Psychological adjustment and health related quality of life in children who have been diagnosed with acute lymphoblastic leukaemia

Thesis

Psychological Adjustment and Health Related Quality of Life in children who have been diagnosed with Acute Lymphoblastic Leukaemia

Caroline Paul, B.Sc.

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ABSTRACT

Objectives: This study aimed to: a) investigate children's adjustment and Health Related Quality of Life (HRQL) in Acute Lymphoblastic Leukaemia (ALL) and b) identify factors associated with, and predicting, adjustment and HRQL in children with ALL. Wallander and Varni's (1992) model of child adjustment was used as a theoretical framework.

Design: A cross-sectional, within-subjects, questionnaire survey design was used.

Method: Forty-four parents and 28 children participated. Parents completed questionnaires assessing child's adjustment and HRQL. A parenting stress measure, to collect data on likely predictors, and a semi-structured interview was administered to parents. Children aged 5-12 years completed a questionnaire assessing HRQL.

Results: ALL was associated with poor adjustment and poor HRQL. Demographic, treatment status, child and parent characteristics and life stress were associated with adjustment and/or HRQL. Child characteristics (i.e. distractability/ hyperactivity, adaptability), gender and parent characteristics were significant predictors of adjustment. Number of siblings, parental isolation and treatment status significantly predicted child-rated HRQL, while child characteristics (i.e. acceptability, mood, adaptability), treatment status, age at diagnosis and life stress predicted parent-rated HRQL.

Conclusion: Results suggest child characteristics, parent characteristics and treatment status, in particular, are important predictors of adjustment and HRQL in ALL. The findings are discussed in relation to previous research, methodological weaknesses and the possible role of cross informant variance. Implications for clinical research, the development of theory and future research are outlined.

ACKNOWLEDGEMENTS

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1. INTRODUCTION

1.1 OVERVIEW OF ALL

This study investigated psychological adjustment and quality of life in children diagnosed with Acute Lymphoblastic Leukaemia (ALL). The introduction will give an overview of ALL, including its characteristics, treatment, survival rates and the implications of these. Research into psychological impact of childhood cancer in terms of adjustment and Health Related Quality of Life (HRQL) is reviewed. A model of child adjustment, used in paediatric chronic physical conditions is introduced; this identifies risk and resistance factors and has been influential in research into child adjustment. Interest in HRQL has increased, with the acknowledgement that more comprehensive measures of outcome are needed for this group of children[1]. Currently literature on HRQL lacks an overarching theoretical framework, however there is a general agreement that HRQL is a multidimensional construct and this is outlined. Methodological limitations of research into adjustment and HRQL in childhood cancer are considered. The need for future research and the rationale for the present study are presented, with specific aims and hypotheses outlined.

1.1.1 Characteristics of ALL

ALL is a common malignancy of childhood, occurring at an annual rate of approximately 31 per million, and accounting for about 23% of cancer diagnoses

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[1] Traditionally the emphasis in outcome studies was on survival rates, however recently studies have focussed on the assessment of children's adjustment.

under the age of 15 years (Ries, Kosary, & Hankey, 1996). The peak incidence occurs from ages three to five years, with ALL affecting slightly more boys than girls (St Jude Children's Research Hospital, 2001). Early symptoms can be similar to those of flu (i.e. fever, feelings of weakness, tiredness, aching bones or joints and swollen lymph nodes). Diagnosis is established by bone marrow examination.

1.1.2 Treatments for All

Chemotherapy is the primary treatment for ALL and comprises of three phases:

a) Remission induction – chemotherapy is used to kill as many leukaemia cells as possible, with the aim of causing the cancer to go into remission.

b) Consolidation and central nervous system (CNS) prophylaxis - chemotherapy consolidates remission and prevents leukaemia cells spreading to the brain and spinal cord.

c) Maintenance therapy - given as an outpatient and lasting 20 months for girls and 36 months for boys, resulting in the whole programme lasting two to three years, respectively. Boys receive longer treatment due to slightly higher risk of relapse.

Trials have shown that cranial irradiation causes long-term cognitive impairments and disruptions in growth; it is therefore no longer the standard treatment for childhood cancer (Eiser, 1998).

During chemotherapy children may experience side effects, for example, alopecia, nausea, skin complaints, sleep disturbance and changes in mood and behaviour.

1.1.3 Survival rates and implications

Given the best current therapy over 70% of children with ALL can expect long-term survival[2], and in the most favourable prognostic sub-groups, the figure rises to around 80% (Medical Research Council, 1999). Treatment outcome has previously focused upon survival rates (mortality and disease-free survival). Since prognosis has improved, it is important to assess children's adjustment and the impact of disease (and treatment) on different areas of functioning.

Increasingly the focus has been upon late physical (e.g. second cancers, abnormal growth, and cardiac dysfunction) and psychological (e.g. cognitive, specific neuropsychological and social functioning) consequences of childhood cancer. Eiser (1998) suggests 'for the most part, survivors must live with the knowledge that they are at a greater risk of a variety of physical problems and there is very little that can be done' (p. 624). Little is known about how survivors react to such information, and the effect on self-esteem and decisions about life-style. Regular follow-ups may be necessary to identify individuals requiring medical, psychological, educational and social interventions.

1.2 PSYCHOLOGICAL IMPACT OF CHILDHOOD CANCER AND HRQL

Many studies have focussed on how children 'adjust' to cancer. Unfortunately, however, the concepts of 'adjustment', 'adaptation', 'coping', 'stress' and 'competence' are used interchangeably in such studies (Rutter, 1981). In addition,

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[2] Long-term survivors are those surviving from diagnosis to five years from diagnosis without recurrence. After five years it is unlikely that ALL will reoccur and is therefore regarded as a 'cure'.

there is often no clear distinction between maladjustment in terms of emotional, behavioural or psychosocial problems (Pless & Stein, 1996).

Similar problems have begun to emerge in the HRQL literature, where there is 'little consensus regarding the appropriate measures, which include assessment of self-esteem, anxiety, depression, social skills, body image...' (Eiser, Hill & Vance, 2000, p. 452). As a result there is confusion in the literature regarding the distinction between adjustment and HRQL. It was assumed in this study that adjustment and HRQL were overlapping constructs, with HRQL being a broader concept, including adjustment.

Definitions of the concepts, theoretical frameworks and empirical findings will be described below.

1.2.1 Adjustment

Adjustment of children with cancer has received considerable attention due to improvements in prognosis and the potentially damaging effects of treatment, with certain cancers (e.g. Hodgkin's disease and ALL) being studied more frequently (Eiser et al., 2000).

1.2.2 Definition

Good adjustment has been defined as:

> behaviour that is age-appropriate, normative, and healthy, and that
>
> follows a trajectory toward positive adult functioning.

While,

> maladjustment is mainly evidenced in behaviour that is
>
> inappropriate for the particular age, especially when this behaviour
>
> is qualitatively pathological or clinical in nature (Wallander &
>
> Thompson, 1995, p.125-126).

It has been suggested children with chronic disease (e.g. cancer, asthma, and juvenile rheumatoid arthritis) are more likely to show maladjustment than healthy children. Pless & Nolan (1991) found a two- to three-fold increased rate of psychological difficulties in children with chronic disease compared to healthy peers, while Eiser (1990) found an increased risk of adjustment problems with CNS involvement or physical disability.

Factors identified as affecting the experience of cancer in children include: physical appearance, interference with activity, peer rejection, integration with school, family support and relations, anxiety about symptoms and relapse, and impact of treatment (Eiser, Havermans, Craft, & Kernahan, 1995).

Childhood cancer has been shown to predispose certain individuals to post traumatic stress symptoms (Eiser, 1998). When compared to matched families, childhood leukaemia survivors suffered a disturbing (yet sub-diagnostic) cluster of anxiety symptoms (Stuber, Christakis, Houskamp, & Kazak, 1996).

Studies into adjustment in children with chronic diseases are now replacing traditional measures (i.e. psychiatric interview and symptom reports) with assessments of behaviour, self-concept, depression, competence, self-esteem and locus of control (Eiser, 1990).

1.2.3 Factors affecting adjustment

The effect of age on adjustment in children with chronic diseases has been reported, with young children experiencing separation anxiety and attachment difficulties. In children (under five years) requiring multiple admissions, an association between relatively brief hospitalisation and increased risk of later behavioural disturbance or delinquency was found (Quinton & Rutter, 1976). Eiser (1990) reviewed the literature and concluded, younger children were more affected with relation to school tasks and achievement, whilst older children experienced disrupted social adjustment.

Studies have found that children diagnosed with cancer at a young age, were less likely to suffer later adjustment difficulties (Koocher, O'Malley, Gogan & Foster, 1980). This may be due to both the child's lack of understanding of the seriousness of the illness, and the developmental tasks characteristic of middle childhood and adolescence being less affected.

As the time since onset of disease increases, many patients become less anxious about recurrence. However, Koocher et al. (1980) found a mixed group of paediatric cancer survivors suffered residual psychosocial sequelae (ranging from 'mild-to-substantial'), with symptoms of depression, anxiety and poor self-esteem. Those patients able to articulate reasons for this described uncertainty about the future, fear of recurrence, and inability to 'forget' stressful aspects of treatment.

A review by Eiser et al. (2000) found few differences between childhood cancer survivors and population norms on standardized measures of anxiety, depression and self-esteem. The only study reviewed to find more symptoms in survivors (of bone tumours) than norms, identified problems relating to physical functioning, physical role performance, pain, general health and social functioning (Eiser, Cool, Grimer, Carter, Ellis, Kopel & Eiser, 1997).

In summary, findings suggest that while most survivors do relatively well, a subset suffer more serious adjustment difficulties (Kazak, 1994). It is important to identify these individuals (both during and after treatment) and establish what factors influence maladjustment. This will have both theoretical and clinical implications, enabling the provision of preventative and therapeutic interventions.

1.2.4   Model of Child Adjustment

Wallander and Varni's conceptual model (1992, adapted from Wallander, Varni, Babani, Banis & Wilcox, 1989) has been influential in this area of research (Figure 1). A non-categorical approach (i.e. commonalities between diseases are greater than their differences) is proposed, and paediatric chronic physical disorders are conceptualised

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as a constant strain for children and their parents. It was suggested that modifiable risk and resistance factors could be empirically identified, thereby providing heuristic guidance for the development of interventions. For example, Quiggins (1996) found that perceived stress and perceived social support had affected adjustment, and therefore suggested implementing interventions aimed at reducing perceived stress (e.g. relaxation) and increasing perceived social support (e.g. social skills training). However, Pless & Stein (1996) purport that 'few findings are sufficiently clear-cut to permit the identification of clinical sub-groups with the precision needed to serve as a basis for more efficient intervention strategies' (p.331).

Risk factors, identified in the model, include: disease/disability parameters, functional independence in activities of daily living and psychosocial stressors, while personal characteristics (e.g. temperament), social-ecological variables and stress processing (e.g. coping strategies) were resistance factors. Due to the model's complexity and the low incidence of chronic physical disorders, most researchers have tested components or detailed sub-models of the framework. Wallander, Pitt & Mellins (1990) argue that it is not possible to validate the model as a whole, since it is only feasible to analyse single or small groups of variables to see if they operate in the hypothesised direction.

Figure 1: Wallander and Varni's (1992) model of child adjustment. Red boxes indicate risk factors; blue boxes indicate resistance factors.
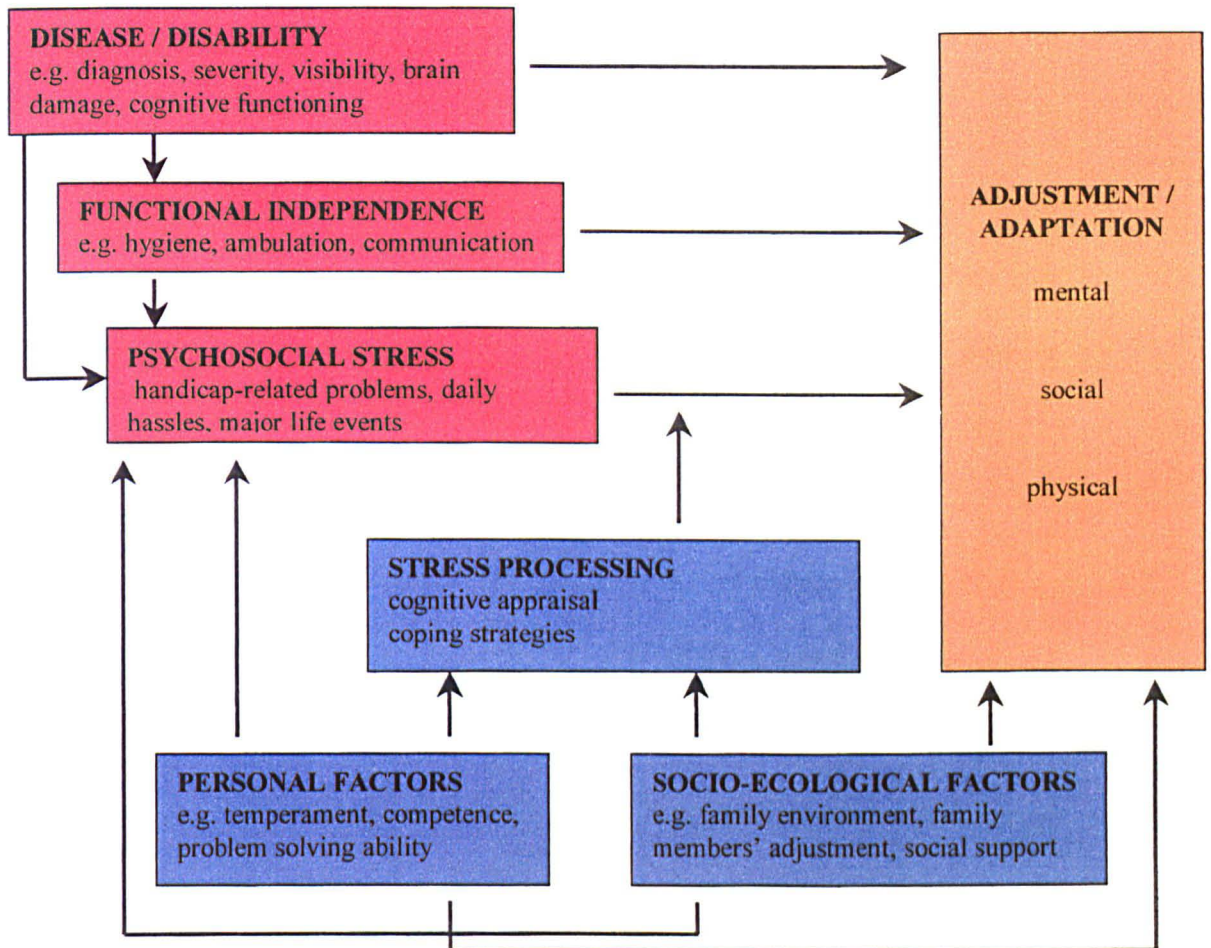
1.2.5 Empirical findings on child adjustment in cancer

1.2.5.1 Risk factors

Research examining risk factors have failed to find consistent association between disease/disability factors and adjustment, suggesting it is not the most influential factor. Age (at diagnosis) and time since diagnosis were not found to be associated with adjustment in a large study of paediatric cancer patients (Quiggins & Varni, 1996). A

study by Varni, Katz, Colegrove, & Dolgin (1995a) found cancer diagnosis (leukaemia versus other cancers) was unrelated to any adjustment dimension in newly diagnosed paediatric patients. However, diagnosis correlated with negative affectivity nine months post-diagnosis and with maladjustment in long-term leukaemia survivors (Varni & Katz, 1997).

Varni, Katz, Friedman-Bender & Quiggins (1996) found paediatric cancer patients on-treatment suffered more problems with functional independence than children off-treatment; these problems were associated with emotional distress and somatic symptoms (Varni, Katz, Quiggins & Friedman-Bender, 1996).

