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Family Impact

and Infant Emotional Outcomes

when an Infant Has Serious Liver Disease:

A Longitudinal Mixed Methods Study

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In memory of my father,
Kenneth Russell Bowden.
He taught me the value of education and provided me
every opportunity to achieve my full potential.

Declaration

This thesis is submitted to the University of Sydney in fulfilment of the requirements for the degree of Doctor of Philosophy.

To the best of my knowledge and belief, the work presented in the thesis is original. I hereby declare that I have not submitted this material, either in full or in part, for a degree at this or any other institution.

Michael R Bowden
01 December 2015

Abstract

Background

Serious liver disease in infancy causes significant morbidity. Up to 80% of children will eventually require transplantation. This study aims to investigate parent and family responses to the diagnosis of serious liver disease in infancy and to identify family factors that are predictive of the infants' emotional and behavioural outcomes.

Methods

The study uses quantitative and qualitative methods. Parents of infants recently diagnosed with serious liver disease completed validated measures of parent stress, family function, impact of the illness on the family, and father engagement, as well as an interview about their experience of the infants' illness. The measures were repeated after one year, with the addition of the Child Behavior Checklist (CBCL).

Results

Parents of 42 infants enrolled, and parents of 37 infants completed the study. Illness severity, liver diagnosis other than Biliary Atresia and parent perceptions of greater impact of the infants' illness on the family predicted poorer infant outcomes. For mothers, the final best-fit model explained 32% of the variation in CBCL ($P = .001$). Fathers' best-fit model explained 44% of the variation in CBCL ($P < .001$).

Thematic analysis of the parent interviews revealed six major themes: uncertainty; awareness of the infant's vulnerability; feelings of isolation; dealing with other aspects of life; the importance of shared experience; and adjustment.

The integrated data analysis demonstrated that lack of extended family support, poor family adjustment to the illness, and financial stress are related to greater impact of the illness on the family.

Conclusions

The study identifies early risk factors for poor emotional and behavioural outcomes for infants with serious liver disease, providing an opportunity for early intervention. Parents who lack support from extended family, who have financial stress, or who report a high impact of the illness on the family, should be referred for psychosocial assessment.

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Preface

The research study presented in this thesis was undertaken by the author under the supervision of Professor Philip Hazell, Concord Clinical School, Discipline of Psychiatry, Sydney Medical School, The University of Sydney.

The research was undertaken at The Children's Hospital Westmead, Sydney Children's Hospital, Royal Children's Hospital Brisbane and Royal Children's Hospital Brisbane. Ethics approval was granted by the Human Research Ethics Committee, Concord Repatriation General Hospital, The Queensland Children's Health Services Human Research Ethics Committee, and the Royal Children's Hospital Melbourne Human Research Ethics Committee (Appendix A).

The author was responsible for all aspects of the study. The author planned, designed and carried out the research, including recruitment of participants, data collection and analysis. The author synthesised the results, and wrote and revised the thesis. The author wrote and revised the manuscript submitted for publication in a peer-reviewed journal, as listed below and contained in Appendix B. The author prepared and presented talks and posters at conferences in Australia and internationally, as listed below.

Publications Arising from the Research Study

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Bowden MR, Hazell P. Poster presentation, 'On the Roller-Coaster: Parent Stress in Infant Liver Disease'. 14th WAIMH World Congress, Edinburgh UK. June 2014

Bowden MR, Hazell P. Oral presentation, 'Family Adjustment when an Infant has a Serious Illness: Putting Research into Practice'. 16th International Congress of ESCAP, Madrid Spain. June 2015

Bowden MR, Hazell P. Poster Presentation, 'Family adjustment and parenting stress when an infant has serious liver disease: an Australian experience'. Royal College of Psychiatrists, International Congress 2015, Birmingham, UK. June 2015

