well siblings themselves. Cognitions were defined as thoughts, feelings and attitudes (Carpenter & Sahler, 1991). Well sibling-parent dyad agreement on well sibling cognitions appeared to vary with the type of cognition. For example, there was higher agreement about cognitions concerned with the illness and ill child, and lower agreement about cognitions concerned with family functioning.

A revised well sibling adjustment model was proposed on the basis of the findings (Figure 5, Results Chapter, p.71). The independent variables were well sibling-parent dyad agreement, maternal report of emotional symptoms, maternal satisfaction with support from friendships and burden of care of the illness. The dependent variable in the model was overall well sibling adjustment. Importantly, the relationship between well sibling-parent agreement scores and overall adjustment scores was not

confounded by their dependence upon parental report. There was no evidence to support the hypothesis that well sibling-parent dyad agreement was a mediating factor between well sibling adjustment and the parental adjustment.

Approximately one third of parents and well siblings reported that well siblings would benefit from psychology input given within a sibling group; approximately one quarter reported that well siblings would benefit from psychology input given individually. Most parents stated that they would prefer to contain their healthy children's distress within the family, and reported that the interview procedure was helpful in orienting them further to their healthy children's needs.

4.2 Interpretation of Results

The impact of chronic illness on healthy siblings has been a neglected area within paediatric psychology. There has been little research into their adjustment, and even fewer investigations of their cognitions about living with an ill child. Little paediatric research has taken a family-oriented perspective. Therefore, research questions in the literature are mostly exploratory and comparison of results between studies can only be tentative.

4.2.1 Well Sibling Adjustment

It is clear that maladjustment in well siblings is not an inevitable outcome of living with a chronically ill brother or sister. They appear to form a low risk group for maladjustment, albeit higher than the general population. Chronic illness is only one

stressor that families contend with in the general population, alongside others such as poor housing, parental separation and so on.

In this study, approximately one quarter of the sample had overall adjustment problems in the borderline or abnormal ranges of the adjustment scale. This compared closely to rates cited within the literature. For example, Sloper and While (1996) and Stallard et al (1997) reported that one quarter of well siblings of children with cancer and life-threatening diseases respectively had overall adjustment scores in the borderline or abnormal ranges.

Well siblings (especially females) were more likely to score within the normal range of the pro-social behaviours sub-scale, in comparison with the adjustment scale's standardisation sample. Many spontaneously expressed a high level of responsibility for their ill brothers and sisters. These results support findings of qualitative studies. For example, Ferrari (1984, 1987) reported that well siblings were more sensitive and socially mature, and Menke (1987) reported that many well siblings felt protective towards their ill sibling. Havermans and Eiser (1994) similarly reported that many well siblings reported positive benefits from their brother or sister being ill, such as developing increased empathy.

Several well siblings in the sample reported experiencing somatic symptoms. This supports previous findings that well siblings are more likely to report physical symptoms (e.g. sleep disturbances, enuresis, appetite problems) and also appear preoccupied with their personal health compared to peers from healthy families

(Lavigne & Ryan, 1979 (cited in Eiser, 1993)). Social Learning theory (Bandura, 1977 (cited in Atkinson, Atkinson, Smith, Bem & Hilgard, 1990)) predicts that well siblings will have learnt that somatic symptoms are an appropriate expression of distress.

4.2.2. Parental Psycho-social Variables

The parental psycho-social variables were burden of care of the ill child, presence of maternal emotional symptoms, and maternal satisfaction with social support. The burden of care variable was the only independent and significant predictor of well sibling adjustment. A hierarchical regression analysis indicated that this finding was not the result of multi-collinearity of the maternal variables of emotional state and satisfaction with social support.

Burden of care has not previously been compared with well sibling adjustment. However, Sloper and While (1996) similarly reported, that level of disruption to family functioning as demanded by a child's cancer (i.e. number of nights child spent in hospital) was significantly associated with well sibling adjustment. The mean score on the Burden of Care Index was similar to other studies in which parents have rated burden of care for a child with a chronic illness. For example, Ray and Ritchie (1993) cited a mean score of 242 (SD=134), compared to this study's mean score of 231 (SD=75). This indicates that the sample was representative of the target population in terms of burden of care demanded by the sick child.

The presence of maternal emotional symptoms was not significantly associated with overall well sibling adjustment when all other parental psycho-social variables were

controlled for (in the multiple regression analysis), although the two variables were significantly correlated. However, maternal emotional symptoms and peer relationship problems were significantly associated when the other variables were controlled for. There is a considerable body of research showing strong relationships between parental emotional state and healthy children's behavioural problems (Richman, Stevenson & Graham, 1982 (cited in Eiser, 1993); Stocker, 1991; Tew & Lawrence, 1973). The lack of a stronger relationship in this study may be partly explained by many siblings being aware of the reasons for their parents' distress in relation to the illness. Sloper and While (1996) hypothesise that the ability to self-reflect is an important aspect of resiliency.

The mean score on the Malaise Inventory of this sample of 6.5 (SD=4.7) was similar to other studies. This indicates that the sample was representative of the target population with respect to this variable. For example, Sloper and While (1996) cited a mean score of 6.8 (SD= 4.6) for mothers with a child with cancer, and Tew and Lawrence (1973) cited a mean score of 6.1 (SD=5.3) for mothers with a child with spina bifida.

No statistical associations were found between maternal satisfaction with intimate and community support and overall well sibling adjustment. Maternal satisfaction with friendship support and well sibling adjustment were significantly correlated, but were not significantly associated when all other variables were controlled for. There is considerable evidence in the literature that these hypothesised relationships exist (Ferrari, 1984; Sloper & While, 1996). These relationships are likely to reduce parenting duties (e.g. looking after well siblings) and also be emotionally supportive.

They are therefore hypothesised to facilitate maternal adjustment and allow parents to be more available to well siblings. The lack of statistical association with well sibling adjustment may have resulted from the poor psychometric properties of the satisfaction scales and response bias of the sample on the Intimate Relationship sub-scale.

4.2.3 Well Sibling-Parent Agreement on Well Siblings' Cognitions about the Illness and its Impact

In this study, mothers over-estimated the number of negative well siblings' cognitions about the illness and its impact on family life. This contrasts with previous literature (Menke 1987), which suggests that parents underestimate their healthy children's concerns. The over-estimation may have reflected mothers' own anxiety about their healthy child's experience of the illness. As one parent said of her 10 year old healthy child, "I fear that she will think back to the day G. [the patient] was diagnosed and think 'that was the day I lost my freedom'". Similarly to previous research, there was no evidence of agreement varying with the gender of the well sibling (Menke, 1987). There was also no evidence of well sibling-parent agreement varying with the age of the well sibling.

The highest levels of agreement were on items concerned with well sibling cognitions about the illness or the ill child. For example, almost all well sibling-parent pairs agreed that the healthy siblings understood (or did not understand) why parents had to spend more time with their ill brother or sister. Mothers were much less reliable at reporting well siblings' cognitions about issues not directly related to the illness, such as family

functioning or the well siblings' emotional functioning.

The upper range of the percentage agreements found in this study were extremely high compared with similar research. Craft and Craft (1989) cited a range of 72 to 87 per cent agreement on well sibling-parent reports of changes in physical functioning since illness onset (e.g. bedwetting). The lower range of these percentage agreements on well sibling cognitions about their emotional reactions and family functioning compares well with Craft and Craft's findings. As discussed in the Method section (p.47), the high agreement rates may have partly resulted from the dichotomous response format of the SDQ and therefore high chance agreement.