Quiggins & Varni (1996) found the third risk factor, psychosocial stress (e.g. perceived disease-related stress), to be associated with higher negative affectivity and total behaviour problems in children with cancer. Varni and Katz (1997) suggest it may take children up to nine months (after diagnosis) to return to some semblance of normal life, and for daily hassles to become more salient than major life events associated with the diagnosis and treatment of cancer.

1.2.5.2 Resistance factors

Research has found family functioning (i.e. a social-ecological factor), specifically, higher family cohesion and expressiveness, to predict better adjustment (Varni, Katz, Colegrove, & Dolgin, 1996) in children with newly diagnosed cancer. Perceived classmate social support was also associated with lower depressive symptoms, lower state, trait and social anxiety, reduced internalising and externalising behaviour

problems and higher levels of general self-esteem (Varni, Katz, Colegrove, & Dolgin, 1994b).

Few studies have investigated personal factors (e.g. personality characteristics) and stress processing in the adjustment of these children. However, disease-related stress processing (e.g. the child's perception of physical appearance) has been shown to relate to depressive symptoms, social anxiety and self-esteem (Varni, Katz, Colegrove, & Dolgin, 1995b).

Demographic factors were excluded from the model, since they failed to account for significant variance in children's adjustment (Wallander & Varni, 1992).

In summary, evidence supports functional independence and psychosocial stress as risk factors, affecting adjustment in childhood cancer, while social-ecological factors (e.g. family functioning) serve to protect against maladjustment.

1.2.6 HRQL

As survival rates in childhood cancer improve, there is increasing recognition that more sensitive, comprehensive measures of outcome are required (Eiser & Jenney, 1996). Previous studies assessing adjustment (traditionally psychiatric disturbance) have focussed upon a relatively narrow area of children's lives. A growing interest in the broader functioning of children with cancer has resulted in research investigating HRQL.

1.2.7 Definition & Theoretical framework for HRQL

Feeny, Furlong, Mulhern, Barr & Hudson (1999) defined HRQL as:

> concerned with the opportunities that a person's health status affords, the constraints that it places upon the person and the value that person places on his or her health status (p.2).

While the HRQL literature lacks an overarching theoretical framework (Gill & Feinstein, 1994), it is generally agreed to be a multi-dimensional construct. The World Health Organization (1947) identified physical, mental and social dimensions in its definition of health. Aaronson (1991) expanded this and suggested that physical, psychological and social functioning, in addition to, disease- and treatment-related symptoms were the 'core' set of domains in a HRQL measure. An example of treatment-related symptoms, perceived physical appearance, was shown to be important in paediatric oncology patients' experience of alopecia (Varni & Setoguchi, 1991).

A debate surrounds the advantages of disease-specific and generic instruments (i.e. those including 'core' domains) in HRQL. The former may allow a more sensitive measurement of problem-specific areas for different patient groups, while generic instruments enable comparisons across groups and with healthy controls. Some measures (e.g. the Peds QL, Varni, Seid & Rode, 1999) integrate generic and disease/symptom-specific approaches.

A bio-behavioural model (Varni, Katz, Seid, Quiggins, Friedman-Bender & Castro, 1998), derived from Wallander and Varni's (1992) model (described above), was influential in the development of the Peds QL (Varni, Seid & Rode, 1999).

1.2.8 Measurement of HRQL

Interest in HRQL in adult cancer patients has resulted in the development of a number of measures with good psychometric properties (Goodwin, Boggs, & Graham-Pole, 1994). However, in 1996, Bradlyn and Pollock reported few HRQL measures were appropriate for use in paediatric cancer trials.

Two examples of widely used cancer-specific measures are described below. The Play Performance Scale for Children (Lansky, List, Lansky, Cohen, & Sinks, 1995) measures only functional status and therefore provides a crude measure of HRQL. A more comprehensive measure, the parent-report Pediatric Oncology Quality of Life Scale (Goodwin et al., 1994), has physical status, emotional status and treatment-related domains, but no parallel form exists for children.

A recently developed measure, the Peds QL (Varni, Seid & Rode, 1999), has generic (i.e. physical, emotional, social & school) and disease/symptom-specific modules (e.g. nausea, perceived physical appearance), allowing comparisons to be made with healthy groups, and measurement of cancer-specific problems. It has been shown to have good psychometric properties and has parallel child and parent forms. The questionnaire is problem-focussed, with it being assumed that health-related problems can be overcome by combining bio-medical and bio-behavioural (e.g. cognitive-behavioural) interventions.

At the International Workshop on assessing HRQL in children with cancer it was suggested that in the future:

A key new step is the demonstration of the usefulness of HRQL measures in paediatric oncology, evidence that using HRQL measures helps investigators and clinicians to achieve their goals of evaluating new therapies and treating patients ......and in the short-term, the focus of HRQL research should be on consolidating the advances that have been made already and on the assessment of the relative strengths and weaknesses, measurement properties and usefulness of existing approaches (Feeny, Barr, Furlong, Hudson, & Mulhern, 1999, p.154).

Mulhern, Horowitz, Ochs, Friedman, Armstrong, Copeland, & Kun (1989) highlighted the need for normative data for children at various stages during and after treatment, to enable the interpretation of HRQL scores.

It is hoped that the development of HRQL measures will enable identification of patients requiring psychosocial interventions, aid clinical decision-making, and allow comparisons to be made among alternative therapies in clinical trials (Pollock, 1999). Such instruments could be used to identify acute psychosocial difficulties secondary to illness or treatment and residual problems in long-term survivors (Spieth & Harris, 1996).

1.2.9   Empirical findings on HRQL

Due to the focus on developing HRQL measures, few studies have assessed HRQL in paediatric populations. Goodwin et al. (1994), however, found children recently diagnosed with cancer had lower overall QOL, poorer physical functioning and more physical discomfort, than children diagnosed more than 30 months previously, or children in remission/off-treatment. Varni, Seid & Rode (1999) also found physical functioning and disease-related scores were poorer for children on-treatment for cancer (ALL accounted for 44% of the sample) than those off-treatment, consistent with findings obtained in studies investigating adjustment in this group.

Varni, Seid, & Kurtin (1999) used the Peds QL measure to compare healthy children, with children suffering acute health conditions (i.e. inpatients/outpatients at hospital or community clinics at least three months previously) and chronic health conditions (i.e. children who had attended speciality clinics for orthopaedics, cardiology, rheumatology and diabetes). The authors found healthy children had better HRQL than children with acute and chronic health conditions.

Finally, evidence of cross-informant variance (i.e. lack of agreement among reporters) by Varni, Seid & Rode (1999) highlighted the importance of using child-report measures.

1.3   SUMMARY OF LITERATURE REVIEW

Adjustment in childhood cancer has received a lot of attention due to increased survival rates and greater awareness of adverse treatment effects. Wallander and Varni's (1992) model of child adjustment has been influential in generating research

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in this area, with studies highlighting the importance of risk (e.g. functional independence) and resistance factors (e.g. social ecological factors). Due to the model's complexity most studies have tested components or sub-models of the framework.

A more recent area of interest in childhood cancer is that of HRQL. The current literature lacks an overarching theoretical framework, although it is accepted that HRQL is multidimensional, with physical, psychological and social functioning, and disease/treatment-related symptoms forming the 'core' domains.

Many studies describe the development of various instruments assessing HRQL but until recently these had not been shown to have acceptable psychometric properties. The development of such a measure (i.e. the Peds QL), designed for paediatric chronic health conditions, with generic and cancer-specific modules is an exciting and promising advance, enabling investigation of HRQL across age groups and the comparison of child- and parent-ratings.

1.4   METHODOLOGICAL CRITICISMS

Overall, studies evaluating adjustment and HRQL in children with cancer are variable in their methodological adequacy (many suffering from problems outlined below). The author is unaware of any studies focussing on both adjustment and the broader concept of HRQL in children with ALL, nor of a study examining predictors of HRQL in this area.

Many studies have not used systematic screening instruments in the measurement of adjustment (Varni & Setoguchi, 1992); without these children's emotional and behavioural problems are likely to be under-diagnosed and under-treated. Until recently no validated, generic paediatric HRQL instrument, with self- and proxy-report, had been designed for paediatric chronic health conditions.

Problems with previous research include limited research on young children, reliance on proxy reporting (problematic due to cross-informant variance), mixed diagnostic patient groups (important to distinguish due to different treatments and sequelae associated with different types of cancers, Eiser et al., 2000), and poor descriptive data (e.g. no information on time since diagnosis, Eiser et al., 2000). Finally, few studies investigated the types of problems (beyond those which are psychiatric and behavioural in nature) children with cancer present with, and most research has been conducted in the USA, with it being unclear to what extent these results are applicable to a British population (Bradford, 1997).

Assessment is complicated by developmental considerations, for example, the child's cognitive and language abilities at diagnosis and their developmental progress during treatment and recovery. Child-report measures need to provide reliable, valid and meaningful responses to be informative. Previously it was assumed young children were unable to provide such information. Recent studies (e.g. Varni, Seid & Rode, 1999) have shown this is not the case, and that parents are not necessarily the most reliable source of information about a child's well-being. Despite this few measures exist for young children.

The measurement of parent's ratings remains important, however, with it sometimes being the only measure available (e.g. when children are too young or too ill), and because of parents' influence on access, direction and priorities relating to their child's medical care (Parsons, Barlow, Levy, Supran, & Kaplan, 1999). Proxy-ratings have been found to be most accurate (i.e. similar to patient ratings) when the proxy and patient live in close proximity (Sprangers & Aaronson, 1992). Maternal distress, marital adjustment and health locus of control have been shown to co-vary with reports of child's behaviour (Parsons et al., 1999), and agreement among observers appears to be lower for internalizing problems (e.g. depression) than externalizing problems (e.g. hyperactivity) in children with cancer (Seid, Varni, Rode, & Katz, 1999).

It is important to acknowledge the limits of the correlational nature of most of this research (i.e. correlation does not prove causation). Wallander and Varni (1992), however, suggest that research with a strong conceptual basis, taking the form of model testing, can provide support for causal hypotheses using correlational findings.

Criticisms have also be raised regarding the measures used, for example, Perrin, Stein, & Drotar (1991) criticised the use of the Child Behaviour Checklist (CBCL, Achenbach & Edelbock, 1983) in the measurement of adjustment, due to it inclusion of items which directly tap physical health problems (e.g. 'wets self'), its insensitivity to mild adjustment problems, and for providing a 'social competence' score (which will be lower in children with chronic diseases due to restricted opportunities to participate in social activities).

Finally, confusion regarding the concepts of adjustment and HRQL has been mentioned. It is hoped by operationalising both concepts through the measures used in this study, this confusion will be minimized. In this study it was assumed adjustment and HRQL were overlapping constructs, with HRQL being a broader concept, including adjustment. The Wallander and Varni (1992) model will be used to guide the search for predictors of HRQL.

1.5 RATIONALE AND AIMS OF THIS STUDY

The study aimed to increase knowledge regarding the impact of ALL and its treatment on children, in terms of adjustment and HRQL and their predictors. Through identifying what predicts adjustment and HRQL, appropriate screening instruments could be used to detect children and parents at risk of developing problems and those requiring interventions.

This study examined adjustment in children using a behavioural screening questionnaire, the Extended Version of the Strengths and Difficulties Questionnaire, (SDQ, Goodman, 1999), thereby generating scores for conduct problems, hyperactivity-inattention, emotional symptoms, peer problems and pro-social behaviour.

The HRQL measure used (i.e. the Peds QL, Varni, Seid, Rode, 1999) included generic and disease-specific scales, enabling disease-sensitive data to be collected, while still allowing comparisons with healthy populations.

Currently there is a lack of research examining adjustment and HRQL in young children, and few studies include child-report HRQL measures. This study aimed to address these issues by investigating HRQL in children as young as two years of age, adjustment in children aged four years and over, and included a child-report measure of HRQL for children aged 5 years and over. It was hypothesised children with ALL would have poorer adjustment and HRQL when compared with healthy norms.

Diagnosis and treatment of ALL has major implications for both the child and the family. This study limited its focus to child adjustment and HRQL, since wider psychosocial considerations were beyond its scope. However, a semi-structured interview was administered. This asked parents about difficult aspects of their child's diagnosis/treatment and their perspective of available services. The interview enabled parents to discuss these issues, and others arising from the questionnaires, further.

The research used Wallander and Varni's (1992) framework to explore predictors of adjustment, and to indicate what factors may influence HRQL. In line with many other studies only a sub-model was tested (described below). This was felt appropriate due to the wide age range being studied and the importance of not burdening participants, particularly young children and children on-treatment.

Treatment status (i.e. whether children were on- or off-treatment) was included as a risk factor, due to previous empirical findings relating to functional independence (Varni et al., 1996). Despite demographic factors previously not being found to explain variance in children's adjustment, gender was included in this study. It was hypothesised that boys diagnosed with ALL might have poorer adjustment and HRQL,

due to them undergoing longer treatment. Few studies have reported an association between adjustment and age, however Koocher et al. (1980) found the younger the child at diagnosis, the fewer adjustment problems experienced later. Due to the current study focussing on young children and the use of a child-report measure, it was hypothesised that a relationship between age (at diagnosis and participation) and adjustment and HRQL might be found. Studies have shown psychosocial stress to be a risk factor in adjustment, therefore life stress (using the Parenting Stress Index (PSI, Abidin, 1995)) was also measured in this study.

Research into resistance factors found family functioning predicted better adjustment. Parent characteristics were measured (using the PSI, Abidin, 1995) and it was hypothesised they would affect adjustment and HRQL. Few studies have investigated stable personality characteristics in adjustment and HRQL in these children, so child characteristics (using the PSI, Abidin, 1995) were also measured. Due to the PSI measuring difficult child characteristics and dysfunctional parent characteristics, both were hypothesised to be associated with, and predict, poor adjustment and poor HRQL. Therefore these were risk factors in the current study.

Stress processing was not included in this study due to the difficulty of measuring cognitive appraisal and coping strategies in very young children and across the wide age group being studied.

The relationship between ethnicity, social economic status, number of siblings, marital status and disease severity and adjustment, and HRQL, were investigated.

1.6   RESEARCH QUESTIONS AND HYPOTHESES

Research Questions:

1. What is the association between ALL (and its treatment) and children's adjustment and HRQL?

2. What factors (i.e. demographic, disease and treatment variables, child and parental characteristics, and life stress) are associated with, and predict, adjustment and HRQL?

Hypotheses:

1. ALL and its treatment will be associated with poor adjustment and poor HRQL in children.

2. A range of demographic, disease and treatment factors will be associated with, and predict, poor adjustment and poor HRQL:

a) Children who are on-treatment will have poorer adjustment and HRQL than children off-treatment.

b) The relationship between gender and adjustment and HRQL will be explored.

It is suggested that boys will have poorer adjustment and HRQL, due to undergoing longer treatment.

c) The relationship between age (at diagnosis and participation) and adjustment, and HRQL will be explored.

Previous research has not found an association between adjustment and age but the current study's inclusion of young children and a child-report measure may uncover a relationship between age and adjustment and HRQL.

3. There will be poor adjustment and HRQL in those children who display difficult child characteristics.

4. There will be poor adjustment and HRQL in those children whose parent's characteristics make functioning as a competent caregiver difficult.

5. There will be poor adjustment and HRQL in those children who experience high levels of life stress.

2. METHOD

2.1 Ethical Approval

Approval was gained from the Multi-Centre Research Ethics Committee (MREC) and relevant local ethic committees (Appendix 1).

2.2 Design

The study employed a within-subjects design to examine adjustment and HRQL in children aged between 2 and 12 years, diagnosed with ALL. The study comprised a cross-sectional questionnaire survey design.