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List of Abbreviations

CBCL	Child Behavior Checklist
CHW	Children's Hospital at Westmead
CNS	Central Nervous System
DADS	Dads' Active Disease Support scale
DASS	Depression Anxiety Stress Scales
ESLD	End-stage liver disease
FAD	Family Assessment Device
FSIQ	Full Scale Intelligence Quotient
HMR	Hierarchical multiple regression
HREC	Human Research Ethics Committee
HRQOL	Health-related quality of life
IFS	Impact on Family Scale
JRA	Juvenile Rheumatoid Arthritis
PELD	Pediatric end-stage liver disease
PIQ	Performance Intelligence Quotient
PTSD	Post-Traumatic Stress Disorder
RCHB	Royal Children's Hospital Brisbane
RCHM	Royal Children's Hospital Melbourne
SCH	Sydney Children's Hospital
SEI	Socio-Economic Index
SES	Socio-economic status
VIQ	Verbal Intelligence Quotient

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1 Introduction

1.1 Synopsis

Serious liver disease presenting in infancy is an example of life-threatening chronic illness with long-term implications for the infant and the family. It is associated with significant morbidity leading to developmental and emotional problems for the child and disruption for the family. Liver transplant is required for many of these cases and has good surgical outcomes. However, the post-transplant course can be variable and children need to remain on lifelong medication.

Despite commonalities between different chronic illnesses, such as functional impairment and visibility of the illness, there has been increasing research interest into the specific effects of individual childhood illnesses. Several meta-analyses have now demonstrated large differences in emotional and behavioural outcomes depending on illness type. Worse emotional and behavioural outcomes are found when the illness affects central nervous system function, when there is functional impairment associated with the illness, and when the illness persists throughout childhood. Serious liver disease in childhood is characterised by each of these factors.

This chapter begins with an outline of the physical and developmental consequences of liver disease in childhood. A description of the treatment services available in Australia is then presented. Theoretical approaches to the study of chronic illness in children are presented, outlining the evolution of thinking in this area. Methodological issues in studying the psychosocial effects of serious illness in infancy are then discussed. Research into the psychosocial aspects of serious liver disease in infancy and childhood is then presented, including the areas of health-related quality of life and emotional, behavioural and psychiatric aspects of liver disease in this age group.

Research examining parent distress, family functioning and father engagement in the care of infants and children with liver disease is presented next, followed by a discussion of the limitations of research to date. Finally, the aims and research hypotheses of the current study are stated.

1.2 Serious Liver Disease in Infants

1.2.1 Physical Aspects of Serious Liver Disease in Infants

Serious liver disease is defined in this thesis as liver disease that may result in the need for liver transplantation. Serious liver disease in infants is rare, with an overall incidence rate of approximately one in 2,500 live births.[1] The majority of cases in childhood are diagnosed early in infancy. The most common identified liver disorders are biliary atresia, metabolic diseases (such as alpha-1 antitrypsin deficiency and citrullinemia), acute fulminant hepatic failure (such as cryptogenic hepatic failure and autoimmune hepatitis), Alagille Syndrome and malignancy (such as hepatoblastoma).[2,3]

Serious liver disease is associated with significant morbidity, including ascites, gastrointestinal bleeding, encephalopathy, endocrine abnormalities and renal dysfunction.[4] Infants with cholestatic disorders such as biliary atresia and Alagille Syndrome are at risk of chronic complications due to bile retention in the liver. Bile retention causes direct damage to hepatic metabolic function, with progressive liver damage, biliary cirrhosis, portal hypertension and eventual liver failure.[5] Lack of bile secretion to the proximal intestine results in poor absorption of dietary fat and fat-soluble vitamins (vitamins A, D, E and K) leading to long-term problems with growth, metabolic bone disease, neuromuscular problems and blood clotting difficulties. Accumulation of bile salts in the tissues can also lead to severe and distressing pruritus.[5]