Well sibling and parent versions of the Sibling Perception Questionnaire were specially developed for the study, and therefore data could not be directly compared with previous studies. In Klein's (1991) review of parent-child agreement studies, she expresses concern that the test-retest reliability of the tool used to assess agreement is usually ignored. In this study, test-retest reliability over one week was high. Although a week is a short space of time, it was decided that many of the illnesses could change significantly over a longer period of time, and thus impact on the well siblings' cognitions about the illness and their life circumstances.

In conclusion, mothers' ability to report on their healthy children's cognitions about the illness did not appear reliable. However, their level of agreement did appear predictive of well siblings' overall adjustment. Importantly, the relationship between well sibling-parent agreement scores and overall adjustment scores was not

confounded by their dependence upon parental report. While the reliance on parental reports of their adjustment largely reflects ethical considerations of involving the children in research, it creates a void in the understanding of well siblings' feelings, beliefs and attitudes.

4.2.4 Revised Well Sibling Adjustment Model

The proposed adjustment model (Figure 5, Results chapter, p.71) was supported by McCubbin and co-workers' systemic model (McCubbin, Joy, Cauble, Comeau, Patterson & Needle, 1980; McCubbin & Patterson, 1983). Their model suggests that successful adaptation to a crisis is a function of two factors: firstly, the manner in which the family handles the differing cognitions of each family member, and secondly, how the family deals with the demands of the illness with available resources. Similarly in this study, well sibling-parent agreement on siblings' illness cognitions and burden of care of the illness were predictive of overall well sibling adjustment and well sibling emotional problems. The fact that they remained significant after the entry of individual parental variables (emotional state and satisfaction with support) underlines the interdependence between well siblings, their parents and the demands placed on the family system. It can be hypothesised that well siblings respond primarily with negative emotions to gain parental attention (emotional or physical) without needing to directly confront the issue that their parents do not appear aware of their attitudes, emotions and beliefs about the illness.

The revised adjustment model was only able to predict problems in adjustment, and not levels of pro-social behaviours. This could be due to the small range in the pro-social

sub-scale scores resulting in lower statistical power in the analysis. It may also be attributable to well siblings displaying an inflated rate of pro-social behaviours as a direct result of the increased opportunities for such behaviours in families with a sick child.

There was no evidence to support the hypothesis that well sibling-parent dyad agreement was a mediating factor between well sibling adjustment and the parental psycho-social variables. It is likely that parental awareness results from the general quality of the relationship between parents and their healthy children, rather than the amount of parental availability (emotional and physical) to them. This hypothesis was supported by the study's finding that well sibling-parent agreement did not vary with the length of illness, although some conditions (e.g. spina bifida) had pre-dated the well sibling's birth. It was also supported by many mothers commenting that they felt the well siblings' adjustment reflected the whole family's approach to living with an ill child. For example, three families with a range of illnesses and well adjusted healthy children talked openly about their shared religious faith.

4.2.5 Illness and Demographic Variables

The diagnoses and associated illness variables (such as prognosis and duration) were not significantly associated with the dependent or the independent variables in the model. This finding supports the non-categorical approach to sampling in psycho-social studies of chronic disease as discussed by Pless and Perrin (1985).

The finding that well sibling adjustment did not vary with the size of the family, did not support previous results. In Eiser's (1993) review of paediatric psycho-social research, she comments that well siblings in general appear to adjust better in larger families. Stocker (1991) suggests this is due to supportive relationships with siblings compensating for problematic or less supportive relationships with parents. As with this study, no clear pattern emerged with ordinal position. However, function of ordinal position in a family is less easy to interpret since age and ordinal position are inextricably linked.

4.2.6 Service-Related Questions

The majority of mothers stated that they would prefer to support their healthy children themselves rather than their children attend Clinical Psychology sessions. Parents in favour of psychological input, either had a critically ill child or their healthy children had already received psychological treatment. Well siblings in favour of group or individualised help mostly advised that the Psychologist should take a psycho-educational role or facilitate well siblings meeting informally together. This supports previous research findings that many healthy siblings report wanting more illness information (Stallard et al, 1997).

Most mothers and well siblings appeared to want to maintain their day-to day life as normal as possible without further hospital-based interventions. Nevertheless, some mothers concurrently reported a lack of time and emotional energy to talk to well siblings, and this was supported by them having higher scores on the burden of care and emotional state measures respectively. As noted above, this lack of availability did

not affect parental awareness of problematic issues for well siblings. Several older siblings stated that they had priorities away from the family (e.g. homework) and wanted to focus on them. Several mothers also commented that they would feel they had other 'ill children', if they were meeting with a Psychologist. They suggested that they could be helped to support their healthy children through psycho-education.

4.3 Limitations of the Study

Three areas of the study will be reviewed: the design, methodology and variables investigated.

4.3.1 Non-Experimental Design

The model used in this study predicted that parents' awareness of well siblings' cognitions and lower burden of care would result in better well sibling adjustment. However, this causal direction could not be inferred from the correlational design used (Barker, Pistrang & Elliott, 1994). It is likely that well sibling-parent agreement and well sibling adjustment will mutually affect each other. Additionally, the relationship between these two independent variables and well sibling adjustment might also stem from another unmeasured source, for example the security of the well siblings' attachment to their parents.

4.3.2 Methodology

4.3.2.1 Sampling

The study's statistical analysis was limited by the small sample size. A larger sample size would have allowed for more detailed study of the effects of demographic and

illness variables on the dependent and independent variables. For example, the sample included well siblings across an eight year age-range. No detailed conclusions could be made as to developmental changes in well siblings' cognitive appraisals of the illness and its impact, the developmental changes in well sibling-parent agreement across ages nor well siblings' clinical psychology requirements over age groups.

The external validity of the findings was likely to be limited by several sampling factors. First, the sample was largely composed of families with a white ethnic origin and from non-manual socio-economic backgrounds. No demographic information was available on non-participating families, and therefore no conclusions could be made as to whether the sample was demographically representative of the target population of families. However, there is no research evidence to suggest that families with a child with a chronic illness are more likely to be of white ethnic origin or be from the non-manual classes. Systemic theory proposes that family functioning and interaction with other social systems (e.g. religious or health care systems) will be based upon cultural values and societal demands (Minuchin, 1974). The homogeneity of the sample precluded examining ethnic and cultural differences in the families' functioning.

The external validity of the findings was likely to be compromised further by the sampling procedure. Twenty-four per cent of eligible families could not be initially contacted. This may have been due to a change in address, but may also have resulted from them having fewer resources (e.g. not having a telephone to be contacted by the researcher or not having the time to return the reply slip). Sixteen per cent of contacted families refused to participate. Of those families which gave a reason for

non-participation, many were experiencing additional stressors to the illness of the child (e.g. bereavement) or the ill child being critically ill. This may have biased the sample further towards those with fewer life stressors and less practical and psychological burdens of care.

Eiser (1993) further suggests that participating families are more likely to engage in open communication about illness issues, compared to families who refuse to participate. Additionally, two hospitals opted to choose eligible families from their patient lists themselves. Although they were urged to select every family which met the inclusion criteria, the selectors may have been biased towards those they felt were more open in their communication about the illness.

However, it was noteworthy that the distribution of well sibling adjustment scores, maternal emotional symptom scores and burden of care scores appeared representative of previous samples used in similar investigations

4.3.2.2 Measures

Well sibling-parent agreement was assessed using an adapted form of the Sibling Perception Questionnaire. The scales' items were intended to be negatively biased, which resulted in no possible assessment as to whether respondents were tending towards acquiescence (Barker, Pistrang & Elliott, 1994). Additionally some items did not appear to be negative in content, although they were scored as such. For example, one item asked whether the well sibling wondered why his brother or sister became ill. This may be an adaptive cognition rather than a negative one.