2.3 Participants

All participants were recruited through the Regional Paediatric Oncology/Haematology Unit. Participants were parents of children (2 - 12 years) and children (5 - 12 years) diagnosed with ALL. Exclusion criteria included: patients diagnosed or relapsed with ALL in the last 6 months, parents or children not fluent in English, and patients considered too ill by their doctors to participate in the study.

2.4 Measures

Parents of children (4 –12 years) completed the Extended Version of the Strengths and Difficulties Questionnaire (Goodman, 1999) (Appendix 2). Children (5 – 12 years) and parents of children (2 –12 years) completed the Peds QL measure (Varni, Seid & Rode, 1999). An example of a child-report (5-7 years) Peds QL questionnaire is shown in Appendix 3. All parents filled in the Parenting Stress Index (Abidin, 1995) (Appendix 4).

A semi-structured interview was administered (Appendix 5). This asked parents about difficult aspects of their child's diagnosis/treatment and their perspective of available services.

Demographic details (e.g. age, ethnic group, social economic status) and medical information (e.g. date of diagnosis, start of treatment, treatment protocol) were obtained from parents and medical notes. Patients were categorised by a Consultant Paediatric Oncologist into standard and high risk.

2.4.1 The Extended Version of the Strengths and Difficulties Questionnaire (SDQ, Goodman, 1999).

This measure of adjustment is a brief behavioural screening questionnaire for children, aged 4 to 16 years, asking about symptoms and positive attributes. The 25 items are divided between five scales, each having five items; generating scores for conduct problems, hyperactivity-inattention, emotional symptoms, peer problems, and pro-social behaviour.

The informant-rated version of the SDQ has been shown to function as well as the established, reliable and valid Rutter questionnaires and CBCL (Achenbach & Edelbock, 1983) at detecting conduct and emotional problems (Goodman, 1997 & Goodman & Scott, 1999). It has been found to be better than the CBCL at detecting inattention and hyperactivity (Goodman & Scott, 1999) and diagnostic predictions based on the SDQ have been shown to agree well with clinical diagnoses in psychiatric clinic samples (Goodman, Renfrew & Mullick, 2000b).

Parents were asked to rate statements relating to their child's behaviour over the last 6 months, or school year, as not true, somewhat true or certainly true. The banding of scores is 'normal', 'borderline' or 'abnormal' (Goodman, 1997). An impact supplement was included, which asks if the respondent thinks the young person has a problem, and if they do its chronicity, distress, social impairment and burden on others.

2.4.2 Peds QL measure (Varni, Seid, & Rode, 1999).

The Peds QL measure is a modular approach to measuring HRQL and can be used in healthy children and those with acute or chronic health conditions. The core scales are Physical (8 items), Emotional (5 items), Social (5 items) and School Functioning (5 items).

Supplementary cancer specific modules include Pain (2 items), Nausea (5 items), Procedural Anxiety (3 items), Treatment Anxiety (3 items), Worry (3 items), Cognitive Problems (5 items), Perceived Physical Appearance (3 items) and Communication with Physician/ Nurse (3 items).

Developmentally appropriate forms exist for children aged 2-4, 5-7, 8-12 years. Paediatric-report was measured in children from 5-12 years, while parent- reports exist for children aged 2-12 years. The instructions ask how much a problem each item has been during the past month. Children over 8 years of age and their parents are given a 5-point response scale ranging from 'never a problem', 'almost never a problem', 'sometimes a problem', 'often a problem', to 'a lot of a problem'. The young child-report (5-7 years) response scale is reworded and simplified to a 3-point

scale, ranging from 'not a problem', 'sometimes a problem' to 'a lot of a problem', with responses anchored to a happy/sad faces scale. The toddler age range (2-4 years) only has a parent-report due to developmental limitations on self-report for children less than 5 years of age, and includes just 3 items on school functioning.

Items are reverse-scored and linearly transformed to 0 - 100 scale (e.g. 0 = 100, 1 = 75, 2 = 50, 3 = 25, and 4 = 0), with higher scores indicating better HRQL. Scale scores are computed as the sum of items divided by the number of items answered. The following summary scores are calculated:

1. Total Scale Score (sum of all items on core scales divided by number of items answered)

2. Physical Health Summary Score (same as the Physical Functioning Scale Score)

3. Psychosocial Health Score (sum of items answered in Emotional, Social and School Functioning Scales)

4. Scale scores exist for the 8 supplementary cancer specific modules.

The Peds QL has been shown to have internal consistency, reliability and demonstrated to have validity through the known-groups method (Varni, Seid & Curtin, 1999).

As recommended by MREC minor modifications were made to items on this questionnaire, which were thought to be too American (e.g. 'angry' replaced 'mad').

2.4.3 Parenting Stress Index (Abidin, 1995)

This has 120 items and was standardised for use with parents of children ranging from 1 month to 12 years. In addition to a total stress score, separate scores and sub-scores for 3 sources of stress (child, parent and life event domains) are obtained. In child and parent domains items are scored on a scale of 1 to 5, while the life event domain consists of a yes or no response to specific events.

The child domain consists of the subscales: Distractibility/Hyperactivity (9 items), Adaptability (11 items), Reinforces Parent (6 items), Demandingness (9 items), Mood (5 items) and Acceptability (7 items). High scores may be associated with children who display qualities that make it difficult for parents to fulfil their parenting roles.

The parent domain consists of the subscales: Competence (13 items), Isolation (6 items), Attachment (7 items), Health (5 items), Role Restriction (7 items), Depression (9 items) and Spouse (7 items). High scores suggest the source of stress and potential dysfunction of the parent-child system may be related to dimensions of the parent's functioning.

The third scale (Life Stress) consists of 19 possible events and provides an index of the amount of stress outside the parent-child relationship the parent is currently experiencing.

This measure has been shown to have test- retest reliability (Hamilton, 1980) and construct and predictive validity (Abidin, 1995) are good.

2.5   Procedure

2.5.1   Pilot Investigation

The study was piloted on 5-10 patients under the care of the Regional Paediatric Oncology/Haematoloy. No unforeseen difficulties were encountered, and these children were included in the main analysis.

2.5.2   Participant recruitment

All participants were children (aged 5 –12 years) and parents of children diagnosed with ALL, who attended the Regional Paediatric Oncology/Haematology unit, or were under the shared-care of the Regional unit and their local hospital. Participants were approached by their Consultant and given an explanation of the study. This ensured no inappropriate families were contacted (e.g. cases where the child had suddenly deteriorated). If interested, parents (and children, if age-appropriate) were given information letters by the researcher. Due to children off-treatment not attending clinic on a regular basis the researcher phoned the parents (after speaking to the Consultant in charge) and explained the study. If they were interested in obtaining more details an information letter was sent.

Information letters described the aims of the study and what participation would involve. A stamped addressed envelope was enclosed and parents (and children) were asked to indicate whether they were interested in participating by returning the reply form (see Appendix 6 for examples of parent and child (aged 5 years and over) information letters). Follow-up letters were sent to parents who had not returned the reply form within a one-month period.

The researcher contacted parents and children who indicated interest in participating, and a visit was organised at a convenient time and place (usually their home). On meeting the participants written consent was obtained, with it being ensured that children understood and were able to consent for themselves (i.e. were 'Gillick' competent - see Appendix 7 for consent forms). On consenting, the participants agreed to their GP being informed of their participation in the study.

All participants (i.e. parents and children aged between 5 and 12 years) completed the Peds QL (Varni, Seid & Rode, 1999), while parents of children over 4 years filled in the Extended Version of the SDQ (Goodman, 1999). All parents completed the Parenting Stress Index (Abidin, 1995). The researcher was available to answer questions regarding parent self-administered instruments, and administered the Peds QL for young children (5-7 years). For the child group (8-12 years) the researcher was available to assist with the self-administered instrument after instructions had been given and clarified.

A semi-structured interview was administered to parents, asking them about the difficulties/distress they experienced with their child's diagnosis/treatment and their perspective on available services.

2.6   Statistical Analysis

The data generated included categorical, ordinal and interval data. The stages of analysis were:

1. Descriptive analysis of demographic and disease variables.

2. One-sample t-tests to analyse the association between ALL and adjustment and HRQL.

3. Independent t-tests, ANOVAS and Pearson correlations.

4. Multiple Regression analyses to determine whether demographic, disease/treatment, child and parent characteristics, and life stress predict adjustment and HRQL.

All analyses were conducted using Statistical Package for Social Sciences for Windows, Version 10 (SPSS Inc. 1999).

3. RESULTS

3.1 Response rates

A total of 51 information sheets were given to parents. Children and parents indicated interest in participating by returning reply slips. Forty-three reply slips were returned, with 41 respondents (80 % response rate) indicating interest in participating.

3.2 Demographic & disease characteristics of the sample

3.2.1 Sample characteristics

Forty-four parents (38 mothers, 6 fathers) of 23 boys and 18 girls, and 28 children (15 boys and 13 girls) participated in the research.

The children's mean age was six years six months (SD = 2.31, range = 2–12 years), with 4.9% of the children having no siblings, 48.8% having one and 46.3% having two or more. The number of siblings included stepsiblings living in the family home. Of the sample 82.9% of parents were married, 12.2% divorced and 2.4% separated. The majority of children were white (82.9%), with 9.8% Asian and 7.2% coming from other ethnic groups.

Occupational data supplied by participants was classified according to an interim version of a government National Statistics Socio-Economic Classification (NS-SEC, Rose & O'Reilly, 1998). Using the three-class version, 56.1% fell in managerial and professional class, 14.7% were intermediate class, and 26.8% were working class.

3.2.2 Disease variables

Mean age of children at diagnosis was 3.7 years (SD = 2.3, range = below 1 year–10 years). On average it was 36.3 months since diagnosis (SD = 17.6, range = 11 months–80 months).

The children studied were divided into two treatment groups, on- and off-treatment. On-treatment was defined as newly diagnosed or relapsed on-treatment, while off-treatment was termed as in remission (i.e. disease-free status accompanied by termination of treatment in past 12 months) or long-term survivor (disease free status and termination of treatment more than 12 months ago). Twenty participants were on-treatment (18 newly diagnosed and two relapsed on-treatment) and 21 were off-treatment (six in remission and 15 long-term survivors).

Patients were categorised by a Consultant Paediatric Oncologist into high risk (i.e. receiving a more intensive treatment protocol due to a poorer prognosis, or children who had relapsed) and standard risk. Twelve children were high risk (of whom 4 were relapsed patients) and 29 children were standard risk patients.

3.3   Analysis

3.3.1   Overview of the data analysis

One-sample t-tests were conducted to investigate hypothesis 1; comparisons were made with norms (i.e. healthy children) for adjustment (i.e. SDQ) and HRQL (i.e. Peds QL), and with acutely and chronically ill children for HRQL.

Kolmogorov-Smirnov tests were used to test normality of distribution (required for parametric tests) for SDQ and Peds QL scales. Distribution was normal for most SDQ scales, however normality could not be assumed for the prosocial behaviour scale ($p = 0.04$), and no t-test was conducted. Results based on the impact score need to be interpreted with caution, as the distribution was only just normal ($p = 0.11$). Distribution was normal for Peds QL paediatric-rated and parent-rated scales.

For the latter hypotheses, where t-tests and ANOVAS were conducted, Kolmogorov-Smirnov tests and Levene tests were used to test normal distribution and homogeneity of variance. Prior to conducting Pearson correlations, scatter plots were used to check that variables were linearly related. Hypotheses were one-tailed when the direction in which differences were expected was specified; non-directional hypotheses were two-tailed.

Following each regression analysis standardised residuals were examined. No evidence was found to suggest the assumptions underlying the regression models were invalid. The exception to this was the analysis of HRQL rated by children, where the

distribution of the number of siblings in the sample is problematic; this will be commented on later.

The large number of t-tests and correlations conducted increases risk of chance significant findings (i.e. Type I errors). In the current study 21 chance significant findings would have been expected with a significance level of 0.05, however 90 t-tests and correlations were found to be significant. It might have been appropriate to increase the threshold at which significance was reported to 0.01. Results significant at the 0.05 level were reported, however, due to the exploratory nature of the study. While they should be interpreted with caution, it was felt more important to identify all children with difficulties, thereby increasing the probability of Type II errors (i.e. not rejecting the null hypothesis when it should be rejected) (Perneger, 1999).

In the following section each hypothesis will be re-stated and followed by relevant results.

3.3.2 Investigation of hypothesis 1

Hypothesis 1: ALL and its treatment will be associated with poor adjustment and poor HRQL in children (one-tailed hypothesis).

In order to test this hypothesis children with ALL were compared with healthy children for both adjustment (SDQ measure) and HRQL (Peds QL), and with acutely and chronically ill children for HRQL. Initially, participant scores on the SDQ will be described; numbers of participants (and percentiles) falling within normal, borderline and abnormal categories are shown in Table 1.

3.3.2.1 Extended Version of the SDQ (Goodman, 1999).

Table 1. Numbers (percentiles) of participants in normal, borderline & abnormal categories of the SDQ.

	Numbers (Percentages)		
Parent SDQ	**Normal**	**Borderline**	**Abnormal**
Total score	22 (62.9)	6 (17.1)	7 (20.0)
Emotional symptoms	17 (48.6)	6 (17.1)	12 (34.3)
Conduct problems	24 (68.6)	4 (11.4)	7 (20.0)
Hyperactivity	27 (77.1)	3 (8.6)	5 (14.3)
Peer problems	22 (62.9)	5 (14.3)	8 (22.9)
Pro-social	32 (94.1)	1 (2.9)	1 (2.9)

SDQ = Strengths & Difficulties Questionnaire

Normative SDQ data came from the first national mental health survey of 10,438 British 5-15 year olds (Meltzer, Gatward, Goodman, & Ford, 2000). Participants' mean scores were compared with means from the survey using one-sample t-tests (Table 2).

Table 2. T-tests comparing participant scores & norms (Meltzer et al., 2000) on the SDQ.

	Participants SDQ (mean)	Norms SDQ (mean)	Significance
Total score	11.4	8.4	.004**
Emotional symptoms	3.7	1.9	.001***
Conduct problems	1.9	1.6	.171
Hyperactivity	3.6	3.5	.417
Peer problems	2.2	1.5	.013*
Impact score	3.4	0.4	.001***

SDQ = Strengths & Difficulties Questionnaire
*p<.05; **p<.01; ***p<.001

The pro-social score on the Parent SDQ (mean) was 8.3, while the norm groups (Meltzer et al., 2000) mean was 8.6.

In terms of adjustment, these results show participants with ALL suffered more total adjustment difficulties, emotional symptoms, peer problems, and had significantly higher impact scores (i.e. assessment of distress and social impairment) compared to the norm group (Meltzer et al., 2000).

3.3.2.2 Peds QL measure (Varni, Seid, & Rode, 1999).

Paediatric-report and parent-report scores on core scales (Physical, Emotional, Social and School Functioning) were compared with mean scores of chronically ill children (i.e. attendees of speciality clinics for orthopaedics, cardiology, rheumatology and diabetes), acutely ill children (i.e. inpatients/outpatients at hospital/community clinics at least three months previously) and healthy children using one-sample t-tests (Varni, Seid & Kurtin, 1999). The results are shown in Tables 3 & 4.

Table 3. T-tests comparing participant scores & scores of chronically ill, acutely ill & healthy children (Varni, Seid & Kurtin, 1999) on child-report Peds QL scores.