Biliary atresia is the most common liver disease presenting in infancy. It has an incidence of between 1/10,000 and 1/15,000 live births, the most common form of which develops in the early postnatal period and consists of obliteration of the extra-hepatic biliary tree.[5] The cause is unknown, but it is thought to be secondary to perinatal insult, such as infection, leading to an immune response and damage to the bile ducts.[5,6] Treatment requires that the hepatoportoenterostomy surgical procedure (known as the Kasai procedure) is performed to allow bile drainage. The Kasai procedure is most effective if performed before the infant is 8 weeks old.[5] Most children with biliary atresia continue to have inflammation of the biliary tree and eventually develop portal hypertension and hepatic failure.[5,6] Between 60 and 80% of children with biliary atresia will eventually need a liver transplant.[1,6] Biliary atresia is the most common indication for paediatric liver transplantation internationally.[2,6-9] In 2013, biliary atresia accounted for 55.6% of all paediatric liver transplants in Australia and New Zealand.[2]

The next most common liver diseases in infants and children are metabolic disorders such as Wilson's disease (copper storage disease), neonatal hemochromatosis (iron storage disease) or α_1-antitrypsin deficiency. Metabolic disorders accounted for 14.2% of all paediatric liver transplants in Australia and New Zealand in 2013.[2] These disorders have variable presentations, treatment and prognosis. Metabolic disorders commonly induce hepatic injury and liver failure, through alterations to the storage of lipids, glycogen or other substances such as copper.[10] For many of these conditions liver transplantation can be curative.[10] The next largest group of conditions leading to the possible need for liver transplantation in infants and children is fulminant hepatic failure, which accounts for 11.1% of paediatric liver transplants in Australia and New Zealand.[2]

At the time of writing this thesis, paediatric liver transplantation in Australia and New Zealand has a 1 year survival rate of 89%, with 84% of children surviving for 5 years post-transplant.[2] Similar survival rates have been reported from the United States [9] and the United Kingdom.[8] Of all liver transplants performed for children under the age of 16 years in Australia, 20 to 25% are performed on children aged under 12 months.[2]

1.2.2 Developmental Aspects of Serious Liver Disease in Infants

There is a gap in the knowledge base regarding the developmental aspects of serious liver disease in infancy, most research focussing on children's development following liver transplantation. To date, much of the research has focused on the effects of the illness on cognitive functioning, motor skills development and, in older children, academic outcomes.

1.2.2.1 Cognitive, motor skills development and academic outcomes in infants and children with serious liver disease

Limited research has been undertaken into the cognitive functioning of infants and children with serious liver disease prior to liver transplant. Although most studies have small sample sizes it has been consistently demonstrated that prior to liver transplant, children with liver diseases have significant developmental problems. Earlier onset of illness and greater severity of illness are significantly correlated with poor developmental outcomes as outlined below.

Three studies from the University of Texas have highlighted that chronic liver disease results in cognitive and motor development problems in infants and children with biliary atresia and end-stage liver disease.[11-13] Stewart and colleagues found that onset of liver disease prior to age 12 months is significantly correlated with worse cognitive outcomes when compared to those with later childhood onset of illness,

regardless of the severity of the liver disease. Infants and children with early onset exhibit cognitive problems that positively correlate with duration of illness.[12]

In addition, infants and children with early onset illness or those who do not receive a liver transplant display cognitive problems that persist into later childhood, when compared to both normative samples and children with another chronic childhood illness, Cystic Fibrosis.[13]

Although the number of participants is small in all of the reviewed studies, the findings suggest that the infant brain may be more vulnerable than that of older children to the metabolic effects of chronic liver disease.[12,14] These studies also raise the question as to whether or not early onset of liver disease has a negative impact on cognitive development that may not be found in other chronic illnesses early in life.

Reed-Knight and colleagues assessed the intellectual and academic performance of 195 children with chronic liver disease, renal disease and heart disease who were being evaluated for solid organ transplantation. The study group included 55 children with chronic liver disease with a mean age of 12.79 years (*SD* 3.60). The children in this study had significantly lower intellectual and academic functioning compared with population norms. The children with liver disease in particular had lower IQ than the normal population ($d = 0.51$, $P = .001$), poor word reading ($d = 0.31$, $P = .04$) and poor math computation ($d = 0.49$, $P = .002$).[15] The findings support the earlier studies that suggest a direct deleterious effect of chronic liver disease on cognitive outcomes and indicate there may be a persistence of the problems with continuing illness.