Parental emotional symptoms were assessed using the Malaise Inventory. This scale has been recently criticised for its dichotomous response format and it focusing mostly on physical signs of stress (Cheung & Buchannan, 1997). Burden of care of the illness was measured using the Clinician's Overall Burden Index. The only concern with this scale was that it did not include care for the ill child's own emotional reactions to having the illness, which several mothers emphasised as an additional aspect of caring for more critical illnesses or those with disfiguring treatment side-effects (e.g. cancer).

Interpretation of the data collected from the parental satisfaction sub-scales was limited. The internal consistency of the intimate relationship sub-scale and the test-retest reliability of the community sub-scale were poor. This is not an uncommon problem with social support measures. Bowling (1997) concludes that their development has suffered from methodological problems and from there being "little attempt to test measures of social support for reliability and validity" (p.94).

4.3.2.3 Self-report Measures

As with all self-report measures, there is the problem of participants responding in a social desirable way, either consciously or unconsciously (Crowne & Marlowe, 1960, (cited in Barker, Pistrang & Elliott, 1994)). This was likely to be high in this study because well siblings were informed for ethical reasons that their parents would be told anything they said of concern.

Additionally, mothers' responses may have been affected by their own emotional state. There is extensive literature on the effects of parental emotional state on parents' reports of children's adjustment problems. For example, Breslau, Davis and Prabucki (1988) (cited in Klein, 1991) reported that maternal depression significantly raised the number of child adjustment problems reported. However, they also reported that it did not affect the accuracy of awareness of their healthy children's cognitions. Nevertheless, mothers' reports of the well siblings' cognitions may have reflected their own anxieties or hopes about the siblings' inner experience of the illness. As one mother said "I think my answers were a projection of my own feelings and fears".

4.3.3 Variables not Investigated

The following variables were not assessed within the present study and may have impacted upon well sibling-parent agreement and well sibling adjustment:

4.3.3.1 Parents' Behaviours towards Well Sibling

The systemic model used in this study suggests that parents need to be both aware and to act upon the differing cognitions of their healthy children to facilitate their adaptation. The present study did not look at parents' actual behaviours in validating the well siblings' sense of worth in the family, but only looked at their awareness of such cognitions.

4.3.3.2 Relationship between Well Sibling and Ill Child

The current relationship between the well sibling and the ill child was not investigated, nor was their previous relationship before the illness onset (if applicable). Both aspects

of their relationship are likely to affect well siblings' cognitive appraisals of the illness and its impact, and also parental awareness of these cognitions. Much research has commented on the strong mutual influence between siblings (Dunn, 1988) throughout development. They experience intense affects including envy, jealousy, love, companionship and empathy, which are likely to be greatly exaggerated when one sibling has a chronic illness (Eiser, 1993). Opportunities for joint activities may be reduced, and concern and anxiety may dominate the relationship. Additionally, parental differential treatment of their children has been related to more conflict and negative behaviour between the siblings themselves (Boer, 1990, (cited in Eiser, 1993)).

4.3.3.3 Relationship between Well Sibling and other Adults

The well siblings' relationships with other adults, and especially fathers was not considered. It cannot be assumed that well sibling adjustment is dependent only on their relationship with their mother. In two-parent families, many parents commented that if only one parent was to be chosen, fathers would be in the best position to reflect on well siblings' cognitions and adjustment. They argued that when the sick child had been acutely ill, the mother had stayed at the hospital and the father had stayed at home with the well siblings.

Additionally, many well siblings mentioned other adults (e.g. grand-parents) who had been particularly helpful. These adults may in some cases have compensated for decreases in emotional or physical availability of parents. Moreover, with the longer-term conditions (e.g. muscular dystrophy, spina bifida), a consistently supportive

relationship with any adult can be protective in traumatic circumstances (Mrazek & Mrazek, 1987).

4.3.3.4 Well Siblings' Adjustment before Illness Onset

Well siblings' adjustment before the onset of the illness was not assessed due to the non-cancer conditions being mostly long-standing and even pre-dating some siblings' births. However, two thirds of the ill children had been acutely ill in the last six months, which systemic theory would hypothesise as impacting on family functioning. In this sub-sample, pre-existing problems in some well siblings might have masked associations changes in their adjustment status over that time. However, it is acknowledged that pre-existing adjustment difficulties would be a further risk factor.

4.3.3.5 Attachment Status of Well Siblings

The security of the well siblings' attachment to their parents in their early years may have affected their later adjustment to chronic illness in a brother or sister and the demands it makes on family functioning (Bowlby, 1979). As one mother of an eight year old (F.) and a four year old with unstable diabetes said "(F) was helped by having four years alone with me before my youngest was born".

4.3.3.6 Well Siblings' Care of the Ill Child

The burden of care demanded by the illness was conceptualised as a responsibility of the parents. However, in reality well siblings often find themselves assuming a large burden of care and responsibility for the ill child, especially if either parent shows signs

of severe depression or maladjustment (Burton, 1975; Gath, 1974 (cited in Eiser, 1993). Parents may also use them as confidantes and sources of social support.

4.4 Methodological Implications

Research into the psycho-social effects of childhood illness on healthy siblings is a relatively new area of study. The results of this study indicate the centrality of both systemic and cognitive factors in the adjustment process. Methodologies based on systemic measures, which assess all family members' cognitions of the illness experience, are essential to build a clearer picture of family adjustment and its impact on well siblings. This and previous study results indicate that mothers are not reliable substitute reporters of their children's cognitions of the illness experience. Longitudinal studies will allow for the further identification of specific factors which place well siblings at risk of poor adjustment or conversely facilitate adaptation.

Further research could also usefully embrace the recent trend in Clinical Psychology to combine quantitative and qualitative methodologies. Qualitative methodology has the advantage of accessing participants' individual experiences through their own narratives, without the restrictions imposed by a simplistic quantitative design. By hearing participants' stories, an exploration of the personal meanings they have given to experiences can be assessed. Additionally, Barker, Pistrang & Elliott (1994) argue that the data are more accessible to readers since it consists of verbal descriptions rather than abstract numerical descriptions.

Eiser (1998) further suggests that children's adjustment is usefully assessed by using quality of life measures. These are currently under development and importantly rely on children's own responses.

4.5 Clinical and Professional Implications

The clinical implications of this study need to be addressed by Clinical Psychologists and other clinicians. The scarcity of Clinical Psychologists in the NHS means it is important to disseminate findings among professionals, especially nursing and medical teams which have most face-to-face contact with families. Additionally, this study indicates that well siblings of children with both shorter- and longer-term conditions may show adjustment difficulties.

There is minimal literature as to the specific types of help families feel would benefit well siblings. By taking part in the study, the participating families indicated that the psycho-social effects of chronic illness are a concern for them. However, parents in the main, appear to specifically want help themselves to empower them in obtaining and mobilising support for their healthy children. Worried parents need to be reassured that although well siblings are at risk of adjustment difficulties, they are a low risk sample and also that many well siblings report benefits from the experience.

Several families acknowledged not understanding what Clinical Psychology is. Many mothers equated receiving psychological help with being 'ill', and both mothers and well siblings reported not wanting to return to the hospital. Clinical Psychologists will promote themselves and their work best by working collaboratively with clinical teams

which have day-to-day contact with families. In this way, psycho-educational work with clinicians, parents and children may be carried out routinely in workshops in non-medical settings, videos and books. This information would aim to help prevent family adjustment difficulties. Suggested topics might be identifying common well sibling concerns, guidance around normalising these emotions, helping siblings to disclose feelings about illness issues and spending special time with siblings.