Participants Peds QL child scores (mean) 0 – 100: 0 = poor, 100= good HRQL		Norm Group (mean)		Significance
Total Score	74.2	Chronically ill	77.2	.133
		Acutely ill	78.7	.051
		Healthy	83.0	.002**
Physical Health	77.9	Chronically ill	77.4	.428
		Acutely ill	78.9	.372
		Healthy	84.4	.018*
Psychosocial Health	72.1	Chronically ill	77.1	.050
		Acutely ill	78.7	.017*
		Healthy	82.4	.001 **
Emotional Functioning	70.5	Chronically ill	76.4	.086
		Acutely ill	77.3	.058
		Healthy	80.9	.010*
Social Functioning	75.2	Chronically ill	81.6	.027*
		Acutely ill	82.8	.012*
		Healthy	87.4	.001**
School Functioning	70.4	Chronically ill	73.4	.185
		Acutely ill	75.7	.063
		Healthy	78.6	.011*

*p<.05; **p<.01; ***p<.001

Table 4. T-tests comparing participants scores & scores of chronically ill, acutely ill & healthy children (Varni, Seid & Kurtin, 1999) on parent-report Peds QL scores.

Participants Peds QL parent scores (mean) 0 – 100: 0 = poor, 100= good HRQL		Norm Group (mean)		Significance
Total Score	68.0	Chronically ill	74.2	.012*
		Acutely ill	80.4	.000***
		Healthy	87.6	.000***
Physical Health	72.8	Chronically ill	73.3	.447
		Acutely ill	81.8	.009**
		Healthy	89.3	.000***
Psychosocial Health	65.4	Chronically ill	74.8	.001***
		Acutely ill	79.6	.000***
		Healthy	86.6	.000***
Emotional Functioning	59.4	Chronically ill	73.1	.000***
		Acutely ill	78.8	.000***
		Healthy	82.6	.000***
Social Functioning	75.9	Chronically ill	79.8	.113
		Acutely ill	83.6	.010**
		Healthy	91.6	.000***
School Functioning	60.5	Chronically ill	71.1	.002**
		Acutely ill	74.7	.000***
		Healthy	85.5	.000***

*p<.05; **p<.01; ***p<.001

Overall, results show participants with ALL suffered significantly more problems in total HRQL than healthy children in paediatric and parent-report measures. In terms of core functioning there were less significant differences between child-related scores and chronically and acutely ill children, than the parent-report measures. Evidence supports hypothesis 1, with leukaemia and its treatment being associated with poor adjustment and poor HRQL.

3.3.3 Investigation of hypothesis 2

Hypothesis 2: A range of demographic, disease and treatment factors will be associated with, and predict, poor adjustment and poor HRQL.

3.3.3.1 Treatment status

Hypothesis 2a): Children who are on-treatment status will have poorer adjustment and HRQL than children falling into the off-treatment status (one- tailed hypothesis).

Significant differences were found between children on- and off-treatment on SDQ emotional symptoms ($t(33) = 2.0$; $p < 0.05$) and peer problems ($t(33) = -2.0$; $p < 0.05$); with children on-treatment having more emotional symptoms, and less peer problems.

Children on-treatment had significantly lower scores (i.e. poorer HRQL) than those off-treatment on child-rated ($t(26) = -1.8$; $p < 0.05$) and parent-rated ($t(39) = -3.3$; $p < 0.01$) Peds QL total score. Significant results on t-tests examining core and cancer scales can be seen below (Table 5). All children on-treatment had lower scores than children off-treatment.

Table 5. Independent–sample t-tests comparing scores for children on- & off-treatment on the Peds QL.

Scale	Mean difference	df	t value	Significance
Child-rated Physical Health summary	-14.137	26	-2.572	0.008**
Child-rated School functioning	-14.064	26	-2.173	0.020*
Parent-rated Physical Health summary	-29.240	34	-5.158	0.001***
Parent-rated School Functioning	-15.270	39	-2.335	0.013*
Parent-rated Pain	-16.964	39	-2.718	0.005**
Parent-rated Nausea	-29.750	29	-4.783	0.001***
Parent-rated Communication with physician / nurse	-17.916	38	-1.938	0.030*

$*p<.05; **p<.01; ***p<.001$

3.3.3.2 Gender

Hypothesis 2b): Boys will have poorer adjustment and HRQL, due to them undergoing longer treatment (one-tailed hypothesis).

No significant differences between boys and girls were found for adjustment.

Significant differences between boys and girls were found when independent samples t-test were conducted on Peds QL child-rated nausea score (t (26) = -1.860; p < 0.05), parent-rated total score (t (39) = -1.729; p < 0.05), parent-rated physical health (t (39) = -2.369; p < 0.01), and parent-rated nausea score (t (38) = -2.232; p < 0.05).

A Mann-Whitney was conducted on child-rated pain score due to the normality assumption not being met (z = -2.273; p < 0.01).

All of the above differences were in the expected direction, with boys having poorer scores, with the exception of parent-rated communication with physician/nurse score (t (38) = 1.737; p <0.05), when girls had poorer scores.

3.3.3.3 Age

Hypothesis 2c): The relationship between age (at diagnosis and participation) and adjustment, and HRQL will be explored (two-tailed hypothesis).

3.3.3.3.1 Age at diagnosis

Significant negative correlations were found for age at diagnosis and total SDQ score (r = -0.439; n = 35; p < 0.01), conduct problems (r = -0.438; n = 35; p < 0.01), and hyperactivity-inattention (r = -0.392; n = 35; p < 0.05), indicating current problems reduced as age increased.

Pro-social behaviour (r = 0.425; n = 34; p < 0.05) was positively correlated with age at diagnosis indicating this increased with age.

Age at diagnosis and Peds QL child-rated nausea (r = -0.385; n = 28; p < 0.05), and parent-rated worry (r = -0.447; n = 41; p < 0.05) were negatively correlated suggesting young children had less difficulties.

3.3.3.3.2 Age at participation

Participation age and total SDQ score (r = - 0.339; n = 35; p < 0.05), and SDQ impact score (r = -0.395; n = 35; p < 0.05) were negatively correlated, indicating current difficulties reduced as age increased.

Parent-rated Peds QL worry ($r = -0.434$; $n = 41$; $p < 0.01$) was negatively correlated, indicating young children had less difficulties.

In summary, findings provide some support for hypothesis 2, with demographic and treatment factors being associated with poorer adjustment and HRQL.

3.3.4   Investigation of hypothesis 3

Hypothesis 3: There will be poor adjustment and HRQL in those children who display difficult child characteristics.

Difficult child characteristics (i.e. child domain of Parenting Stress Index (PSI), Abidin, 1995) correlated positively with adjustment difficulties on all SDQ subscales (apart from pro-social behaviour, which was negatively correlated) and negatively with some of the Peds QL scales. On the whole this indicates that difficult child characteristics were associated with poorer adjustment and poorer HRQL. See Appendix 8 for significant correlations.

3.3.5   Investigation of hypothesis 4

Hypothesis 4: There will be poor adjustment and HRQL in those children whose parent's characteristics make functioning as a competent caregiver difficult.

Dysfunctional parent characteristics (i.e. parent domain of PSI, Abidin, 1995) correlated positively with adjustment difficulties on hyperactivity-inattention, and correlated negatively with pro-social behaviour and some Peds QL scales, suggesting

dysfunctional parent characteristics were associated with specific adjustment and HRQL difficulties. See Appendix 8 for significant correlations.

3.3.6 Investigation of hypothesis 5

Hypothesis 5: There will be poor adjustment and HRQL in those children who experience high levels of life stress.

High life stress (i.e. life stress domain of PSI, Abidin, 1995) correlated positively with emotional symptoms on the SDQ and negatively with some parent-rated Peds QL scales, providing some support for hypothesis 5. Significant correlations can be seen in Appendix 8.

3.3.7 Additional demographic & disease characteristics

The associations between socio-economic class, marital status of parents, ethnicity, number of siblings, disease status (i.e. high or standard risk) and adjustment (i.e. total SDQ) and HRQL (child- and parent-rated total Peds QL scores) were investigated.

Child-rated total Peds QL was found to be different for children with one sibling when compared with children with two or more siblings (t (25) = 2.034; p = 0.05). When the 4 children with more than 2 siblings were taken out of the analysis the difference disappeared, suggesting it may be those families with more than 2 children who had more problems. This finding needs to be interpreted with caution, due to a small number of children having a large influence on the results.

3.3.8 Variables predicting adjustment & HRQL

Treatment status, gender, age (at diagnosis and participation), child and parent

characteristics and life stress were entered into the following stepwise multiple

regression analyses, to determine whether they predicted adjustment (i.e. total SDQ)

and HRQL (child- and parent-rated Peds QL score).

3.3.8.1 Adjustment

Three significant predictors accounted for 72.2% of the variance in adjustment: child

characteristics (61.1%), gender (6%) and parent characteristics (5.1%) (Table 6).

Table 6. Summary statistics for the adjustment multiple regression analysis.

Dependent variable	Independent variables in order selected	B	T	R²	F
SDQ total score	Child characteristics Gender Parent characteristics	0.213 -3.742 -0.068	8.179*** -3.076** -2.342*	0.722	25.937***

SDQ = Strengths & Difficulties Questionnaire
Significant at *p<.05; **p<.01; ***p<.001

These findings further support part of hypothesis 2, hypothesis 3 and hypothesis 4,

with gender (a demographic factor), difficult child characteristics and dysfunctional

parent characteristics predicting adjustment. Disease and treatment factors did not

make a significant contribution to the variance.

A multiple regression analysis was conducted to establish whether child and parent

characteristic subscales, when entered into the analysis with treatment status, gender,

age (at diagnosis and participation) and life stress, predicted adjustment. Only the

child characteristics distractibility / hyperactivity (61.2%) and adaptability (7%)

significantly predicted variance in adjustment (Table 7). Gender and parent

characteristics did not make a significant contribution.

Table 7. Summary statistics for the multiple regression analysis examining the importance of child & parent characteristic subscales in predicting adjustment.

Dependent variable	Independent variables in order selected	B	T	R²	F
SDQ total score	Child Distractibility / Hyperactivity Child Adaptability	0.572 0.249	4.561*** 2.603**	0.682	33.236***

SDQ = Strengths & Difficulties Questionnaire
Significant at *p<.05; **p<.01; ***p<.001

3.3.8.2 HRQL rated by children

Number of siblings was included (in addition to, treatment status, gender, age (at

diagnosis and participation), child and parent characteristics and life stress) in this

analysis due to the significant result with child-rated total Peds QL. Only number of

siblings predicted variance in child-rated total Peds QL (Table 8). The distribution of

the number of siblings, however, was not normal, with most children having one or

two siblings, and only a small number having more than two. While the result may

not therefore be reliable, it suggests children with many siblings may have poorer

HRQL. A larger sample is needed to establish whether this is a reliable finding.

Table 8. Summary statistics for the child-rated HRQL (core scales) multiple regression analysis.

Dependent variable	Independent variables	B	T	R²	F
Child Peds QL total core score	Number of siblings	-10.226	-3.639***	0.346	13.244***

Significant at *p<.05; **p<.01; ***p<.001

A multiple regression analysis was conducted to examine whether child and parent characteristic subscales, when entered into the analysis with treatment status, gender, age (at diagnosis and participation) and life stress, were important in predicting child-rated HRQL (Table 9). Number of siblings was removed due to the problems discussed. Parental isolation, a parent characteristic, (24%) and treatment status (13.5%) significantly predicted HRQL.

These findings provide further partial support for part of hypothesis 2 and hypothesis 4: treatment status and dysfunctional parent characteristics predict poorer HRQL.

Table 9. Summary statistics for the multiple regression analysis examining the importance of child & parent characteristic subscales in predicting child-rated HRQL (core scales).

Dependent variable	Independent variables in order selected	B	T	R²	F
Child Peds QL total core score	Parent isolation	-1.997	-3.106**		
	Treatment status	10.792	2.275*	0.375	7.199**

Significant at *p<.05; **p<.01; ***p<.001

The cancer modules were added together to give an overall cancer/treatment measure and stepwise multiple regression analyses were conducted for

a) demographic and treatment status, total child and parent characteristic scores, and life stress.

b) demographic and treatment status, subscales for child and parent characteristics, and life stress.

When total child and parent characteristics were put into the analysis (i.e. a) above)) only total child characteristic score made a significant contribution (20.8%). When child and parent characteristics subscales were examined parental isolation explained 35.9% of the variance.

Similar findings were obtained when child core and cancer modules were totalled (dependent variable) and a) and b) were conducted.

3.3.8.3 HRQL rated by parents

Two significant predictors accounted for 46.8% of the variance in parent-rated Peds total core QL: child characteristics (22.2%) and treatment status (24.6%) (Table 10).

Table 10. Summary statistics for the parent-rated HRQL (core scales) multiple regression analysis.

Dependent variable	Independent variables in order selected	B	T	R^2	F
Parent Peds QL total core score	Child characteristics Treatment status	-0.327 16.944	-4.272*** 4.135***	0.468	16.251***

Significant at *p<.05; **p<.01; ***p<.001

A multiple regression analysis was conducted to examine whether child and parent characteristic subscales, when entered into the analysis with treatment status, gender, age (at diagnosis and participation) and life stress, predicted parent-rated HRQL (Table 11). Acceptability (a child characteristic) (22.5%) and treatment status (21.2%) significantly predicted variance.

Table 11. Summary statistics for the multiple regression analysis examining the importance of child & parent characteristic subscales in predicting parent-rated HRQL (core scales).

Dependent variable	Independent variables in order selected	B	T	R^2	F
Parent Peds QL total core score	Child acceptability Treatment status	-1.584 15.659	-3.901*** 3.729***	0.437	14.345***

Significant at *p<.05; **p<.01; ***p<.001

The cancer modules were added together to give an overall cancer/treatment measure and stepwise multiple regression analyses were conducted (in the same manner as for the analyses of the child-rated data). When total child and parent characteristics were put into the analyses, child characteristics (21.7%), age at diagnosis (29.3%), life stress (9.6%) and treatment status (7.6%) predicted significant amounts of variance.

When child and parent characteristic subscales were examined child acceptability (25.4%), life stress (15.6%), age at diagnosis (14.1%), treatment status (8.9%) and child mood (4.6%) made significant contributions to explaining the variance.

Similar findings were obtained when child core and cancer modules were totalled (dependent variable) and total child and parent characteristics, treatment status, gender, age (at diagnosis and participation) and life stress, were entered into a stepwise multiple regression.

Child acceptability (26.7%), life stress (14.4%), treatment status (16.2%), age of child at diagnosis (7.5%) and child adaptability (4.9%) predicted significant variance when core and cancer modules were totalled (dependent variable) and child and parent characteristic subscales were examined.

3.4 Additional findings

Pearson correlations were conducted to examine the relationship between child- and parent-rated Peds QL on core and disease/treatment specific modules. Emotional functioning, social functioning, treatment anxiety and worry were not correlated. Examination of data showed child-rated mean scores were higher (i.e. better HRQL) than parent-rated mean scores for emotional functioning and treatment anxiety, and lower for social functioning and worry.

The sample's mean scores on total child and parent characteristics, life stress and child and parent subscales on the PSI were compared with normative data (Abidin, 1995), using one-sample t-tests. Participants had higher scores on child adaptability ($t(40) = 2.248$; $p < 0.05$), child demandingness ($t(40) = 4.014$; $p < 0.001$), and parent health ($t(40) = 2.659$; $p < 0.01$), indicating more problems than the normative sample.

3.5   Qualitative Data

A semi-structured interview with parents produced information regarding what families and children found difficult regarding the diagnosis and treatment of leukaemia, and their perspective of available services.

Grounded theory was used to analyse the qualitative material generated, using a method outlined in Pidgeon & Henwood (1996). This involves coding material using index cards, thereby enabling sorting and re-representation of the material, with the aim of identifying significant concepts. Categories were drawn up to reflect overall themes.

A summary of the main categories (in relation to the six questions) will be given below; only categories with more than two responses will be listed. For a more detailed analysis and examples of participant responses see Appendix 9.

Question 1: What have been the hardest parts of the whole experience for you and your family of your child's diagnosis?

Diagnosis; family disruption, separation, isolation & strain; coping with seeing child ill; negative impact on family; and reliance & trust in medical staff were the main themes generated.

Question 2: What have been the hardest parts of the whole experience for your child of their diagnosis?