Motor and language development are other areas that may be affected by liver disease arising in infancy. Burgess and colleagues examined 20 infants who had the Kasai procedure for biliary atresia without a liver transplant. They found that the infants had normal cognitive development but borderline motor development in the

first 8 months of life, with worsening performance in both cognitive and motor development thereafter.[16] Hopkins and colleagues studied 14 infants with biliary atresia at pre-transplantation using the Bayley Scales of Infant Development. The investigators found the infants' mean motor development scores and mean mental development scores to be significantly lower than test norms.[17] Caudle and colleagues conducted two studies of infants who had biliary atresia who had not had a transplant. They reported significant developmental problems in the infants, including reduced gross and fine motor skills development, impaired visual reception, and expressive and receptive language difficulties in the infants with biliary atresia when compared to test norms. Overall, the authors concluded that increasing severity of liver disease is significantly correlated with higher levels of impairment in both gross and fine-motor skills.[18,19]

In Summary, research to date appears to indicate a correlation between severe liver disease in children and infants and a wide range of difficulties in cognitive, developmental and academic delay.

1.2.2.2 Cognitive, motor skills development and academic outcomes in infants and children following liver transplant

There is an extensive knowledge base about cognitive and academic outcomes in children post-liver transplant. In overview, early studies that examined the cognitive outcomes for children who have had a liver transplant typically concentrated on pre- and post-transplant comparisons. These studies indicate that there is no change in cognitive performance between the pre- and post-transplant period.[20,21]

Studies of children up to 9 years post-transplant have similarly shown continued poor cognitive outcomes in infants and children with liver transplants, indicating high rates of delayed intellectual development in children with liver disease compared with population norms.[22-26] However, small sample sizes, differences in the ages of the

children, variability of cognitive assessment instruments used and heterogeneous results for individuals in the sample make the results difficult to interpret.

A multi-centre study found clear evidence of cognitive delay, academic difficulties and executive functioning problems in 144 children aged 5 to 7 years of age who had had a liver transplant at least 2 years previously. Findings included significantly reduced Full Scale IQ (FSIQ), Performance IQ (PIQ) and Verbal IQ (VIQ) scores in the liver transplanted children compared with test norms, as well as executive functioning difficulties and working memory problems.[27] The same group of children was assessed 18 to 36 months later, with a 65% follow up rate. The investigators found that the intellectual, academic and executive functioning deficits persisted over time, though reading skills had improved to within the normative range.[28]

An Australian study, limited by a small sample size of 13 patients matched with 6 sibling controls, reported similar findings of FSIQ within the normal range but executive functioning problems persisting at least 5 years post-transplant.[29] Kaller and colleagues also reported similar findings in their study of 137 children who had had a liver transplant at least 12 months previously. They reported problems in attention and executive functioning in this group of children.[30]

Pinquart and Teubert conducted a meta-analysis of 954 studies (a total number of 104,867 children) published before November 2011. They examined the academic, physical and social functioning of children with chronic physical illnesses, compared with either healthy controls or normative data. The analysis included 45 studies of children with chronic kidney or liver disease (the number of participants was not reported for the subgroup). It is not possible to separate the findings for liver disease from those for kidney disease in the meta-analysis. However, the authors reported a moderate to large effect size (ES) of -0.78 (Confidence interval [*CI*] -0.90 to -0.66) for poor academic functioning in the group of children with chronic kidney or liver

disease, indicating that significant academic impairments are associated with these chronic conditions. In addition, there was little heterogeneity between samples, indicating consistency in the findings across studies of these conditions.[31]

Primary type of liver disease is also a significant factor in children's developmental outcomes. For example, metabolic disorders are associated with brain damage and delayed mental and psychomotor development pre-transplant, with improvements in psychomotor development, but not in mental development, two years post-transplant. Children with metabolic disorders show significantly poorer mental development when compared with children who had transplants due to biliary atresia.[32]

1.3 Infants and Children with Serious Liver Disease: the Australian Context

Children residing in the state of New South Wales (NSW) who have serious liver disease diagnosed in infancy are managed in one of two tertiary treatment centres in Sydney, located at the Children's Hospital at Westmead (CHW) or at the Sydney Children's Hospital (SCH) at Randwick. In other states in Australia children with serious liver disease are treated in the capital city tertiary children's hospital.