The strong association between parental awareness and well sibling adjustment, emphasises the importance of families with significant adjustment difficulties, being treated together using a systemic framework. The aim would be to help re-organise families' perceptions of their relationships and roles in order to meet the direct and indirect demands of the illness. A useful exercise would be to externalise the discrepancies in well siblings' and parents' perceptions of the illness and its impact, and then assess any change in the well siblings' adjustment.

Finally, clinicians must be made aware of the ease of reinforcement of well siblings' perceptions of a special relationship existing between parents and the ill child and not the well sibling. Stewart, Stein, Forrest and Clark (1992) (cited in Eiser, 1993) interviewed ten healthy siblings each of children with a terminal illness. None of these children had ever discussed the illness with a doctor, although three of the older children would have liked to.

4.6 Future Research

Future research is recommended to build upon several of the ideas discussed. Firstly, the concept of child adjustment being a function of parent-child agreement on problematic issues could be tested more systematically with paediatric and non-paediatric samples. Levels of well sibling-parent agreement and adjustment could be compared between healthy siblings in the same family, where the burden of care of the illness is constant.

Secondly, an investigation of the relationship between mutual well sibling-parent awareness of problematic issues and family adjustment would be interesting. In the present study, this idea would have translated as well siblings additionally reporting on their parents' adjustment state. However, it is unclear whether it would be developmentally possible for children to think about and report adult cognitions.

Thirdly, it would be useful to investigate what parents actually do to validate well siblings' sense of worth in the family and what well siblings find useful. For example, well siblings and parents could be asked about what activities they find helpful to do together (e.g. joint hospital visits), or what issues they would like to discuss with each other.

4.7 Conclusions

The findings indicate that well siblings, of children with a range of chronic conditions, are a low risk group for overall adjustment problems. However, compared to the normal population many experience emotional and conduct difficulties, but many also display pro-social behaviours.

The results support McCubbin and Patterson's (1983) systemic model of family adjustment. The factors most strongly associated with well sibling adjustment were parental awareness of their cognitive responses to the illness and the burden of care demanded by the illness. The strong association between parent-child agreement about children's cognitions and child adjustment is an interesting avenue for further research in both paediatric and non-paediatric areas.

In practice, mothers' abilities to report on their healthy children's cognitions about the illness did not appear reliable. Whenever possible, parents should not be used for proxy assessments of their healthy children's emotions, attitudes and feelings about specific issues. Most mothers and well siblings indicated that they would prefer not to use Clinical Psychology services, but would prefer well siblings to be helped indirectly through psycho-education or meeting other families in similar situations.

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APPENDIX 1:

Introductory Letter and Information Sheets



Sub-Department of Clinical Health Psychology

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CONFIDENTIAL

(Date)

Dear (Name of Parent),

We are psychologists interested in the experiences of families in which a child is ill. This study is under the supervision of xxx (Consultant Pediatrician) and xxx (Child Clinical Psychologist) at the xxx Hospital.

We are writing to invite your family to take part in this research. It involves an interview with yourself and also a brother and /or sister of the child being treated at the hospital. The interviews will be informal, and are intended to be helpful and supportive. The interviews could take place either at the xxx Hospital or at your home, depending upon which suits you best.

Please find enclosed information sheets which tell you more about the project. If you have any questions, please feel free to contact xxx (Child Clinical Psychologist, xxx Hospital) or (Researcher's name) on (Telephone number).

At the bottom of this letter is a 'Reply Form'. We would be grateful if you would complete it and send it to the address below in the stamped addressed envelope. Alternatively we will contact you on the telephone.

We hope you will feel able to take part in this study. Thank you for your help.

Yours sincerely,

xxxx
Clinical Psychologist in Training

xxxx
Child Clinical Psychologist

I am happy to be contacted to discuss taking part in this research YES/NO

Name of Parent.....

Names (and ages) of brothers /sisters of child being treated at the xxx Hospital

1).....

2).....

3).....

Address and /or Telephone Number

.....
.....

RETURN ADDRESS: Researcher's name, Department, University Name, Street, City.



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CONFIDENTIAL

INFORMATION SHEET FOR PARENTS

Title of Research Study

Healthy Siblings' Adjustment To Chronic Illness In A Brother Or Sister

Researchers

Name, Clinical Psychologist in Training, Name of University

Name (Supervisor of Study), Professional Title, Name of University

About the research study

Families react in many different ways to living with and caring for an ill child. Sometimes parents, brothers and sisters report it is difficult; sometimes they report positive consequences, for example the family becoming closer.

Over the last few years, hospital teams (e.g. doctors, nurses, psychologists) have started to provide more individualised support to families. For example, some hospitals provide groups for brothers and sisters to discuss their feelings. In order that hospital teams continue to improve such services and provide the best types of support for all family members, we are interviewing families about their day-to day life with an ill child.

If you take part in this research study

(Name of Consultant Pediatrician) has given us permission to invite some families of children being treated at the xxx Hospital to help us with this research. If you decide to take part, you and a brother /sister of your child will be interviewed at the hospital or at your home, at a time which is convenient to you. All the information collected in this study will be confidential and your family's name will not be used.

You and your child do not have to take part in this study if you do not want to. If you decide to take part, you may withdraw at any time without having to give a reason. If you withdraw, your child's treatment will not be affected in any way. The interview with you takes about one hour and the interview with the child's brother /sister takes about 20 minutes. In the unlikely event that either you or your child are upset by the interview, it would be stopped immediately.

All proposals for research using human subjects are reviewed by an ethics committee before they can proceed. This proposal was reviewed by the xx Hospital Research and Ethics Committees. This project is covered by xxx No Fault Indemnity insurance protection.

If you would like any more information about the research study, please feel free to contact (researcher name) on (telephone number). Alternatively, you can telephone (field supervisor name) on (telephone number) or (academic supervisor name) on (telephone number).



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CONFIDENTIAL

INFORMATION SHEET FOR BROTHERS AND SISTERS

Title of Research Study

Healthy Siblings' Adjustment To Chronic Illness In A Brother Or Sister

Researchers

Name, Clinical Psychologist in Training, Name of University

Name (Supervisor of Study), Professional Title, Name of University

About the study

Brothers and sisters react in many different ways to living with an ill child. Sometimes they say it is difficult; sometimes they say it is OK. Over the last few years, doctors, nurses and psychologists have helped children who feel upset about their brother or sister being ill. For example, some hospitals run groups for brothers and sisters to discuss their feelings. This study will help hospitals to know how they can further help brothers and sisters of ill children.

You do not have to take part in this study if you do not want to. Your decision will not affect your sister's /brother's hospital care in any way.

If you decide to take part, you may change your mind without giving a reason. I will meet with you for about 15 minutes and your Mum /Dad for about one hour. This could take place at the hospital or at your home. We will be discussing a number of aspects of how your brother or sister's illness has affected you. The information that we get will be confidential, although if the psychologists involved decide that it is important that your parents are aware of what has been discussed, then they may be informed.

If you would like any more information about the research study, please feel free to contact (researcher name) on (telephone number). Alternatively, you can telephone (field supervisor name) on (telephone number) or (academic supervisor name) on (telephone number).

APPENDIX 2:

Example Letter Granting Ethical Approval

RIVERSIDE RESEARCH ETHICS COMMITTEE

CHELSEA & WESTMINSTER HOSPITAL

Lower Ground Floor Pharmacy Offices

369 Fulham Road London SW10 9NH

Tel: 0181 846 6855 Fax: 0181 846 6860

4 August 1997

Miss Victoria Smith (Clinical Psychologist in Training, University College London)

Dear Miss Smith

RREC 1467 - An exploration of parent-sibling understanding in families of a child with a serious illness.

I am writing to inform you that the above study has been considered and now approved by Chairman's action.