The main themes identified were treatment (& condition) frightening & painful; coping with side effects; isolation from friends & family; uncertainty and fear of

future/prognosis; hospital visits and interaction with medical staff; missing school; and being different from others (siblings & peers).

Question 3: What has been the most helpful support given to you and your family?

Medical & nursing staff, social worker, friends & family, other parents, support group, psychological support/counselling and charities were the main themes generated.

Question 4: What has been the most helpful support given to your child?

The main themes identified were medical/nursing staff & social workers, family, playroom/therapist, friends, and school/nursery.

Question 5: What could have been done to improve the support given to you and your family?

Themes generated were nothing; misinformed/lack of information regarding diagnosis, treatment & entitlements; emotional support for parents; would have liked all care in Oxford; support groups; and improvements with current service.

Question 6: What could have been done to improve the support given to your child?

The main themes identified were nothing, psychological input/support, key worker, and more access to play specialist.

3.6   Summary of findings

Hypothesis 1 was supported, with leukaemia and its treatment being associated with poor adjustment and poor HRQL.

Hypothesis 2 was partly supported with demographic (i.e. gender associated with HRQL, and age at diagnosis & participation being associated with adjustment & HRQL) and treatment status being associated with poorer adjustment and HRQL.

Demographic and treatment factors predicted poor adjustment and/or HRQL: gender was a significant predictor in adjustment, while number of siblings explained variance in child-rated HRQL. Age at diagnosis was important in predicting variance in parent-rated cancer-specific HRQL, and treatment status was found to predict child- and parent-rated HRQL.

Hypothesis 3 was supported with difficult child characteristics being associated with poorer adjustment and HRQL. In addition, these characteristics explained a significant amount of variance in adjustment, child-rated cancer-specific HRQL and parent-rated HRQL. The child characteristics of distractibility / hyperactivity and adaptability predicted adjustment, while child acceptability, child mood, and child adaptability significantly predicted parent-rated core and cancer-specific HRQL.

Hypothesis 4 was supported with dysfunctional parental characteristics being associated with poorer adjustment on two subscales, and poorer child- and parent-rated HRQL in some domains. These characteristics explained a significant amount

of variance in adjustment, and child-rated HRQL. Parental isolation significantly predicted child-rated core and cancer-specific HRQL.

Hypothesis 5 was partly supported with high life stress being associated with poorer emotional adjustment and poorer parent-rated HRQL in some domains. Life stress explained a significant amount of variance in parent-rated cancer-specific HRQL.

On the parent–rated measures (i.e. SDQ and parent-rated HRQL) child characteristics (and treatment status) significantly predicted variance, whereas number of siblings and parent characteristics (in addition to treatment status) were important in predicting child-rated HRQL.

4. DISCUSSION

The main aims of the present study were a) to investigate children's adjustment and HRQL in ALL and b) establish what factors were associated with, and predicted, poor adjustment and HRQL. In the discussion that follows a brief description of the sample's characteristics will be given, followed by a summary of the findings on adjustment and HRQL. The relative contribution of demographic, disease and treatment variables, child and parental characteristics, and life stress to adjustment and HRQL will then be discussed. This will be followed by methodological considerations, the clinical and theoretical implications of the current study and suggestions for future research.

4.1 Demographic & disease characteristics of the sample

The gender mix, and mean age of diagnosis, was representative of the ALL population described in the literature. The majority of parents participating were white, married females, with more than one child.

The sample's NS-SEC (Rose & O'Reilly, 1998) scores were compared to those obtained using the Labour Force Survey (i.e. national employment statistics; Institute for Social and Economic Research, 2001). From this it was apparent the sample was skewed towards class one.

4.2 The impact of ALL. Hypothesis 1: *ALL and its treatment will be associated with poor adjustment and poor HRQL in children.*

4.2.1 Adjustment

Sixty-three per cent of the sample's SDQ total scores fell within the 'normal' category, consistent with other studies, indicating that most children with cancer adjust well, and only a subset suffer serious adjustment difficulties (Kazak, 1994). The current sample had fewer difficulties compared with children attending a psychiatric clinic, but more than a community sample (Goodman & Scott, 1999).

When participants' scores were compared with results from a national mental health survey (Meltzer et al., 2000), they had more emotional problems, consistent with findings by Bennett (1994) that children with chronic medical problems were more vulnerable to internalising problems. Koocher et al. (1980) also found that paediatric cancer survivors suffered symptoms of depression, anxiety and poor self-esteem.

Children with ALL were also found to suffer more peer problems, which may reflect the disruption ALL and its treatment has on the development of peer relations. Between the ages of six and 11 years there is usually an intensification of peer relationships (Rowland, 1990). Frequent school absences as a result of hospital appointments probably interfere with development in this area. Katz and Varni (1993) suggested children newly diagnosed with cancer risked facing difficulties on their return to school and in social situations.

Finally, the elevated impact supplement score (i.e. whether difficulties interfere with home life, friendships, education, and leisure) suggests ALL and its treatment disrupts participant's daily life. Qualitative findings support this, with children reporting isolation from friends and family, missing school, and being 'different' from siblings and peers as difficult.

4.2.2 HRQL

Child- and parent-reports of physical, emotional, social and school functioning were compared with mean scores of healthy, chronically, and acutely ill children (Varni, Seid & Kurtin, 1999).

4.2.2.1 Child-rated HRQL

Children with ALL rated their HRQL as poorer than healthy children. Social functioning was lower than chronically and acutely ill children, consistent with reports of peer problems on the SDQ.

4.2.2.2 Parent-rated HRQL

Parents rated their children's HRQL as poorer than healthy and acutely ill children. When compared with chronically ill children, participants were rated as having poorer HRQL on all scales, apart from physical and social functioning.

Overall, parents described their child's HRQL as poorer than the children did themselves. A review by Sprangers & Aaronson (1992) supports this, with 'significant others' being found to underestimate patient's (with chronic disease) QOL. This may be a result of the high levels of distress typically experienced in

parenting a child with cancer, with parents focussing more on their child's distress (Canning, Hanser, Shade & Boyce, 1993). Alternatively, children may minimize reports of their symptoms of distress in the process of adapting to their illness (Canning, Canning & Boyce, 1992a).

4.2.2.3 Specific differences between child- & parent-rated HRQL

Children rated their emotional functioning as better and treatment anxiety as less problematic than parents, consistent with studies in which parents (of children with cancer) reported more psychological disorders (internalising and behavioural disorders) in their children than the children themselves. Previous research indicated agreement among observers is lower for internalising problems than externalising problems, in childhood cancer (Seid et al., 1999).

Children rated their social functioning as poorer, and worry as greater, than parents. Children with ALL are likely to spend more time with adults than peers during their illness, with frequent school absences and hospital appointments. As a result parents may be less aware of their child's problems with this measure of social functioning, which assesses primarily peer relationships. It was apparent during the administration of the Peds QL questionnaire that many parents avoided discussing relapse (an item in the worry scale) with their child, which may result in children feeling unable to discuss their worries with parents, and account for the discrepancy on this scale.

In summary, findings support Hypothesis 1, with ALL and its treatment being associated with poor adjustment and poor HRQL, when participants were compared

with healthy, acutely- and chronically-ill children. Cross-informant variance highlighted the importance of examining child- and parent-reports of HRQL.

4.3   Variables associated with & predicting adjustment and HRQL

Hypotheses 2, 3, 4 and 5 will be looked at together in this section (i.e. whether demographic, disease, child and parent characteristics and life stress, were associated with, and predicted, adjustment and HRQL).

4.3.1   Adjustment

Variables associated with adjustment

The elevation of emotional problems experienced by children on-treatment corresponds to findings reported by Varni, Seid & Rode (1999). However, children off-treatment were found to have more peer problems than children on-treatment. Previously, survivors of childhood cancer have reported difficulties initiating and maintaining personal relationships, characterised by heightened sensitivity and cautiousness (Gray, Doan, Shermer, Vatter-Fitzgerald, Berry, Jenkins & Collins, 1992). Such problems may be linked to less time spent with peers during the illness, with many hospital appointments and school absences. Varni and Katz (1997) suggest it may take up to nine months post-diagnosis for problems with school and social integration to arise. This may coincide with the children returning to some semblance of normal life.

No significant differences in adjustment were found for gender, consistent with studies reporting non-significant gender differences on parent-reported behaviour problems (Wallander & Thompson, 1995).

Younger children (at diagnosis) were found to have more total adjustment problems, and more difficulties with conduct and hyperactivity/inattention. This is consistent with the finding that multiple hospital admissions in children under five years of age are associated with increased risk of later behavioural disturbance (Quinton & Rutter, 1976). Younger children (at participation) were also found to have more current difficulties.

These findings are not consistent with Davis' (1993) conclusions, in a review of psychosocial factors in adjustment, where psychological problems increased as children got older. The current finding that younger children had more problems may reflect the behavioural measure used in this study. Young children may suffer emotional problems due to interrupted care, extended hospitalisations, and lack of understanding (Eiser & Jenney, 1996). Distress under such circumstances may be expressed in their behaviour.

Difficult child characteristics, dysfunctional parent characteristics and life stress were found to be associated with poor adjustment, supporting Hypothesis 3, 4 and 5.

Variables predicting adjustment

When predictors were examined, child characteristics predicted 61.1% of the variance in adjustment, while gender and parent characteristics explained a further 6% and 5.1%, respectively.

In a 25-year longitudinal study examining child temperament, infants were classified into 'easy-temperament', 'slow-to-warm up' and 'difficult-temperament' (Chess & Thomas, 1995). The 10% classified 'difficult' (e.g. those with difficulties establishing routines, disliking change and avoiding new situations) were at risk of developing psychological difficulties. Intuitively, difficult child characteristics might result in more distress and problems with adjustment in children with ALL, with treatment causing many disruptions. The author is unaware of any studies where child characteristics have been found to predict adjustment, in children with ALL.

Gender was not directly associated with adjustment, however it predicted a significant amount of variance in the analysis, together with child and parent characteristics. This may be due to gender having an indirect effect on adjustment.

The importance of parent characteristics is consistent with previous studies (e.g. Varni, Katz, Colegrove & Dolgin, 1996), which have found that family relationship dimensions predict adjustment in children with newly diagnosed cancer. Parent and/or family functioning have typically explained 10-15% of the variance in children's psychological outcomes (Drotar, 1992). Less variance was explained in this study. This may be due to the parent-rated measure focussing on parent characteristics and not family functioning.

When child and parent characteristic subscales were entered into a stepwise multiple regression only the child characteristics of distractibility/hyperactivity (61%) and adaptability (7%) predicted variance in adjustment. High scores on distractibility/hyperactivity subscale (e.g. 'compared to most, my child has more

difficulty concentrating and paying attention') are associated with children who display many of the behaviours associated with Attention Deficit Disorder with Hyperactivity (Abidin, 1995).

Few studies have looked at the influence of child characteristics on adjustment. However, in line with the current finding, Wallander, Hubert & Varni (1988) found mother-reported child activity levels and reactivity (in children with spina bifida and cerebral palsy) were related to behaviour problems.

Participant's adaptability subscale scores were higher than norms (Abidin, 1995). Such scores are associated with children being unable to adjust to changes in their environment. An ability to adjust to a changing environment could be seen as a necessity for children with ALL, with them having to cope with many disruptions caused by treatment. High scores on this measure are likely to cause problems with adjustment.

It is possible that the high percentage of adjustment explained was due to the dependent variable and the independent variable (i.e. child characteristics) both measuring the same construct. This will be discussed under methodological considerations.

In summary, part of hypothesis 2, hypotheses 3, 4 and 5 were supported, with demographic (i.e. age at diagnosis and participation) and treatment status, child and parent characteristics, and life stress associated with poorer adjustment. Child characteristics, gender and parent characteristics also predicted adjustment. When

analysis included child and parent characteristics subscales, distractibility/hyperactivity and adaptability predicted adjustment.

4.3.2 HRQL

The association between demographic, disease, child and parent characteristics and life stress and HRQL, and their ability to predict HRQL, will be discussed below.

4.3.2.1 Child-rated Peds QL

Variables associated with child-rated HRQL

Children on-treatment rated their physical health as poorer than children off-treatment, in line with Varni, Seid & Rode's (1999) findings. School functioning was rated poorer by children on-treatment, and may be caused by frequent school absences due to treatment.

Boys had poorer HRQL than girls on nausea and pain scales. Anticipatory nausea and vomiting are known to be conditioned responses to intensive chemotherapy (Burish, Carey, Krozely & Greco, 1987). Longer treatment in boys may increase the probability of developing such responses.

Older children (at diagnosis) experienced more difficulties with nausea, consistent with findings by Dolgin, Katz, Zeltzer & Landsverk, (1989) that older children encountered more adverse reactions to chemotherapy.

Hypotheses 3 and 4 were supported, with difficult child and dysfunctional parent characteristics being associated with poorer HRQL in certain domains. Hypothesis 5 (i.e. poorer HRQL in children where life stress was high) was not supported.

Child-rated total Peds QL was significantly different for children with one, or two or more siblings. When the four children with more than two siblings were removed from analysis the difference disappeared, suggesting it was those children who experienced more problems. This finding needs to be interpreted with caution due to a small number of individuals influencing the results.

Variables predicting child-rated HRQL

Number of siblings was the only predictor of child-rated HRQL, explaining 34.6% of the variance. Caution need to be taken when interpreting this finding, due to the abnormal distribution of number of siblings and the assumptions underlying the regression model not being fulfilled. Intuitively, however, children from large families may receive less parental physical and psychological attention due to an increase on parental demands. This fits with previous research, for example, Rutter (1979) investigated risk factors in chronic illness, and found large family size was associated with subsequent psychiatric disorders.

When child and parent characteristic subscales were entered into the multiple regression analysis, parental isolation and treatment status, explained 24% and 13.5% of the variance, respectively. Total child characteristic score (20.8%) and parental isolation (35.9%) predicted variance in cancer-specific child-rated Peds QL.

Parent isolation (e.g. 'I feel alone and without friends') is a parent characteristic, examining parent's social isolation and availability of social support with parenting. Qualitative findings support the significance of parental isolation. Parents reported family disruption, separation, isolation and strain, and lack of family support to be difficult aspects of their child's diagnosis and treatment. This is consistent with other studies. When comparisons were made with parents of healthy children, parents of chronically ill children were found to have fewer friendships (Kazak, 1991). Parents reported relatives' and friends' support was often lost after diagnosis of their child's life-threatening condition, which may be due to families 'internally regrouping' and excluding outside support systems (Mastroyannopoulou, Stallard, Lewis & Lenton, 1997). This increases risk of isolation, particularly for mothers who are less likely to be in employment.

Parents of children off-treatment have also been found to report feelings of loneliness and uncertainty (Van Dongen-Melman, Pruyn, De Groot, Koot, Hahlen & Verhulst, 1995). Parents in the current study, whose children were about to finish treatment, described feeling anxious about the reduced contact with medical staff. This may have contributed to feelings of isolation.

In summary, part of hypothesis 2, and hypotheses 3, and 4 were supported, with demographic (i.e. gender and age at diagnosis) and treatment status, and child and parent characteristics being associated with child-rated HRQL. Number of siblings was the only variable to predict child-rated HRQL. When the analysis included child and parent subscales, parental isolation and treatment status predicted 37.5% of the variance. When cancer modules (and core and cancer modules) were totalled as the

dependent variable, total child characteristics and parental isolation predicted child-rated variance.

4.3.2.2 Parent-rated Peds QL

Variables associated with parent-rated HRQL

Parents rated physical health, school functioning, pain, nausea, and communication with physician/nurse as poorer in children on-treatment. Varni, Seid & Rode (1999) also reported parents rated children on-treatment as having poorer physical functioning and disease-specific functioning.