Australia has three paediatric liver transplant units, located in Sydney, Melbourne and Brisbane. Children from other regions of Australia who require a liver transplant must travel to one of these major metropolitan centres. The Children's Hospital at Westmead (CHW) (located in Sydney) provides liver transplantation for children from throughout NSW, from South Australia, and also from Western Australia.

Children with serious liver disease typically experience prolonged periods of illness, have multiple hospital admissions for medical and/or surgical interventions, and become increasingly unwell over an extended period of time before proceeding to transplant. If transplantation is required, hospital admission for the transplant itself is

usually of the order of 4 to 6 weeks, but children and families typically are required to remain in close proximity to the hospital for follow up treatment for several more months after discharge (personal communication, O'Loughlin EV, Stormon M, 2015).

A review of current clinical practice revealed that psychosocial assessment at the point of diagnosis of serious liver disease is not routine at any of the specialist liver transplant units in Australia (personal communications, Hardikar W. 2015, Ee L. 2015, Krishnan U. 2015.). Rather, referral is made to the psychosocial team only if the treating medical team considers that the infant's parents are particularly distressed. With respect to preparation for paediatric liver transplantation, practices vary across treatment centres in Australia, dependent on the availability of psychosocial staffing resources in each hospital. Within Australia, the transplant service at CHW is the only service that includes routine psychosocial assessment of the child and family by a Child Psychiatrist and/or Child Psychologist and a Social Worker. Assessments typically occur when an infant or child is being actively considered for liver transplantation. Variability in the course of liver disease in this age group often results in the assessment occurring several years after the infant's initial diagnosis.

The current PhD study arose from the author's experience working as a Child Psychiatrist consulting to the liver transplant team at CHW. Consultation at the time of the child's assessment for transplant revealed that many of the children presented with long-standing but previously unidentified and untreated emotional distress. In addition, parents and families frequently reported ongoing distress as a result of the child's illness. The clinical experience therefore raised the question as to whether or not earlier intervention would be useful in the routine management of these children and families, with the aim of improving long-term outcomes for both the children and their families following the diagnosis of serious liver disease in infancy.

1.4 Approaches to Studying Chronic Illness in Children and Families

Before examining the psychosocial aspects of serious liver disease in infants, a brief description of the main theoretical approaches to studying chronic illness in children and families will be presented. The section begins with an overview of the findings from epidemiological studies, followed by a summary of the main theoretical approaches. The study of chronic illness in childhood has focussed mostly on children and adolescents, with little work conducted into the responses of families to a new diagnosis in infancy. Despite the influence of theoretical models such as the non-categorical approach to studying chronic illness described below, recent scientific evidence shows clear differences between chronic illnesses in relation to their impact on the child. The research findings therefore justify the study of specific individual illnesses such as serious liver disease.

1.4.1 Epidemiology of Chronic Illness in Children

Epidemiological studies have shown that chronic illnesses are common in childhood despite the rarity of many individual illnesses. As outlined below, chronic illness in childhood is associated with problems in psychosocial adjustment, especially when there is functional impairment and Central Nervous System involvement. If the illness persists, adult psychosocial adjustment is also affected. As noted previously, serious liver disease in infancy is characterised by all three of these features, resulting in elevated levels of risk for the ongoing adjustment of affected infants.