Please note the following conditions which form part of this approval:

- [1] This approval is for one year only. For projects with an expected duration of more than one year, a letter from the principal investigator will be required in order to further extend consent. This will enable the Committee to maintain a full record of research.
- [2] Any changes to the protocol must be notified to the Committee. Such changes may not be implemented without the Committee's approval.
- [3] The Committee should be notified immediately of any serious adverse events or if the entire study is terminated prematurely.
- [4] You are responsible for consulting with colleagues and/or other groups who may be involved or affected by the research, e.g., extra work for laboratories. Approval by the Committee for your project does not remove your responsibility to negotiate such factors with your colleagues.

Cont/2...

Cont/2...RREC 1467 - An exploration of parent-sibling understanding in families of a child with a serious illness.

- [5] You must ensure that nursing and other staff are made aware that research in progress on patients with whom they are concerned has been approved by the Committee.
- [6] Pharmacy must be told about any drugs and all drug trials, and must be given the responsibility of receiving and dispensing any trial drug.
- [7] The Committee must be advised when a project is concluded and should be sent one copy of any publication arising from your study, or a summary if there is to be no publication.

May I take this opportunity to wish you well in your research. However, if any doubts or problems of an unexpected nature arise, please feel free to contact me at any time.

Yours sincerely



J Nigel Harcourt-Webster MD FRCPATH
Chairman - RREC

Seen and Approved	
Submission	18-06-97
Protocol	Signed Initials: —
Information Sheet	Signed Initials: J.N.H.W.
Consent Form	Signed Initials: J.N.H.W.
Questionnaires	Signed Initials: J.N.H.W.
Letter of Indemnity	Signed Initials: —
CTX/DDX/Licence	Signed Initials: —

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APPENDIX 3:

Consent Forms (Parent and Well Sibling Versions)



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CONFIDENTIAL

CONSENT FORM FOR PARENTS

Title of Research Study

Healthy Siblings' Adjustment To Chronic Illness In A Brother Or Sister

Researchers

Name, Clinical Psychologist in Training, Name of University

Name (Supervisor of Study), Professional Title, Name of University

To be completed by parents:

Delete as appropriate

- 1) Have you read the information sheet about the study? YES /NO
- 2) Have you had the opportunity to ask questions and discuss this study? YES /NO
- 3) Have you received satisfactory answers to all your questions? YES /NO
- 4) Have you received enough information about this study? YES /NO
- 5) Who have you spoken to about this study?.....
- 6) Do you understand that you and your child are free to withdraw from this study:
 - at any time YES /NO
 - without having to give a reason YES /NO
 - without affecting your child's future medical care YES /NO
- 7) Do you agree for you and your child to take part in this study? YES /NO

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

Your Name in Block Letters.....

Names of siblings (between 8 and 16 years old) of the child being treated at XX Hospital

1).....

2).....

3).....

Researcher's Name.....Signed.....Date.....



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CONFIDENTIAL

CONSENT FORM FOR CHILDREN (WELL SIBLINGS)

Title of Research Study

Healthy Siblings' Adjustment To Chronic Illness In a Brother or Sister

Researchers

Name, Clinical Psychologist in Training, Name of University

Name (Supervisor of Study), Professional Title, Name of University

To be completed by the child:

Delete as appropriate

1) The study has been explained to me, and I understand it YES /NO

2) I agree to take part in this study and know that I can come out at any time YES /NO

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

Your Name in Block Letters.....

Researcher's Name.....Signed.....Date.....

APPENDIX 4:

Parent Questionnaires and Strengths and Difficulties Questionnaire Norms

MALaise INVENTORY

Interview Number:

COMPLETED BY RESEARCHER

“Here are some things parents have told us about how they feel. You may feel that some items used to apply, but do not now. We only want to know about the last 6 months.”

<u>Do you often have back-ache ?</u>	<u>YES</u>	<u>NO</u>
<u>Do you feel tired most of the time ?</u>	<u>YES</u>	<u>NO</u>
<u>Do you often feels miserable ?</u>	<u>YES</u>	<u>NO</u>
<u>Do you often have bad head-aches ?</u>	<u>YES</u>	<u>NO</u>
<u>Do you often get worried about things ?</u>	<u>YES</u>	<u>NO</u>
<u>Do you usually have difficulty falling asleep or staying asleep ?</u>	<u>YES</u>	<u>NO</u>
<u>Do you wake early in the morning for no special reason ?</u>	<u>YES</u>	<u>NO</u>
<u>Do you wear yourself out worrying about your health ?</u>	<u>YES</u>	<u>NO</u>
<u>Do you often get into a violent rage ?</u>	<u>YES</u>	<u>NO</u>
<u>Do people often irritate or annoy you ?</u>	<u>YES</u>	<u>NO</u>
<u>Have you at times had twitching of the face, head or shoulders ?</u>	<u>YES</u>	<u>NO</u>
<u>Do you often become scared for no good reason ?</u>	<u>YES</u>	<u>NO</u>
<u>Are you scared to be alone when there are no friends near you ?</u>	<u>YES</u>	<u>NO</u>
<u>Are you easily upset or irritated ?</u>	<u>YES</u>	<u>NO</u>
<u>Are you frightened of going out alone or of meeting people ?</u>	<u>YES</u>	<u>NO</u>
<u>Are you constantly keyed up and jittery ?</u>	<u>YES</u>	<u>NO</u>
<u>Do you suffer from indigestion ?</u>	<u>YES</u>	<u>NO</u>
<u>Do you suffer from an upset stomach ?</u>	<u>YES</u>	<u>NO</u>
<u>Is your appetite poor ?</u>	<u>YES</u>	<u>NO</u>
<u>Does every little thing get on your nerves, and wears you out ?</u>	<u>YES</u>	<u>NO</u>
<u>Does your heart often race like mad ?</u>	<u>YES</u>	<u>NO</u>
<u>Do you often have bad pains in your eyes ?</u>	<u>YES</u>	<u>NO</u>
<u>Are you troubled with rheumatism or fibrosis ?</u>	<u>YES</u>	<u>NO</u>
<u>Have you had a nervous breakdown ?</u>	<u>YES</u>	<u>NO</u>

SIBLING PERCEPTION QUESTIONNAIRE (PARENT VERSION)

Interview Number:

COMPLETED BY RESEARCHER

“Here are some things children have told us about how their sister’s or brother’s illness affects them. We’d like you to put yourself in your (healthy) child’s position, and indicate which of these thoughts s/he has had in the last 6 months”.

Wishes there was something s/he could do about his her brother’s or sister’s illness	YES	NO
Wishes you spent less time with his /her brother or sister	YES	NO
Wishes you spent more time with him /her	YES	NO
Feels sad about his /her brother or sister being ill	YES	NO
Feels people are more interested in his /her brother or sister than him /her	YES	NO
Feels that because of his /her brother’s /sister’s illness, people don’t care how s/he feels	YES	NO
Thinks about his /her brother’s or sister’s illness	YES	NO
Feels that because of his /her brother’s /sister’s illness, you ignore him /her	YES	NO
Feels that because of his /her brother’s /sister’s illness, s/he has too much to do in the house	YES	NO
Wonders why his /her brother or sister got sick	YES	NO
Worries that s/he can catch the illness from his /her brother or sister	YES	NO
Doesn’t want to bother you with his /her worries	YES	NO
Feels angry about his /her brother or sister being ill	YES	NO
Feels afraid of his /her brother’s or sister’s illness	YES	NO
Wishes s/he knew someone who understands how s/he feels	YES	NO
Understands why you have to spend more time with his /her brother or sister	YES	NO
Feels his /her friends worry that they can catch the illness from his /her brother or sister	YES	NO
Forgets that his /her brother or sister is ill	YES	NO
Feels that because of his /her brother’s /sister’s illness, you don’t do much as a family	YES	NO
Feels s/he cannot talk to his /her friends about his /her brother’s /sister’s illness	YES	NO
Feels s/he cannot talk to you about his /her brother’s /sister’s illness	YES	NO
Feels s/he cannot talk to you about his /her school work	YES	NO
Feels s/he cannot talk to other adults about his /her brother’s /sister’s illness	YES	NO