Boys' Peds QL total score, physical health, and nausea was poorer. This may be due to them typically undergoing longer treatment, thereby supporting the hypothesis relating to gender. Boys, however, had better HRQL on communication with physician/nurse, which may reflect increased familiarity with staff as a result of undergoing longer treatment.

Worry in the current study was found to increase with age. Eiser & Jenney (1996) suggest older children experience embarrassment associated with alopecia, problems due to interrupted school attendance, and peer and family problems. As children grow older their increasing cognitive abilities results in them asking more questions about their disability or long-term survival. All of these may cause increased distress. Comments made by parents of very young children in the interview support this, with them describing their children as having adapted very well to their diagnosis. They thought this was due to their child not understanding the life-threatening nature of

their illness. Young children, however, were reported to have increased behavioural problems, and it was suggested that these may indicate emotional distress. A possible explanation of these findings is that children of all ages experience anxiety and emotional distress but that older children are more able to label these emotions.

Piagetian theory has guided the work relating to children's understanding of death. It has often been assumed children under seven years old (pre-operational stage) are unable to understand the irreversibility of death. Studies (e.g. Spence & Brent, 1984) however, have shown that there are exceptions, with exposure to death-related experiences (e.g. chronic illness) enabling younger children to understand the key features of death. While younger children's understanding should not be under-estimated, older children are more likely to have a greater understanding of their disability, which may increase the amount their parents think they worry.

Hypotheses 3, 4 and 5 were supported, with poorer parent-rated HRQL associated with difficult child characteristics, dysfunctional parent characteristics, and high levels of life stress.

Variables predicting parent-rated HRQL

Child characteristics (22.2%) and treatment status (24.6%) predicted variance in parent-rated Peds QL. When child and parent characteristic subscales were investigated, child acceptability (22.5%) and treatment status (21.2%) predicted parent-rated HRQL.

Various multiple regression analyses were conducted, with cancer modules and core/ cancer modules totalled as the dependent variable; child characteristics, age at diagnosis, life stress and treatment status were found to predict variance. Examination of child and parent characteristic subscales identified child acceptability, child mood, and child adaptability as significant predictors, in addition to those outlined above.

Problems in child acceptability is assumed to reflect a mismatch in parental expectations of attractiveness, intelligence, or how pleasant the parent had expected their child to be (e.g. 'my child does not like to be cuddled or touched very much'). Difficulties may reflect poor attachment, rejection or issues in the child-parent relationship (Abidin, 1995). Attachment may have been affected in some participants, with the diagnosis of ALL. Most parents will not have expected to have to care for a 'sick' child.

High scores on the mood subscale are associated with difficulties in affective functioning. Emotional difficulties were also identified in participants on the SDQ and HRQL measures. Finally, poor adaptability was found to also be important in child-rated HRQL, and relates to children's inability to adjust to changes in their environment. This is likely to make the disruptions, associated with treatment, difficult.

The importance of life stress supports Hypothesis 5, and is consistent with predictions made in Wallander and Varni's (1992) model, where 'general stress' (i.e. stress indirectly or not at all related to their condition) was thought to increase the risk of adjustment problems. In the interview parents frequently discussed the financial

strain of their child's diagnosis, with one/or both parents temporarily or permanently giving up work.

In summary, part of hypothesis 2, and hypotheses 3, 4 and 5 were supported, with demographic (i.e. gender, and age at diagnosis) and treatment status, child and parent characteristics and life stress being associated with poor parent-rated HRQL. Child characteristics and treatment status were significant predictors of parent-rated HRQL. Child acceptability and treatment status were significant predictors when child and parent subscales were analysed.

When cancer and cancer/core modules were totalled as the dependent variable in the multiple regression analyses, child characteristics, age at diagnosis, life stress, and treatment status were significant predictors. Child acceptability, child mood, and child adaptability were important in explaining the variance in the cancer and cancer/core modules (in addition to, age at diagnosis, life stress, and treatment status), when parent and child characteristic subscales were examined.

Overall, ALL and its treatment has been found to be associated with poor adjustment and poor HRQL. Demographic and treatment factors were associated with adjustment and HRQL, and predicted adjustment and HRQL. Difficult child characteristics, dysfunctional parental characteristics and life stress were also associated with adjustment and/or HRQL, and predicted adjustment and/or HRQL.

On the parent-rated measures, child characteristics (and treatment status) significantly predicted variance, whereas a family characteristic (i.e. number of siblings) and a

parent characteristic (and treatment status) were important in predicting child-rated core HRQL. Child characteristics were, however, important in predicting the cancer-specific modules of the child-rated Peds QL.

4.4 Methodological considerations

4.4.1 Sample

A very high response rate was achieved. This may reflect the recruitment method and high participant satisfaction with services (evident in the positive responses regarding current services in the interview). Despite this the sample size was relatively small and would have benefited from more participants.

The majority of parent participants were mothers, Caucasian and from high socio-economic classes, limiting the generalisability of the findings. Participants did not include children diagnosed or relapsed within the previous six months. This criterion was included due to increased levels of distress for families during this time. However, their exclusion may result in an underestimation of the need for services.

4.4.2 Measures

The SDQ was quick to administer and previous studies have found it to have good reliability and validity. However, a disadvantage of the measure, which was not developed specifically for children with physical illnesses, is that items ask about physical symptoms (e.g. 'often complains of headaches, stomach-aches or sickness'). Due to overlap with treatment side-effects, the number of children with psychological problems may be overestimated (Perrin et al., 1991).

Adjustment difficulties were only assessed in children aged four years and over. This was due to the informant-rated version of the SDQ only being suitable for parents of children aged four years and over. In hindsight, Rickman's (1987) Behaviour Checklist could have been used in the younger age group.

In this study, 61% of adjustment was found to be attributable to child characteristics. It is possible that the same constructs were being measured by dependent (i.e. SDQ) and independent variable (i.e. child domain, PSI, Abidin, 1995). While this may account for the high amount of variance explained it does not explain why only distractibility/hyperactivity and adaptability significantly explained adjustment. In addition, child characteristics were also found to be important in explaining variance in HRQL (i.e. total Peds QL, which consists of physical, emotional, social and school functioning), where the likelihood of overlap was less.

4.4.3 Design & statistical analyses

This study used a cross-sectional design, thereby limiting the conclusions that can be drawn about direction of causality. While a longitudinal design would have been more informative this was not possible due to the time constraints of the study. Children on- and off-treatment were also combined in this study, in order to obtain adequate numbers. The resulting heterogeneity may reduce the precision of findings, although significant predictors were found for adjustment and HRQL.

Results significant at the 0.05 level were reported in this study, thereby increasing the possibility of Type I errors. It was felt, however, important to identify all children

with difficulties and examine possible reasons for this, while acknowledging the increased risk of not rejecting the null hypothesis when appropriate.

Finally, no controls were used so differences (e.g. on between-group comparisons) and patterns in the results cannot be attributed to ALL per se, and may instead be attributable to having a chronic disease.

4.5   Implications of present study

4.5.1   Clinical implications

It was hoped that through identifying problem-specific situations for individual children, patient/treatment matches could be better implemented. One area identified as problematic in this study was peer relations and social functioning. It may be appropriate to consider social skills training and school reintegration cognitive-behaviour therapy (Varni, Blount & Quiggins, 1998) for such children.

If future research confirmed the importance of child (e.g. distractibility/hyperactivity) and parent characteristics (e.g. parental isolation) in predicting children's adjustment and HRQL, these could be identified in assessment and appropriate interventions developed and used to decrease difficulties encountered in these areas.

Children's problems (i.e. peer problems) were not found to resolve when treatment finished. This has implications in terms of follow-up, and is especially important due to increased survival rates. Eiser (1998) concluded from a review into long-term

consequences of childhood cancer that it was 'imperative that all survivors are offered the opportunity for systematic follow-up and advice' (p.30)

Cross-informant variance in this study highlights the importance of obtaining children's perceptions of HRQL, especially in relation to internalising problems. If one assumes children were providing reliable and valid estimates of their HRQL, educating parents about their child's HRQL may relieve some of their own distress.

It was apparent during the interview that many parents wanted to talk about their experience of having a child with cancer. This may relate to their perceived isolation (reported in this study). Kazak, Stuber, Barakat, Meeshe, Guthrie & Meadows (1998) described parents being eager to talk about their experience of cancer, reporting that few people understood their ongoing distress. In the current study a few parents (mainly those with children off-treatment), however, said that talking about their child's illness was difficult, as they recalled how difficult it had been. This is in line with reports that disclosure can cause distress (Kelly et al., 1997, cited in Eiser 2000). Informed consent and de-briefing need, therefore, to be included in research studies of this type, in order to minimise distress.

In the current study some parents described feeling they should have been provided with more support. In particular, one parent commented that there was 'no emotional support for parents'. A multi-modal treatment approach (Frank, Blount & Brown, 1997) is therefore recommended, with the focus on both child and family. Support groups for parents could be set-up (and existing ones improved), with the aim of reducing isolation in parents of children diagnosed with ALL.

Qualitative findings highlighted the importance of assessing the individual needs of each family. For example, some parents described previously wanting more information (e.g. 'wanted to be more informed of treatment, downs as well as ups, explanation of why medication was needed and what if it doesn't work').

A summary of the results, including qualitative findings, will be sent to all participants and the findings will be fed back to the Regional Paediatric Oncology / Haematology Unit. The importance of disseminating studies findings was highlighted by Eiser et al. (2000).

4.5.2 Theoretical implications

Research into child adjustment needs to focus on coping and adjustment, instead of deficits and maladjustment. The current findings are consistent with previous research, with most children adjusting well to their diagnosis.

Future research needs to clearly define adjustment and HRQL and be guided by explicit theoretical frameworks. There is a long way to go before such frameworks adequately explain and predict which children will experience problems.

Modifications may need to be made to Varni & Wallander's (1992) model of adjustment. For example, the hypothesised relationship between severity of illness and adjustment was not supported in the current study, in line with previous findings (Lavigne & Faier-Routman, 1993). In the current study difficult child characteristics were found to be a 'risk factor' in child adjustment, while in previous studies, certain family environments were associated with childhood behavioural problems and delays

in social competence (Bradford, 1997). In addition, future research needs to clarify the relationship between adjustment and HRQL.

4.6   Future research

Larger samples, through multi-centre collaboration, and longitudinal designs need to be used, enabling an understanding of how the course of ALL and treatment interacts with individual and family development. The current study supported the recommendation that multiple methods and informants are required in the assessment of adjustment (Thompson, Merritt, Keith, Murphy & Johndrow, 1993).

Future studies need to identify which factors (e.g. coping strategies) protect children and families from developing problems in relation to adjustment and HRQL. While the focus should not be one of maladjustment there remains a need to identify and treat the significant minority of children and families requiring support and to evaluate the effectiveness of such interventions.

The qualitative data in this study was analysed using grounded theory and categories were drawn up to reflect themes. Support from medical/nursing staff was a category generated from parents' responses to the question asking what support they had received. Bradford (1997) also highlighted the importance of 'doctor-patient' communication and the health care environment. Future research could more formally test how these affect adjustment in children and their families and incorporate them into the theoretical framework if they were found to be influential.

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The current study found the Peds QL easy to use, fast to administer and score, and its interpretation logical, all of which are necessary in a HRQL measure (Mulhern et al., 1989). Pollock (1999) highlights the need for HRQL measures to make sense to physicians, patients and families. However, to aid interpretation of the Peds QL results normative data needs to be collected.

The effect of siblings and parental adjustment should also be investigated further. The importance of such research is highlighted in a study by Kaplan et al. (cited in Parsons et al., 1999) which found parent's assessment of their own level of functioning correlated strongly with parent's assessment of their child's functioning.

4.7   Conclusion

In conclusion, the current study aimed to investigate children's adjustment and HRQL in ALL, and establish what factors were associated with, and predicted poor adjustment and HRQL. In line with other studies many children with ALL had adjusted well to their diagnosis, however 34.3% and 22.9% suffered from emotional and peer problems, respectively. Their HRQL was also poorer than healthy children and worse than acutely- and chronically-ill children on some scales.

Demographic, treatment status, child and parent characteristics and life stress were all found to be associated with adjustment and/or HRQL. Child characteristics (in particular, distractability/hyperactivity and adaptability), gender and parent characteristics were found to predict adjustment. Number of siblings, parental isolation and treatment status predicted child-rated core HRQL. Child characteristics

(i.e. acceptability, mood and adaptability), treatment status, age at diagnosis and life stress all predicted parent-rated HRQL.

The importance of cross informant variance was apparent with child characteristics (and treatment status) significantly predicting variance on parent-rated measures, while family (i.e. number of siblings) and parent characteristics (in addition to treatment status) predicted child-rated core HRQL.

The methodological weaknesses limit the extent to which conclusions can be drawn and need to be addressed in future research. Nevertheless, results suggest difficult child characteristics, dysfunctional parent characteristics and treatment status, in particular, increase the risk of poor adjustment and HRQL.

Future multi-centre, longitudinal studies need to use clear definitions of adjustment and HRQL. Guided by explicit theoretical frameworks, it is hoped that they will be able to adequately explain and predict which children will experience problems, allowing services to be increasingly needs driven.

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APPENDICES

Appendix 1:   Confirmation of Ethical Approval

Appendix 2:   The Extended Version of the Strengths & Difficulties Questionnaire

Appendix 3:   The Peds QL Questionnaire

Appendix 4:   The Parenting Stress Index

Appendix 5:   Semi-structured Interview

Appendix 6:   Information letters

Appendix 7:   Consent Forms

Appendix 8:   Significant Correlations

Appendix 9:   Qualitative data

TEXT BOUND CLOSE TO

THE SPINE IN THE

ORIGINAL THESIS

APPENDIX 1

OUR REF:SAPS00/5/40/resubmission/caoct00.doc
18 October 2000

MREC Response Form

MULTI CENTRE RESEARCH ETHICS COMMITTEE RESPONSE FORM

MREC reference: 00/5/40. An investigation into psychological adjustment in children who have been diagnosed with acute lymphoblastic leukaemia. Resubmission after rejection, for 13/9/00. Approved subject to amendment 13/9/00 & applicants amendments approved by Chairman's Action.

DETAILS OF APPLICANT

1.   Name and Address of Principal Researcher:

Mr R Scott
Consultant Neuropsychologist
Russell Cairns Unit
Radcliffe Infirmary
Oxford

Qualifications

Psychology undergraduate degree, PhD, and Post graduate clinical psychology qualification

2.   Title of Project:

An investigation into psychological adjustment in children who have been diagnosed with acute lymphoblastic leukaemia.

3.   Name and Address of sponsor

Being funded by the Oxford Doctoral Course in Clinical psychology

DETAILS OF MREC

4.   Name and address of MREC:

Anglia & Oxford MREC, Cambridgeshire Health Authority, St John's, Thorpe Rd, Peterborough, PE3 6JG

5.   MREC reference number: 5. Study 00/5/40

Manor House
Headley Way, Headington
Oxford OX3 9DZ

Tel: 01865 222547
Fax: 01865 222699

RJ/lmp/OM00.68

16 November 2000

Dr Richard Scott
Consultant Neuropsychologist
Russell Cairns Unit
RADCLIFFE INFIRMARY

Dear Dr Scott

Re: OM00.68 – An investigation into psychological adjustment in children who have been diagnosed with acute lymphoblastic leukaemia MREC/00/5/40

Thank you for submitting your MREC approved research application to OPREC for local approval. It was reviewed by the Executive Sub-Committee at their meeting on the 13 November 2000.

In accordance with the interim guidance set down in September 1998 the suitability of the local researcher, the site, the Patient Information Sheet & Consent Form were considered, and I am now please to confirm local approval for your study.

Please note:

- Ethical approval is valid for three years from the date of this letter.

- No significant changes to the research protocol should be made without appropriate research ethics committee/chairman's approval. Any deviations from or changes to the protocol which increase the risk to subjects, or affect the conduct of the research, or are made to eliminate hazards to the research subjects, should be made known to OPREC.