Pless and Roghmann analysed the data from three major epidemiological surveys conducted from the 1940s through the 1960s, the National Survey of Health and Development, the Isle of Wight study and the Rochester Child Health Survey. The rates of chronic illness varied between the studies due to differences in the definition of chronic illness used in each study. The analysis found that approximately 10% of

children experienced at least one chronic illness by the age of 15 years. Up to 30% of the chronically ill children were found to suffer from psychosocial maladjustment. Difficulties in behavioural and psychological adjustment were found to be greater in children whose illness was persistent and in those children with functional impairment. The authors identified the importance of family structure and function and also saw poor self esteem as a mediating factor in the development of psychosocial problems in the children. The analysis demonstrated that the management needs of chronically ill children were different from those of children with acute disorders.[33]

Epidemiological studies have also provided data for long-term outcomes and follow up, in order to assess whether adjustment problems due to chronic childhood illness continue into adolescence and adulthood. Follow up data from the National Survey of Health and Development,[34,35] the Rochester Child Health Survey[36] and the National Child Development Study[37] each found that persistence of childhood chronic illness was associated with increased rates of psychosocial problems in older adolescents and young adults. Although there was some evidence that many of the differences between the chronically ill group and healthy individuals were no longer apparent by the time the participants were aged in their mid-30s, this was only the case for adults whose chronic illness had resolved by early adulthood. The adults whose illness persisted through adulthood continued to suffer significantly higher rates of psychiatric disorder and social difficulties.[34]

The Ontario Child Health population study also found high rates of psychiatric disorders in chronically ill children compared with healthy children, particularly when the chronic illness was accompanied by disability. Children with functional impairment were more than three times more likely to have a psychiatric disorder

than healthy children. Those with a chronic illness without associated disability were more than twice as likely as healthy children to have a psychiatric disorder.[38]

The Bergen Child Study provided further evidence of increased rates of psychosocial problems, particularly amongst children whose chronic illness led to functional impairment or when there was CNS involvement.[39,40]

A population study from the United States, using data from the National Health Interview Surveys, Disability Supplement, demonstrated that functional impairment was demonstrated to lead to adverse emotional and behavioural outcomes when combined with family stress, maternal distress or poverty.[41]

These epidemiological studies are important in providing critical evidence that chronic illness in children is common and is associated with significant psychological morbidity. The studies also demonstrated that illnesses that persist into adulthood, such as chronic liver disease, are associated with significant long-term psychological morbidity through adolescence and into adulthood. Studying serious liver disease in infancy therefore offers the possibility of identifying early risk factors for later psychological morbidity and the opportunity to devise preventive strategies.

1.4.2 The Non-Categorical Approach to Chronic Childhood Illness

Early researchers focussed on psychopathology arising in the child as a result of chronic illness. The child's response to the illness was conceptualised as being influenced by the illness and the family's reactions, resulting in behaviour changes and psychiatric illness.[42,43] The child's resultant psychopathology could, in turn, lead to complications in the illness, such as fluctuations in blood glucose control in adolescents with diabetes associated with rebellious behaviour[44] and depression.[45] There were unsuccessful attempts to link specific psychopathological outcomes to specific disorders.

Stein and Jessop proposed a 'non-categorical' approach to studying children with chronic illness.[46,47] The approach was built upon the earlier work by Pless and Pinkerton[48] that noted the common problems encountered by chronically ill children and their families regardless of the child's diagnosis. Although recognising that different diseases present their own specific issues, Stein and Jessop argued that the commonalities across disease categories allows for health workers to generalise past experience with specific disorders to different types of chronic conditions. These 'generic dimensions' of illness include aspects of the illness such as the visibility, stability or life-threatening nature of the condition, the amount and intrusiveness of care required, the presence of associated mental retardation, and whether there is involvement of sensory or motor systems. This model has demonstrated the importance of functional impairment, child adjustment and service needs of children who suffer from rare illnesses. Given the rarity of many chronic illnesses in childhood, the non-categorical approach assists with the development of health services to meet the needs of children presenting with a range of diverse conditions. For example, psychological management, prevention and rehabilitation services could focus on adjustment and functional impairment regardless of the child's primary illness. Stein and Jessop also argued that the non-categorical view is less stigmatising to people with physical illnesses, as problems associated with the illness could be seen along a continuum between healthy and severely ill.[46] Using data from both a population study and from an institutional study, they reported that there was 'as much or more variation *within* the diagnostic groups as there is *between* them'[47] (italics in original). This social science view was at odds with prevailing medical views, but has been influential over much research into the psychosocial effects of chronic childhood illness and has contributed to a new way of examining chronic illness in children.