(Adapted From: Sibling Perception Questionnaire of Illness Experience (Carpenter & Sahler, 1991))

STRENGTHS AND DIFFICULTIES QUESTIONNAIRE

Interview Number:

COMPLETED BY RESEARCHER

“Please give your answers on the basis of your child’s (i.e. sibling of ill child) behaviour over the last six months. It would help us if you answered all items as best as you can even if you are not absolutely certain or the item seems daft”

	NOT TRUE	SOMEWHAT TRUE	CERTAINLY TRUE
MY CHILD (i.e. WELL SIBLING):			
<u>Considerate of other people’s feelings</u>	_____	_____	_____
<u>Restless, overactive, cannot stay still for long</u>	_____	_____	_____
<u>Often complains of headaches, stomach aches or sickness</u>	_____	_____	_____
<u>Shares readily with other children (food, toys, pencils etc.)</u>	_____	_____	_____
<u>Often has temper tantrums or hot tempers</u>	_____	_____	_____
<u>Rather solitary, tends to play alone</u>	_____	_____	_____
<u>Generally obedient, usually does what adults request</u>	_____	_____	_____
<u>Many worries, often seems worried</u>	_____	_____	_____
<u>Helpful if someone is hurt, upset or feeling low</u>	_____	_____	_____
<u>Constantly fidgeting or squirming</u>	_____	_____	_____
<u>Has at least one good friend</u>	_____	_____	_____
<u>Often fights with other children or bullies them</u>	_____	_____	_____
<u>Often unhappy, down-hearted or tearful</u>	_____	_____	_____
<u>Generally liked by other children</u>	_____	_____	_____
<u>Easily distracted, concentration wanders</u>	_____	_____	_____
<u>Nervous or clingy in new situations, easily loses confidence</u>	_____	_____	_____
<u>Kind to younger children</u>	_____	_____	_____
<u>Often argumentative with adults</u>	_____	_____	_____
<u>Picked on or bullied by other children</u>	_____	_____	_____
<u>Often volunteers to help others (parents, teachers, children)</u>	_____	_____	_____
<u>Can stop and think things over before acting</u>	_____	_____	_____
<u>Can be spiteful to others</u>	_____	_____	_____
<u>Gets on better with adults than with other children</u>	_____	_____	_____
<u>Many fears, easily scared</u>	_____	_____	_____
<u>Sees tasks through to the end, good attention span</u>	_____	_____	_____

STRENGTHS AND DIFFICULTIES QUESTIONNAIRE NORMS

	Normal	Borderline	Abnormal
Total Difficulties Score	0 - 13	14 - 16	17 - 40
Emotional Symptoms Score	0 - 3	4	5 - 10
Conduct Problems Score	0 - 2	3	4 - 10
Hyperactivity Score	0 - 5	6	7 - 10
Peer Problems Score	0 - 2	3	4 - 10
Pro-social Behaviour Score	6 - 10	5	0 - 4

(Strengths and Difficulties Questionnaire (Goodman, 1997))

SOCIAL SUPPORT INDEX

Interview Number:

COMPLETED BY RESEARCHER

“Here are some things parents have told us about the amount of social support they receive. You may feel that some items used to apply, but do not now. We only want to know about the last 6 months.”

1. How involved are you in your neighbourhood?

- Not at all
 - Somewhat
 - Very Involved
 - Other (please explain).....
-

2. How satisfied are you with this situation ?

- Very dissatisfied (I wish things were very different)
 - Somewhat dissatisfied (I would like some changes)
 - Somewhat satisfied (OK for now, pretty good)
 - Very satisfied (I'm really pleased)
 - Other (please explain).....
-

3. Are there any organised groups that are a source of support for you?

- None
 - Some
 - Many
 - Other (please explain).....
-

4. How satisfied are you with this situation ?

- Very dissatisfied (I wish things were very different)
 - Somewhat dissatisfied (I would like some changes)
 - Somewhat satisfied (OK for now, pretty good)
 - Very satisfied (I'm really pleased)
 - Other (please explain).....
-

5. Think of a typical week. About how many times did you talk on the phone with your friends or family ?

- No talks
- 1 talk
- 2 -3 talks
- 4 - 7 talks
- More than 7 talks
- Other (please explain).....

6. How satisfied are you with this amount of telephone contact ?

- Very dissatisfied (I wish things were very different)
- Somewhat dissatisfied (I would like some changes)
- Somewhat satisfied (OK for now, pretty good)
- Very satisfied (I'm really pleased)
- Other (please explain).....

7. In the last week, how many times have you been visited by your friends?

8. How satisfied are you with this amount of visiting ?

- Very dissatisfied (I wish things were very different)
- Somewhat dissatisfied (I would like some changes)
- Somewhat satisfied (OK for now, pretty good)
- Very satisfied (I'm really pleased)
- Other (please explain).....

9. If you were to become angry or upset, would you have someone to talk honestly to, who is not involved? How many people?

- No-one
- 1 person
- 2 people
- 3 - 4 people
- More than 4 people
- Other (please explain).....

10. How satisfied are you with this ?

- Very dissatisfied (I wish things were very different)
- Somewhat dissatisfied (I would like some changes)
- Somewhat satisfied (OK for now, pretty good)
- Very satisfied (I'm really pleased)
- Other (please explain).....

(From: The Inventory of Parent's Experiences (Crnic & Greenberg, 1983))

11. When you are happy, is there someone you can share it with - someone who will be happy just because you are ?

- No
- Yes
- Other (please explain).....

12. How satisfied are you with this situation ?

- Very dissatisfied (I wish things were very different)
- Somewhat dissatisfied (I would like some changes)
- Somewhat satisfied (OK for now, pretty good)
- Very satisfied (I'm really pleased)
- Other (please explain).....

13. Do you currently have a relationship with a spouse or partner ? Do you expect it will continue for the years to come ?

- I don't have a relationship
- I don't expect the relationship to last
- I feel the relationship probably will last
- I feel the relationship definitely will last
- Other (please explain).....

14. How satisfied are you with this situation ?

- Very dissatisfied (I wish things were very different)
- Somewhat dissatisfied (I would like some changes)
- Somewhat satisfied (OK for now, pretty good)
- Very satisfied (I'm really pleased)
- Other (please explain).....

15. At present, do you have someone you can share your most private feelings with ?

- No
- Yes
- Other (please explain).....

16. How satisfied are you with this situation ?

- Very dissatisfied (I wish things were very different)
- Somewhat dissatisfied (I would like some changes)
- Somewhat satisfied (OK for now, pretty good)
- Very satisfied (I'm really pleased)
- Other (please explain).....