- OPREC should be made aware of any serious adverse events.

- Whilst the study has received approval on ethical grounds, it is necessary for you to obtain management approval from the relevant Clinical Directors and/or Chief Executive of the Trusts (or Health Boards/DHAs) in which the work will be done.

I should be very grateful if you could send me a copy of any publication which may arise from this study.

NB: Any research which will be conducted on NHS patients or staff, and which has been approved by a research ethics committee must carry the appropriate indemnity. May I remind you that OPREC final approval is contingent on the appropriate indemnity being in place.

Yours sincerely,

Professor Robin Jacoby
Chairman
Oxfordshire Psychiatric Research Ethics Committee

Oxfordshire Psychiatric Research Ethics (OPREC)			
OPREC No: OM00.68			
Title of Project: An investigation into psychological adjustment in children who have been diagnosed with acute lymphoblastic leukaemia MREC/00/5/40			
Members in Attendance at the Executive Sub-Committee meeting on: 13 November 2000			
Professor Robin Jacoby	4	Professor Paul Harrison	4
Dr Jenny McCleery	4		
The following documents have been reviewed and approved by OPREC			
		Date/Version	Approved
Annexe D		13 November 2000	4
Protocol:		13 November 2000	4
MREC Correspondence		13 November 2000	4
MREC Application Form		13 November 2000	4
MREC Approval Letter		13 November 2000	4
Information Sheet		13 November 2000	4
Consent Form		13 November 2000	4
GP Letter		13 November 2000	4
Questionnaire		13 November 2000	4
Interview Schedules		13 November 2000	4

Terms of Reference, Standard Operating Procedures and a list of members of the Ethics Committee are available from the Research & Development office on request.

INDEMNITY
The purpose of an indemnity arrangement for a researcher is to provide legal protection in the event of a researcher-led unforeseen adverse circumstance, however minimal the risk, arising during the course of a research project. The indemnity applies to the Senior Investigator in the project and automatically covers any other generally more junior colleagues associated with the project. There are various types of indemnity dependent on the circumstances of the researcher and the nature of the research project. Staff with contracts or honorary contracts of employment in NHS Trusts should ensure that they are properly protected by the appropriate indemnity approved by the Trust Chief Executive or Medical Director.

East Berkshire Research Ethics Committee

John Lister Postgraduate Medical Centre, Wexham Park Hospital, Slough,
Berkshire SL2 4HL

9th February 2001

Caroline Paul
Trainee Clinical Psychologist
Oxford Doctoral Course in Clinical Psychology
Isis Education Centre
Warneford Hospital
Oxford
OX3 7JX

Dear Ms Paul,

**Re: East Berkshire Research Ethics Committee Application No: 2268
Psychological adjustment in children who have been diagnosed with acute
lymphoblastic leukaemia (ALL). MREC/00/5/40**

The East Berkshire research ethics Committee received and approved the above
study at the meeting held on the 8th February 2001.

The following Committee members were present at the meeting:

> Dr. A. Macaulay (Chairman)
> Mrs. M. Barwick
> Mr A. Desai
> Mr. S. Dimitry
> Mr. J. McAllister
> Dr. I. Mower
> Mr. A. Prosser
> Dr. I. Walker
> Dr. G. Odds OBE

For record keeping purposes, the following documentation was received:
- Letter to Dr Macaulay from Ms Paul
- Annexe D
- MREC response form
- MREC application
- Study protocol, version 2, August 2000
- Information sheet for Parents of children over 5 years of age, version 3
  September 2000
- Information sheet for children over 5 years of age, version 3 September 2000

Chairman: Dr. A Macaulay
Telephone: (01753) 634670

Administrators: Ms Vicki Gedge
Mrs. Margaret Duffill
Telephone: (01753) 634364
Fax: (01753) 634189
Email: vicki.gedge@hwph-tr.nhs.uk

APPENDIX 2

Strengths and Difficulties Questionnaire

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all times as best as you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of your child's behaviour over the last six months or this school year.

	Not true	Somewhat True	Certainly True	
Considerate of other people's feelings.........				PS
Restless, overactive, cannot stay still for long...				H
Often complains of headaches, stomach-aches or sickness.......................................				E
Shares readily with other children (treats, toys, pencils, etc.).......................................				PS
Often has temper tantrums or hot tempers......				CD
Rather solitary, tends to play alone.............				PP
Generally obedient, usually does what adults request.......................................				(CD)
Many worries, often seems worried.............				E
Helpful if someone is hurt, upset or feeling ill				PS
Constantly fidgeting or squirming..............				H
Has at least one good friend.......................				(PP)
Often fights with other children or bullies them				CD
Often unhappy, down-hearted or tearful........				E
Generally liked by other children.................				(PP)
Easily distracted, concentration wonders......				H
Nervous or clingy in new situations, easily loses confidence....................................				E
Kind to younger children........................				PS
Often lies or cheats................................				CD
Picked on or bullied by other children.........				PP
Often volunteers to help others (parents, teachers, other children)...........................				PS
Thinks things out before acting..................				(H)
Steals from home, school or elsewhere.........				CD
Gets on better with adults than with other children.				PP
Many fears, easily scared.......................				E
Sees tasks through to the end, good attention span				(H)

Signature...................................... Date..............................
Parent/Teacher/Other (Please specify)

The Parent-rated Impact Supplement

Overall, do you think that your child has difficulties in one or more of the following areas: emotions, concentration, behaviour or being able to get on with other people?

No	Yes – minor difficulties	Yes – definite difficulties	Yes – severe difficulties
☐	☐	☐	☐

If you have answered 'Yes', please answer the following questions about these difficulties:

- How long have these difficulties been present?

Less than a mth.	1 – 5 mths.	6 –12 mths.	Over 1 year
☐	☐	☐	☐

- Do the difficulties upset or distress your child?

Not at all	Only a little	Quite a lot	A great deal
☐	☐	☐	☐

- Do the difficulties interfere with you child's everyday life in the following areas?

	Not at all	Only a little	Quite a lot	A great deal
Home life	☐	☐	☐	☐
Friendships	☐	☐	☐	☐
Classroom learning	☐	☐	☐	☐
Leisure Activities	☐	☐	☐	☐

- Do the difficulties put a burden on you or the family as a whole?

Not at all	Only a little	Quite a lot	A great deal
☐	☐	☐	☐

APPENDIX 3

Peds QL – YOUNG CHILD REPORT (5-7)

Instruction for the interviewer:

I am going to ask you some questions about things that might be a problem for some children. I want to know how much of a problem any of these might be for you.

Show the child the template and point to the response as you read.

If is __not at all__ a problem for you, point to the smiling face

If is __sometimes__ a problem for you, point to the middle face

If it is a problem for you __a lot__, point to the frowning face

I will read each question. Point to the pictures to show me how much of a problem it is for you. Let's try a practice one first.

	Not at all	Sometimes	A lot
Is it hard for you to snap your fingers			

Ask the child to demonstrate snapping his or her fingers to determine whether or not the question was answered correctly. Repeat the question if the child demonstrates a response that is different from his or her action.

Think about how you have been doing for the last few weeks. Please listen carefully to each sentence and tell me how much of a problem this if for you.

After reading the item, gesture to the template. If the child hesitates or does not seem to understand how to answer, read the response options while pointing at the faces.

PHYSICAL FUNCTIONING (problems with....)	Not at all	Some-times	A lot
1. Is it hard for you to walk	0	2	4
2. Is it hard for you to run	0	2	4
3. Is it hard for you to play sports or exercise	0	2	4
4. Is it hard for you to pick up big things	0	2	4
5. Is it hard for you to take a bath or shower	0	2	4
6. Is it hard for you to do chores (like pick up your toys)	0	2	4
7. Do you ache (Where?_____)	0	2	4
8. Do you ever feel too tired to play	0	2	4

EMOTIONAL FUNCTIONING (problems with....)	Not at all	Some-times	A lot
1. Do you feel scared	0	2	4
2. Do you feel sad	0	2	4
3. Do you feel angry	0	2	4
4. Do you have trouble sleeping	0	2	4
5. Do you worry about what will happen to you	0	2	4

SOCIAL FUNCTIONING (problems with....)	Not at all	Some-times	A lot
1. Is it hard for you to get along with other children	0	2	4
2. Do other children say they do not want to play with you	0	2	4
3. Do other children tease you	0	2	4
4. Can other children do things that you cannot do	0	2	4
5. Is it hard for you to keep up when you play with other children	0	2	4

SCHOOL FUNCTIONING (problems with....)	Not at all	Some-times	A lot
1. Is it hard for you to pay attention in school	0	2	4
2. Do you forget things	0	2	4
3. Is it hard to keep up with schoolwork	0	2	4
4. Do you miss school because you do not feel well	0	2	4
5. Do you miss school because you have to go to the doctors or hospital	0	2	4

Think about how you have been doing for the last few weeks. Please listen carefully to each sentence and tell me how much of a problem this if for you.

After reading the item, gesture to the template. If the child hesitates or does not seem to understand how to answer, read the response options while pointing at the faces.

PAIN AND HURT (problems with....)	Not at all	Some-times	A lot
1. Do you ache or hurt in you joints and/or muscles	0	2	4
2. Do you hurt a lot	0	2	4

SICKNESS (problems with....)	Not at all	Some-times	A lot
1. Do you get sick when you have medical treatments	0	2	4
2. Does food taste bad to you	0	2	4
3. Do you get sick when you think about medical treatments	0	2	4
4. Do you not feel hungry	0	2	4
5. Do some foods and smells make your stomach upset	0	2	4

PROCEDURAL ANXIETY (problems with....)	Not at all	Some-times	A lot
1. Do needles (i.e. injections, blood tests, IVs) hurt you	0	2	4
2. Do you get scared when you have to have blood tests	0	2	4
3. Do you get scared about needles (i.e. injections, blood tests, IVs)	0	2	4

TREATMENT ANXIETY (problems with....)	Not at all	Some-times	A lot
1. Do you get scared when you are waiting to see the doctor	0	2	4
2. Do you get scared when you have to go to the doctor	0	2	4
3. Do you get scared when you have to go to the hospital	0	2	4

WORRY (problems with....)	Not at all	Some-times	A lot
1. Do you worry about side effects from medical treatments	0	2	4
2. Do you worry about whether or not your medical treatments are working	0	2	4
3. Do you worry that your leukaemia will come back	0	2	4

Think about how you have been doing for the last few weeks. Please listen carefully to each sentence and tell me how much of a problem this if for you.

COGNITIVE PROBLEMS (problems with....)	Not at all	Some-times	A lot
1. Is it hard for you to figure out what to do when something bothers you	0	2	4
2. Do you have trouble solving math problems	0	2	4
3. Do you have trouble writing at school	0	2	4
4. Is it hard for you to pay attention to things	0	2	4
5. Is it hard for you to remember what is read to you	0	2	4

PERCEIVED PHYSICAL APPEARANCE (problems with....)	Not at all	Some-times	A lot
1. Do you feel you are not good looking	0	2	4
2. Do you not like other people to see your scars	0	2	4
3. Are you embarrassed when others see your body	0	2	4

COMMUNICATION (problems with....)	Not at all	Some-times	A lot
1. Is it hard for you to tell the doctors and nurses how you feel	0	2	4
2. Is it hard for you to ask the doctors and nurses questions	0	2	4
3. Is it hard for you to explain your illness to other people	0	2	4

How much of a problem is this for you?

Not at all

Sometimes

A lot.

Parenting Stress Index (PSI, Abidin, 1995)

Only examples of the sub-scales for child, parent and life event domains will be given for the PSI due to the questionnaire being very long (120 items). Parents were asked whether they strongly agreed, agreed, were not sure, disagreed, or strongly disagreed with the statements below.

<u>Child Domain</u>

Distractibility / Hyperactivity Subscale
Examples:

3. My child appears disorganized and is easily distracted.

4. Compared to most, my child has more difficulty concentrating and paying attention.

Reinforces Parent

Examples:

10. My child rarely does things for me that make me feel good.

11. Most times I feel that my child likes me and wants to be close to me.

Mood

Examples:

17. My child seems to cry and fuss more often than most children.

20. I feel that my child is very moody and easily upset.

Acceptability

Examples:

25. My child does a few things which bother me a great deal.

27. My child does not like to be cuddled or touched very much.

Adaptability

Examples:

38. It takes a long time and it is very hard for my child to get used to new things.

39. My child doesn't seem comfortable when meeting strangers.

Demandingness

Examples:

44. There are some things my child does that really bother me a lot.

46. As my child has grown older and become more independent, I find myself more worried that my child will get hurt or into trouble.

Parent Domain

Competence

Examples:

52. I have had many more problems raising children *than I expected.*

54. I feel that I am successful most of the time when I try to get my child to do or not to do something.

Attachment

Examples:

63. I expected to have closer and warmer feelings for my child than I do and this bothers me.

66. My child knows I am his or her parent and wants me more than other people.

Role Restriction

Examples:

69. I find myself giving up more of my life to meet my children's needs than I ever expected.

72. Since having this child, I have been unable to do new and different things.

Depression

Examples:

75. When I think about the kind of parent I am, I often feel guilty or bad about myself.

82. I wind up feeling guilty when I get angry at my child and this bothers me.

Spouse

Examples:

84. Since having my child, my spouse (male/female friend) has not given me as much help and support as I expected.

85. Having a child has caused more problems than I expected in my relationship with my spouse (male/female friend).

Isolation

Examples:

91. I feel alone and without friends.

95. When I run into a problem taking care of my children, I have a lot of people to whom I can talk to get help or advice.

Health

Examples:

97. During the past six months, I have been sicker than usual or have had more aches and pains than I normally do.

98. Physically, I feel good most of the time.

Life Stress Domain

During the last 12 months, have any of the following events occurred in your immediate family?
(Yes / No)

102. Divorce

112. Income decreased substantially

Name   **Date**

Below are some questions relating to your child's illness. It is hoped that through examining what you, your family and your child
- found difficult
- found helpful
- would have found helpful

that it will enable us in the future to support children and families suffering similar problems.

1. What have been the hardest parts of the whole experience for **you and your family** of your child's diagnosis?

-
-
-

2. What have been the hardest parts of the whole experience for your **child** of their diagnosis?

-
-
-

3. What has been the most helpful support given to **you and your family**?

-
-
-

4. What has been the most helpful support given to your **child**?

-
-
-

5. What could have been done to improve the support given to **you and your family**?

-
-
-

6. What could have been done to improve the support given to your **child**?

-
-
-

Oxford Radcliffe HOSPITAL

PAEDIATRIC HAEMATOLOGY/ONCOLOGY UNIT

Dr C.D. Mitchell, PhD, FRCP
Dr K.A. Wheeler, MRCP, FRCPCH
Dr G.W. Hall, PhD, MRCP, MRCPath

Headley Way
Headington
OXFORD
OX3 9DU

Telephone: *(01865) 741166*
Direct Line: *(01865) 221057*
Fax: *(01865) 221083*

A study into the adjustment of children with leukaemia

Information Sheet for Patients

You are being asked to help in some work we are doing, which we will tell you about in this letter. Please read this letter, talk to your parents and friends, and speak to us if you do not understand anything.

We are asking you because we know that you have leukaemia and we would like to know how this has affected you. We want to know more about how children get along with their illness as this would tell us how to help other children and families who are ill too.

If you want to help, a lady called Caroline Paul will ask you some questions about things like, what you find easy and difficult at home, at school, or in the hospital. This would take about 10 to 15 minutes.

What you tell us will only be used to help us in our work. That means we won't tell anyone else what you have said.

If you want to help us please tick 'Yes' on the Reply Sheet and give it to a grown-up at home, who will send it to Caroline Paul. She will then come and see you and ask you some questions.

If you do help we would like to tell your doctor that you are going to and what we will be doing.