Lavigne and Faier-Routman highlighted the limitations of the non-categorical view. They noted overlap between the identified dimensions of illness and the likelihood that children with chronic conditions will score highly on at least one of the dimensions. The non-categorical theory does not define the relative importance of each dimension, or how the dimensions may interact to produce maladjustment. The model therefore provides limited value for discriminating differing levels of need or focussing intervention.[49] While the non-categorical approach may be most helpful in service design for management of rare disorders, it also provides a helpful framework for assessing the illness issues facing children and families with specific rare disorders.

1.4.3 The Risk and Resistance Model

Research attention to adaptation in both child and parents led to chronic illness being seen as a stress that increased the risk of adjustment problems, but only in interaction with other variables including the child's development, and the family and social context.[50-52]

Wallander and Varni developed a 'risk and resistance' model of childhood chronic illness, in which the disorder itself, any associated disability, functional dependence and psychosocial stressors represent risk factors for maladjustment. The risk and resistance model emphasises the importance of the developmental perspective. Wallander and Varni noted that although maladjustment is common in chronically ill children, the majority of sick children are resilient. In this model, resistance factors are categorised as intrapersonal (such as temperament), socio-ecological (such as family environment) and stress processing (such as cognitive processing).[53,54] Wallander and Varni's model therefore builds on the non-categorical approach, by contextualising the child's adaptation to illness through an understanding of systems theory and child development. The model fits within a systems framework: the

individual child's adjustment arises from the interaction between the illness, the child's characteristics and the child's social environment. A more complex view of childhood chronic illness is therefore taken that includes the developmental perspective, individual adaptation, family functioning and socialisation.

1.4.4 Childhood Illness as Trauma for the Child and the Family

Medical illnesses were first included as possible triggers for the development of Post-Traumatic Stress Disorder (PTSD) in 1994 in DSM-IV.[55] This nosological development resulted in a significant increase of studies of PTSD due to chronic childhood illness being published in the late 1990s and 2000s.[56-62] Chronic illness was studied as a source of chronic trauma for the child and parents, with the development of PTSD being dependent on the interaction between a complex set of stressors: the specific impact of the illness on child and parents; the interaction between these impacts; child and parent coping strategies; and direct effects of the illness or its treatment.[58] Researchers attempted to identify risk factors for the development of PTSD in these children, noting the importance of subjective appraisal of threat rather than objective measures of the threat per se, demographic variables and parent stress amongst others.[59,60] PTSD in these children was conceptualised in a systemic, social ecological and developmental model. The model took into account the wide range of normative reactions to trauma, pre-existing psychological issues for individual children, their parents and families, the child's developmental stage, illness and treatment characteristics and wider social supports.[59]

Cabizuca and colleagues conducted a meta-analysis of 16 studies published prior to January 2006 (total number of participants not reported) examining PTSD in parents of children with chronic illness. The meta-analysis included one study of parents of children who had had a solid organ transplant. The meta-analysis found a pooled prevalence of current PTSD for both parents of 22.8% (N = 1845, 95% CI = 16.4 to

29.0%). However, the pooled prevalence of current PTSD in mothers was 19.6% (*N* = 941, 95% *CI* = 14.3 to 24.9%), significantly higher than the pooled prevalence of current PTSD in fathers of 11.6% (*N* = 429, 95% *CI* = 6.4 to 16.7%). Four studies, all of mothers of children with cancer, assessed mothers separately in comparison to mothers of healthy controls and found that the proportion of mothers with PTSD was more than four times higher in the mothers whose children had cancer than in the mothers of healthy children. The meta-analysis also included one study that assessed fathers of children with cancer compared with fathers of a healthy control group. The study found PTSD in 7.1% of fathers of children with cancer and no cases of PTSD in the fathers of the healthy control group.[63]