(From: The Inventory of Parent's Experiences (Crnic & Greenberg, 1983))

OBJECTIVE BURDEN INDEX
 PEDIATRIC AMBULATORY CARE TREATMENT STUDY
 Albert Einstein College of Medicine
 Bronx, New York 10461

	No = 0 Yes = 1	Coding value	0 if No, coding value if Yes
Does _____ (CHILD) have any of the following conditions?			
1) impaired vision?			
IF YES: a) Is the child blind?	<input checked="" type="checkbox"/>	20	<input type="checkbox"/>
	(Col. 18)		
b) Does the child wear glasses? ...	<input type="checkbox"/>	1	<input type="checkbox"/>
	(Col. 19)		
2) impaired hearing?			
IF YES: a) Is the child deaf?	<input type="checkbox"/>	17	<input type="checkbox"/>
	(Col. 20)		
b) Does the child use a hearing aid?	<input type="checkbox"/>	3	<input type="checkbox"/>
	(Col. 21)		
3) limited use of or absence of upper limb?	<input type="checkbox"/>	16	<input type="checkbox"/>
	(Col. 22)		
IF YES: a) Does the child use a prosthesis?	<input type="checkbox"/>	10	<input type="checkbox"/>
	(Col. 23)		
4) limited use of or absence of lower limb?	<input type="checkbox"/>	16	<input type="checkbox"/>
	(Col. 24)		

IF YES: Do any of the following apply?

(HAND RESPONDENT CARD A)

a) child uses a wheelchair	<input type="checkbox"/>	9	<input type="checkbox"/>
	(Col. 25)		
b) child uses braces	<input type="checkbox"/>	4	<input type="checkbox"/>
	(Col. 26)		
c) child uses crutches	<input type="checkbox"/>	4	<input type="checkbox"/>
	(Col. 27)		
d) child uses prosthesis	<input type="checkbox"/>	10	<input type="checkbox"/>
	(Col. 28)		
e) mobility is limited	<input type="checkbox"/>	13	<input type="checkbox"/>
	(Col. 29)		
f) child cannot use public transportation ..	<input type="checkbox"/>	12	<input type="checkbox"/>
	(Col. 30)		

PAGE TOTAL

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Does _____ (CHILD) have any of the following conditions?

(HAND RESPONDENT CARD B)

	No = 0 Yes = 1	Coding value	0 if No, coding value if Yes
5) recurrent infections?	<input type="checkbox"/>	10	<input type="checkbox"/>
	(Col. 31)		
6) addisonian crisis?	<input type="checkbox"/>	17	<input type="checkbox"/>
	(Col. 32)		
7) seizures?	<input type="checkbox"/>	7	<input type="checkbox"/>
	(Col. 33)		
8) abnormal appearance?	<input type="checkbox"/>	16	<input type="checkbox"/>
	(Col. 34)		
9) mild retardation?	<input type="checkbox"/>	10	<input type="checkbox"/>
	(Col. 35)		
10) severe retardation?	<input type="checkbox"/>	22	<input type="checkbox"/>
	(Col. 36)		
11) a genetic, familial, or congenital disease?	<input type="checkbox"/>	16	<input type="checkbox"/>
	(Col. 37)		
<u>IF CHILD IS OVER 3</u>			
12) lack of bladder control?	<input type="checkbox"/>	13	<input type="checkbox"/>
	(Col. 38)		
13) lack of bowel control?	<input type="checkbox"/>	14	<input type="checkbox"/>
	(Col. 39)		
14) is behavior sufficiently abnormal to be obvious in casual contact with an untrained person?	<input type="checkbox"/>	16	<input type="checkbox"/>
	(Col. 40)		
15) is surgery planned?	<input type="checkbox"/>	12	<input type="checkbox"/>
	(Col. 41)		
16) are child's language skills inappropri- ately developed for his/her age?	<input type="checkbox"/>	12	<input type="checkbox"/>
	(Col. 42)		
17) is motor strength inappropriate for child's age?	<input type="checkbox"/>	12	<input type="checkbox"/>
	(Col. 43)		
		PAGE TOTAL	<input type="checkbox"/>

18) Which one of the following best applies --

(HAND RESPONDENT CARD C)

	No = 0 Yes = 1	Coding value	0 if No, coding value if Yes
a) full recovery is anticipated	<input type="checkbox"/>	2	<input checked="" type="checkbox"/>
	(Col. 44)		
b) improvement is anticipated but not full recovery	<input type="checkbox"/>	7	<input type="checkbox"/>
	(Col. 45)		
c) condition is expected to remain the same	<input type="checkbox"/>	12	<input type="checkbox"/>
	(Col. 46)		
d) condition is expected to get somewhat worse	<input type="checkbox"/>	18	<input type="checkbox"/>
	(Col. 47)		
e) significant deterioration is expected ...	<input type="checkbox"/>	23	<input type="checkbox"/>
	(Col. 48)		
f) child is expected to die before adulthood	<input type="checkbox"/>	24	<input type="checkbox"/>
	(Col. 49)		
g) prognosis is unpredictable or unknown ...	<input type="checkbox"/>	20	<input type="checkbox"/>
	(Col. 50)		
19) Is child expected to have a life long dependency on another for daily care?	<input type="checkbox"/>	24	<input type="checkbox"/>
	(Col. 51)		
20) Is child expected to have a life long dependency on daily <u>self</u> care?	<input type="checkbox"/>	14	<input type="checkbox"/>
	(Col. 52)		

PAGE TOTAL

21) Is the child able to do age appropriate . . .

(HAND RESPONDENT CARD D)

	NOTE <u>REVERSAL</u>	No = 1 Yes = 0	Coding value	Transfer coding value if 1st column = 5
feeding	No = 1 Yes = 0	<input type="checkbox"/>	5	<input type="checkbox"/>
		(Col. 53)		
dressing	No = 1 Yes = 0	<input type="checkbox"/>	5	<input type="checkbox"/>
		(Col. 54)		
bathing	No = 1 Yes = 0	<input type="checkbox"/>	5	<input type="checkbox"/>
		(Col. 55)		
toileting	No = 1 Yes = 0	<input type="checkbox"/>	7	<input type="checkbox"/>
		(Col. 56)		

ASK ONLY IF CHILD IS OVER 3

22) Does he/she play with other children?	No = 1 Yes = 0	<input type="checkbox"/>	16	<input type="checkbox"/>
		(Col. 57)		

ASK ONLY IF CHILD IS OVER 5

23) Does he/she attend school?	No = 1 Yes = 0	<input type="checkbox"/>	15	<input type="checkbox"/>
		(Col. 58)		

IF YES:

	NOTE <u>REVERSAL</u>	No = 0 Yes = 1		
a) special school	No = 0 Yes = 1	<input type="checkbox"/>	8	<input type="checkbox"/>
		(Col. 59)		

PAGE TOTAL

MEDICATIONS

Are the parents expected to administer or supervise any type of medications . . .

(HAND RESPONDENT CARD E)

0 if No,
coding
value
if Yes

IF YES, HOW FREQUENTLY?

No = 0
Yes = 1

Coding
rule

- | | | | |
|------------------------|--------------------------|--|--------------------------|
| 24) a) by vein? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| | (Col. 60) | 11 | |
| b) by injection? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| | (Col. 61) | ≤ once/week = 4
1/week-1/day = 7
≥ once/day = 10 | |
| c) by mouth? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| | (Col. 62) | ≤ twice/day = 2
> twice/day = 5 | |
| d) in the eye? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| | (Col. 63) | ≤ twice/day = 2
> twice/day = 5 | |
| e) on the skin? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| | (Col. 64) | ≤ twice/day = 2
> twice/day = 6 | |
| f) an inhalant? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| | (Col. 65) | ≤ twice/day = 2
> twice/day = 6 | |

IF NO TO ALL THE ABOVE, GO TO QUESTION 31

- | | | | |
|---|--------------------------|--------------------------|--------------------------|
| 25) Are medications adjusted by physician or nurse more than once a month? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| | (Col. 66) | 6 | |
| 26) Does parent or child have to regulate the medications? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| | (Col. 67) | 9 | |
| 27) Does parent or child have to be alert for special risks: reactions, toxicity? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| | (Col. 68) | 10 | |
| 28) Are medications experimental? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| | (Col. 69) | 15 | |
| 29) Is the child's appearance altered by the medication? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| | (Col. 70) | 14 | |
| 30) Are medications given on different schedules? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| | (Col. 71) | 7 | |
| | <input type="checkbox"/> | | <input type="checkbox"/> |
| | (Col. 80) | 1 | |

PAGE TOTAL

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BEGIN CARD 2

TREATMENTS

31) Are parents expected to monitor child's . . .