If you don't want to you don't have to and that's okay. Could you then please tick 'No' on the Reply sheet and give it to a grown-up at home.

Thank you for reading this letter.

The people that are doing this work are:

Caroline Paul, Trainee Clinical Psychologist

Dr Richard Scott, Consultant Clinical Psychologist

Dr Kate Wheeler, Consultant Paediatric Oncologist

Dr Chris Mitchell, Consultant Paediatric Oncologist

Dr Georgina Hall, Consultant Paediatric Haematologist

Record Number ☐

Child's name _____

REPLY SHEET

Please tick one of the boxes:

☐ **Yes**

☐ **No**

Please give this to a grown-up at home.

PAEDIATRIC HAEMATOLOGY/ONCOLOGY UNIT

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A study into the adjustment of children with leukaemia

Information Sheet for Parents

You are being invited to take part in a research study. Before you decide if you would like to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with friends, relatives and your G.P. if you wish. Ask us if there is anything, which is not clear to you, or if you would like more information. Take time to decide whether or not you wish to take part.

<u>Introduction</u>

This study is investigating how children adjust to the diagnosis and treatment of leukaemia. We are of course aware that the diagnosis and subsequent treatment of leukaemia has a major impact on children and families in many ways, not all directly related to the diagnosis. We hope to learn more about what particular things help some children and their families to cope better with the diagnosis and treatment of leukaemia. This would then enable us in the future to support children and families who are suffering similar problems.

Why have I been chosen?

We understand that your child has been diagnosed with having leukaemia, and would like your opinion on how your child is adjusting to the diagnosis and treatment of leukaemia. We have also attached a letter for you child, as we would like to find out how they feel they are managing and in what areas they are (and are not) having problems.

What would I have to do if I take part?

If you agree to take part, it would involve a single meeting with Caroline Paul. During this meeting you would be asked to complete three questionnaires. These ask, for example, about your child's functioning in physical and social domains. The completion of these questionnaires will take between 30 and 40 minutes.

In addition, we ask your child to complete a questionnaire, which would take you between 10 to 15 minutes, and would allow us to see what things they find difficult or easy in areas such as physical and social domains.

What about confidentiality?

The information provided on the questionnaires will only be used for research purposes, with the results being stored (on a computer) and reported anonymously. The information would be completely confidential to the researchers and your identity would be protected.

Do I have to take part?

No. Taking part is voluntary. If you would prefer not to take part, you do not have to give a reason. If you do take part but later change your mind, you can withdraw consent at any time.

If you are agreeable, we would wish to inform your G.P. that you are taking part in the study. This would be a short letter explaining what the study is about.

What will happen to the results of the research study?

The results of the study will be written up as a doctoral dissertation and may be published in the future. You will not be identified in any report or publication. If you would like a summary of the results in the future you would be able to let us know.

Who has reviewed the study?

The study has been reviewed by the Multi-Centre Research Ethics Committee for Anglia and Oxfordshire.

How do I contact you?

If you are interested in participating in this study, please tick 'Yes' on the Reply Sheet and return it in the stamped, addressed envelope provided. Caroline Paul would then contact you to discuss the study further and, if you are still interested, arrange to meet with you at a convenient time and place (probably your home) to complete the questionnaires.

If you would prefer not to take part in the study then we would be grateful if you would tick 'No' on the Reply sheet and return it in the enclosed postage paid envelope. This will save you from receiving any further correspondence.

If on receipt of this letter you would like the opportunity to discuss any aspect of this study, or any concerns you may have, then please feel free to contact Caroline Paul at the Isis Education Centre, Warneford Hospital, Oxford (**Tel: 01865 226431**), or Dr Kate Wheeler at the John Radcliffe Hospital, Oxford (**Tel: 01865 221057**).

We do appreciate that you may not wish to take part. Your participation, however, would enhance our understanding of these important issues. Thank you.

Caroline Paul, Trainee Clinical Psychologist

Dr Richard Scott, Consultant Clinical Psychologist

Dr Kate Wheeler, Consultant Paediatric Oncologist

Dr Chris Mitchell, Consultant Paediatric Oncologist

Dr Georgina Hall, Consultant Paediatric Haematologist

Record Number ☐

Parent's name _____

REPLY SHEET

Please tick one of the following boxes:

☐   **Yes, I am interested in participating in the study**

I can be contacted at the following address or phone number:

Address   _____
               _____

Phone number   _____

(Convenient times _____)

☐   **No, I am not interested in participating in the study**

Please return in the enclosed envelope to:

Caroline Paul
Trainee Clinical Psychologist
Oxford Doctoral *Course in* Clinical Psychologist
Isis Education Centre
Warneford Hospital
Oxford
OX3 7JX

Oxford
Radcliffe
H O S P I T A L

THE JOHN RADCLIFFE

Centre Number:
Study Number:
Patient Identification Number for this trial:

PATIENT CONSENT FORM

Title of the study: A study into the adjustment of children with leukaemia

Name of Researchers: Caroline Paul, Trainee Clinical Psychologist
Dr Richard Scott, Consultant Clinical Psychologist
Dr Kate Wheeler, Consultant Paediatric Oncologist
Dr Chris Mitchell, Consultant Paediatric Oncologist
Dr Georgina Hall, Consultant Paediatric Haematologist

1. I have read the Information Sheet, and asked questions about anything I don't understand.

2. I know I don't have to take part and that it won't affect my treatment if I don't.

3. I don't mind the people named above looking at my medical notes when needed for the study.

4. I agree for my doctor to be told that I am taking part in the study.

5. I agree to take part in the above study.

Name of Patient	Date	Signature

Name of Parent	Date	Signature

Researcher	Date	Signature

1 for parent; 1 for researcher; 1 to be kept with the hospital notes

The Oxford Radcliffe Hospital
A National Health Service Trust

WTA 486

Oxford
Radcliffe

Centre Number:
Study Number:
Patient Identification Number for this trial:

PARENT CONSENT FORM

Title of the study: **A study into the adjustment of children with leukaemia**

(An investigation into psychological adjustment in children who have
been diagnosed with acute lymphoblastic leukaemia)

Name of Researchers: Caroline Paul, Trainee Clinical Psychologist
Dr Richard Scott, Consultant Clinical Psychologist
Dr Kate Wheeler, Consultant Paediatric Oncologist
Dr Chris Mitchell, Consultant Paediatric Oncologist
Dr Georgina Hall, Consultant Paediatric Haematologist

1. I confirm that I have read and understand the Information Sheet
dated..............(version......) for the above study, and have had the opportunity to ask
questions.

2. I understand that my participation in the study is voluntary and that I am free to withdraw
my consent at any time, without giving any reason, without my child's medical care or legal
rights being affected (i.e. their treatment will not be affected in any way).

3. I understand that the Researchers may look at sections of my child's medical notes, where
it is relevant to the research. I give permission for these individuals to have access to these
records.

4. I agree for my General Practitioner to be informed of my participation.

5. I agree to take part in the above study.

Name of Parent Participant	Date	Signature

Researcher	Date	Signature

1 for parent; 1 for researcher; 1 to be kept with the hospital notes

The Oxford Radcliffe Hospital
A National Health Service Trust

WTA 486

Table 1. Significant Pearson correlations – PSI child domain & SDQ and Peds child- and parent-rated QL.

SDQ scale	N	Significance (1 tailed)
Total	35	0.731***
Emotional symptoms	35	0.518***
Conduct problems	35	0.508**
Hyperactivity-inattention	35	0.695***
Peer problem	35	0.341*
Prosocial Behaviour	34	-0.440**
Impact	35	0.480**
Peds QL scale		
Child nausea	28	-0.326*
Child worry	28	-0.428*
Child cognitive problems	28	-0.389*
Parent total score	41	-0.480***
Parent psychosocial health summary	41	-0.642***
Parent emotional functioning	41	-0.513***
Parent social functioning	41	-0.481***
Parent school functioning	41	-0.518***
Parent pain	41	-0.324*
Parent procedural anxiety	41	-0.326*
Parent treatment anxiety	41	-0.523***
Parent cognitive problems	41	-0.506***

SDQ = Strengths & Difficulties Questionnaire
*p<.05; **p<.01; ***p<.001

Table 2. Significant Pearson correlations – PSI parent domain & SDQ and Peds child- and parent-rated QL.

SDQ scale	N	Significance (1 tailed)
Hyperactivity-inattention	34	0.330*
Prosocial Behaviour	33	-0.535***
Impact	34	0.393*
Peds QL scale		
Child emotional functioning	27	-0.356*
Child treatment anxiety	27	-0.328*
Child communicating with physician / nurse	27	-0.337*
Parent total score	40	-0.273*
Parent psychosocial health summary	40	-0.356*
Parent school functioning	40	-0.382**
Parent treatment anxiety	40	-0.345*
Parent cognitive problems	40	-0.426**

SDQ = Strengths & Difficulties Questionnaire
*p<.05; **p<.01; ***p<.001

Table 3. Significant Pearson correlations – PSI life stress & SDQ and Peds child-and parent-rated QL.

SDQ scale	N	Significance (1 tailed)
Emotional symptoms	35	0.361*
Peds QL scale		
Parent emotional functioning	41	-0.295*
Parent procedural anxiety	41	-0.469**
Parent treatment anxiety	41	-0.498***
Parent worry	41	-0.366**

SDQ = Strengths & Difficulties Questionnaire
*p<.05; **p<.01; ***p<.001

Table 1. Summary of the responses made to question 1. What have been the hardest parts of the whole experience for you and your family of your child's diagnosis?

Category		Number of responses		Total number
		On-treatment	Off-treatment	
Diagnosis	Initial shock & fear associated with diagnosis & prognosis	15	17	34
	Relief		2	
Family disruption, separation, isolation & strain (including financial)		14	15	29
Coping with seeing child ill (treatment & side-effects)		9	11	20
Negative impact on family	Siblings	5	5	13
	other members	2	1	
Reliance & trust in medical staff		2	1	3
Lack of family support			1	1

Some examples of participant comments for question 1 are summarised below.

Diagnosis
- 'Initially didn't know much about leukaemia & what to expect, very scared' (7 years, On).
- 'Doctor said it was tonsillitis, I thought there was more wrong, was terrified, had to visit doctor nine times, diagnosis was a relief' (7 years, Off).

Family Disruption
- 'Holding family together, trying to be in two places at once' (8 years, On).

Coping with seeing child ill
- 'Coping with change in....., dramatic changes in personality, loss of hair, inability to do anything for self, always tired, couldn't walk, needed 24 hour care & attention', (7 years, Off).

Negative impact on family
- '(Sibling) worried that ..... will die, not the same brother as before, can't play & more aggressive when on treatment' (7 years, On).
- 'We (parents) didn't get on, I (mother) dwelt on diagnosis, while the father denied it' (9 years, Off).

Table 2. Summary of the responses made to question 2. What have been the hardest parts of the whole experience for your child of their diagnosis?

Category	Number of responses		Total number
	On-treatment	Off-treatment	
Treatment (& condition) frightening & painful	11	13	24
Coping with side-effects	9	9	18
Isolation from friends & family	6	8	14
Uncertainty and fear of future / prognosis	6	6	12
Hospital visits and interaction with medical staff	5	1	6
Missing school	4	1	5
Being different from others (sibling & peers)	3	1	4
Relapsing	1		1
Additional family stressors (e.g. divorce)		1	1
Making friends in hospital		1	1

Some examples of participant comments for question 2 are summarised below.

Coping with side-effects
- 'Accepting way she looked very difficult – bald, round & yellow ('chemo-grey')' (6 years, Off).

Hospital visits
- 'Doctors / nurses prodding and poking, he didn't know what they were doing' (7 years, On).

Isolation
- 'Missing out on things – school, parties, friends (6 years, Off).

Uncertainty
- 'Thought he was going to die, still thinks this on & off, needs a lot of reassuring' (7 years, On).

Missing school
- 'Frustration at being unable to get on with normal life, always 'new girl' at school' (10 years, On).

Being different
- 'Found it difficult being different from other, unable to do swimming' (6 years, On).

Table 3. Summary of the responses made to question 3. What has been the most helpful support given to you and your family?

Category	Number of responses		Total number
	On-treatment	Off-treatment	
Medical & nursing staff	18	20	38
Social worker	12	10	22
Friends & Family	8	5	13
Other parents	3	4	7
Support group	2	1	3
Psychological support / Counselling	2	1	3
Charities	2	1	3
Playleader	1	1	2
Additional support (e.g. GP, vicar, school/ nursery)	2	3	5

Some examples of participant comments for question 3 are summarised below.

Medical & Nursing Staff
• '(Consultant) very supportive' (7 years, Off).
• 'Nurses very good, easy to ask questions, approachable, always on end of phone, put mind at ease' (7 years, Off).
Friends
• 'Friends who've looked after animal, packed bags, offered practical support' (3 years, Off).
Other parents
• 'Support from other parents at clinic, best emotional support' (4 years, On).
Charities
'CLIC provided house in Oxford, enabled four of us to stay together' (3 years, On).

Table 4. Summary of the responses made to question 4. What has been the most helpful support given to your child?

	Number of responses		Total number
Category	On-treatment	Off-treatment	
Medical / nursing staff & social workers	8	11	19
Family	10	7	17
Play room / therapist	6	9	15
Friends	2	3	5
School / nursery – in & out of hospital	2	3	5
Individual responses: special events on ward, professionals (e.g. clinical psychologist, art therapist), respite & High Dependency Unit	2	3	5

Some examples of participant comments for question 4 are summarised below.

Medical / nursing staff
• 'MacMillan nurses and medical staff good at answering questions that I couldn't answer' (8 years, Off).
Play room
• 'Nice play room, feels at home, accessible, not daunting' (6 years, On).
Friends
• 'Friends of same age, understood is she's too tired & can't play, & visit in hospital' (4 years, On).

Table 5. Summary of the responses made to question 5. What could have been done to improve the support given to you and your family?

Category	Number of responses On-treatment	Number of responses Off-treatment	Total number
Nothing	9	9	18
Misinformed / lack of information regarding diagnosis, treatment & entitlements	5	4	9
Emotional support for parents	3	3	6
Would have liked all care in Oxford	2	3	5
Support groups (needed or existing ones improved)	1	3	4
Improvements with current service (i.e better accommodation for parents, reduced waiting times, more access to play specialist, more visits by community nurses, more practical support & access to respite)		5	5
Need to assess family's individual needs		1	1
National register for children enabling support to come to families	1		1
Individual responses: would have liked to have been shown how to take blood earlier, information regarding study could have been given in a less blunt, cold manner, & need council to re-house family.	2	1	3

Some examples of participant comments for question 5 are summarised below.

Lack of information
- 'Wanted to be more informed of treatment, downs as well as ups, explanations of why medication was needed and what if it doesn't work. Wanted information about whole protocol. Needed to ask nurses for explanation due to doctors not explaining things fully' (6 years, Off).

Emotional support for parents
- 'No emotional support for parents (e.g. counsellors), would have liked someone to talk to' (3 years, Off).

Care in Oxford
- 'Would have liked all care at Oxford, care in district general not as good, not geared for sick children' (3 years, On).

Table 6. Summary of the responses made to question 6. What could have been done to improve the support given to your child?

Category	Number of responses		Total number
	On-treatment	Off-treatment	
Nothing	12	12	24
Psychological input / support, key worker	2	3	5
More access to play specialist		3	3
Meal times & food more child focussed		2	2
Individual responses: More information about how to tell child diagnosis, nurse (not child) should tell school of their diagnosis/ treatment, felt treatment could have been administered more gently, council needs to re-house family, & hospital need more funding for oncology support workers.	5		5

Some examples of participant comments for question 6 are summarised below.

> **Meal times**
> - 'Need to be more flexible, always hungry when on steroids' (6 years, Off).
>
> **Key worker**
> - 'Would have liked constant friend / nurse (allocated key worker) to trust throughout treatment (7 years, Off).