(HAND RESPONDENT CARD F)

<u>IF YES, HOW FREQUENTLY?</u>	No = 0 Yes = 1	Coding rule	0 if No, coding value if Yes
a) blood pressure?.....	<input type="text"/> (Col. 7)	\leq twice/day = 2 $>$ twice/day = 6	<input type="text"/>
b) pulse?	<input type="text"/> (Col. 8)	\leq twice/day = 2 $>$ twice/day = 6	<input type="text"/>
c) respirations?	<input type="text"/> (Col. 9)	\leq twice/day = 2 $>$ twice/day = 5	<input type="text"/>
d) temperature?	<input type="text"/> (Col. 10)	\leq twice/day = 1 $>$ twice/day = 4	<input type="text"/>

32) Will they need to do . . .

a) postural drainage?	<input type="text"/> (Col. 11)	\leq twice/day = 4 $>$ twice/day = 10	<input type="text"/>
b) urine testing?	<input type="text"/> (Col. 12)	\leq twice/day = 2 $>$ twice/day = 5	<input type="text"/>
c) collection of urine for hospital assay?	<input type="text"/> (Col. 13)	\leq once/week = 2 1/week-1/day = 4 \geq once/day = 6	<input type="text"/>
d) trach change?	<input type="text"/> (Col. 14)	\leq once/week = 8 1/week-1/day = 11 \geq once/day = 14	<input type="text"/>

PAGE TOTAL

e) suctioning or trach care? \leq twice/day = 7
 $>$ twice/day = 13
(Col. 15)

f) stoma care? \leq once/week = 5
 1/week-1/day = 8
 \geq once/day = 11
(Col. 16)

g) dressing change? \leq once/week = 3
 1/week-1/day = 5
 \geq once/day = 8
(Col. 17)

h) passage of feeding tube? \leq once/week = 9
 1/week-1/day = 12
 \geq once/day = 16
(Col. 18)

i) feeding through a tube? \leq twice/day = 5
 $>$ twice/day = 10
(Col. 19)

j) crede? 8
(Col. 20)

k) physical therapy? 7
(Col. 21)

l) shunt testing? 7
(Col. 22)

m) care for wound or decubitis? 9
(Col. 23)

PAGE TOTAL

33) Do they:

a) have to learn to respond to emergencies (e.g., vagal massage, recognition and treatment of hypoglycemia)? 11

(Col. 24)

b) need to provide a special diet? ... 5

(Col. 25)

c) need to use a stethoscope?
IF YES, HOW FREQUENTLY? _____ \leq twice/day = 2
 $>$ twice/day = 5

(Col. 26)

d) use respiratory equipment?
IF YES, HOW FREQUENTLY? _____ \leq twice/day = 6
 $>$ twice/day = 13

(Col. 27)

e) have to expect periodic changes in child's clinical condition, i.e., sickle cell crisis, asthma attacks, relapses of nephrotic syndrome? 15

(Col. 28)

34) Do medications or treatments administered by the parents cause the child any pain? 14

(Col. 29)

35) Will the parents need to keep records?
IF YES, HOW FREQUENTLY? _____ \leq twice/day = 4
 $>$ twice/day = 9

(Col. 30)

36) Does the child have to come to clinic regularly? IF NO, GO TO QUESTION 39
IF YES, HOW FREQUENTLY? _____ \geq twice/week = 14
 \geq once/month = 5

(Col. 31)

37) Does the child have to come for special treatment once a week or more often than that? 11

(Col. 32)

PAGE TOTAL

38) Do clinic visits or treatments require:

a) rearrangement of parent's work schedule? 12
(Col. 33)

b) special babysitting? 9
(Col. 34)

~~c)~~ frequent absences from work or school? 17
(Col. 35)

39) Will the child need to spend all or part of a day in the hospital for special treatments? 17
(Col. 36)

40) Does the child's condition necessitate frequent admissions to the hospital's in-patient service? 21
(Col. 37)

41) Does child's condition necessitate a change in the home environment? 8
(Col. 38)

42) Does the illness require that the child be left only with a person prepared for emergencies or with a person prepared to do special procedures? 17
(Col. 39)

43) Does the illness require a change in parental sleeping patterns for night time supervision? 11
(Col. 40)

44) Is care divided between two or more medical facilities? 13
(Col. 41)

2 PAGE TOTAL
(Col. 80)

APPENDIX 5:

Well Sibling Questionnaire

SIBLING PERCEPTION QUESTIONNAIRE (SIBLING VERSION)

Interview Number:

COMPLETED BY RESEARCHER

"Here are some things other children have told us about how their sister's or brother's illness affects them. You may feel that some did apply when you were younger, but not now. We want you to tell us which of these thoughts you have had in the last six months"

<u>I wish there was something I could do about my brother's or sister's illness</u>	<u>YES</u>	<u>NO</u>
<u>I wish my parents would spend less time with my brother or sister</u>	<u>YES</u>	<u>NO</u>
<u>I wish my parents would spend more time with me</u>	<u>YES</u>	<u>NO</u>
<u>I feel sad about my brother or sister or being ill</u>	<u>YES</u>	<u>NO</u>
<u>People are more interested in my brother or sister than me</u>	<u>YES</u>	<u>NO</u>
<u>Because of my brother's /sister's illness, people don't care how I feel</u>	<u>YES</u>	<u>NO</u>
<u>I think about my brother's or sister's illness</u>	<u>YES</u>	<u>NO</u>
<u>Because of my brother's /sister's illness, my parents ignore me</u>	<u>YES</u>	<u>NO</u>
<u>Because of my brother's /sister's illness, I have too much to do in the house</u>	<u>YES</u>	<u>NO</u>
<u>I wonder my brother or sister got sick</u>	<u>YES</u>	<u>NO</u>
<u>I worry that I can catch the illness from my brother or sister</u>	<u>YES</u>	<u>NO</u>
<u>I don't want to bother my parents with my worries</u>	<u>YES</u>	<u>NO</u>
<u>I feel angry about my brother or sister being ill</u>	<u>YES</u>	<u>NO</u>
<u>I am afraid of my brother's or sister's illness</u>	<u>YES</u>	<u>NO</u>
<u>I wish I knew someone who understands how I feel</u>	<u>YES</u>	<u>NO</u>
<u>I understand why my parents have to spend more time with my brother or sister</u>	<u>YES</u>	<u>NO</u>
<u>My friends worry that they can catch the illness from my brother or sister</u>	<u>YES</u>	<u>NO</u>
<u>I can forget that my brother or sister is ill</u>	<u>YES</u>	<u>NO</u>
<u>Because of my brother's /sister's illness, we don't do much as a family</u>	<u>YES</u>	<u>NO</u>
<u>I cannot talk to my friends about my brother's /sister's illness</u>	<u>YES</u>	<u>NO</u>
<u>I cannot talk to my parents about my brother's /sister's illness</u>	<u>YES</u>	<u>NO</u>
<u>I cannot talk to my parents about my school work</u>	<u>YES</u>	<u>NO</u>
<u>I cannot talk to other adults about my brother's /sister's illness</u>	<u>YES</u>	<u>NO</u>

(Adapted from: Sibling Perception Questionnaire of Illness Experience (Carpenter & Sahler, 1991))