Parent psychological distress has been identified as a risk factor for PTSD in traumatised children, [64,65] and parental PTSD is associated with child distress whether or not the child has experienced a traumatic event.[66]

Children may show positive adaptation to illness and so-called post-traumatic growth. Phipps and colleagues demonstrated that many children with cancer and other chronic diseases were well adjusted. The researchers suggested a 'repressive adaptive' style as a resilience factor in such children. In this model, measures of anxiety and defensiveness are used to allocate children into one of four groups (high or low anxiety combined with high or low defensiveness). Repressors are defined as children who are high in defensiveness but low in anxiety, a pattern commonly found in children with cancer. The repressive adaptive style was found to be associated with good psychosocial outcomes in these children and was not associated with adverse health outcomes, hence was seen as a pathway to resilience.[67-70] Research into resilience following diagnosis of serious physical illness in a child has been extended more recently to examine resilience factors in parents and families, demonstrating a similar pattern.[71,72]

In summary, PTSD is common in children with chronic illness and in their parents. Mothers are more likely than fathers to develop PTSD, but rates of PTSD in both parents are higher than in healthy populations. Parental PTSD is associated with child distress and child PTSD regardless of whether the child has experienced a traumatic event. In the situation of early childhood serious illness, rates of PTSD can be expected to be high in both the child and the parents. However, there is also recent research demonstrating positive adaptation of both children and parents to the adversity of serious illness in children.

1.4.5 Psychological Effects of Chronic Illness

Researchers have found increased rates of psychosocial problems in children with chronic illnesses compared with healthy controls,[36,73-76] particularly when the illness affects central nervous system (CNS) function.[77-79] Poor psychological outcomes are also associated with disease severity, functional disability, and family stress.[80]

Adjustment problems and internalising symptoms have been demonstrated in children with a range of chronic childhood illnesses including asthma,[81] arthritis,[82] renal failure,[74] diabetes,[83] and cancer.[84,85] Psychiatric disorders are more common in chronically ill children whose parents report high levels of personal stress and low levels of support.[74]

Research has also examined specific emotional outcomes in children with chronic illness. Meta-analyses have been conducted into the prevalence of depression,[86,87] anxiety,[88] internalising and externalising problems,[89] social competence,[90] body image disturbance,[91] and impaired self esteem.[92]

Strikingly, each meta-analysis has demonstrated differences in emotional outcomes depending on disease type. For example, Pinquart and Shen conducted a meta-analysis of 340 studies published up to September 2010, examining depressive

symptoms in a total of 33,047 children with chronic illnesses. The authors found a small to very small overall effect size (*d* = 0.19, 95% *CI* = 0.15 to 0.23) for depressive symptoms in chronic illness. However, they found much higher rates of depression in illnesses with CNS involvement (for example the effect size for epilepsy was 0.39), craniofacial abnormalities (*d* = 0.54, 95% *CI* = 0.21 to 0.86) and chronic fatigue syndrome (*d* = 0.94, 95% *CI* = 0.67 to 1.21).[87] The latter may be due to the significant overlap between chronic fatigue and depressive symptoms. Interestingly, the meta-analysis revealed no significant difference in children with cancer or diabetes compared with either healthy controls or normative data.

Despite the commonalities that are present across disorders, as emphasised by the non-categorical approach to studying chronic illness in children, these meta-analyses demonstrate that individual chronic illnesses have unique patterns of association with mental health problems that warrant investigation. Illness-specific factors are important in providing individualised care to children with chronic illness.[93]

More recently, research has focussed on longer-term psychosocial outcomes, often using multi-centre trials to generate larger sample sizes to overcome recruitment issues for uncommon diseases. Such research has identified that improved survival rates may result in developmental problems, psychological adjustment problems or physical morbidity that may persist throughout childhood and affect adult adjustment.[94,95] However, positive long-term psychological outcomes of illness in childhood, such as post-traumatic growth, have also come to attention.[96]

Child health problems at age 2 to 3 years, along with parenting stress and family psychopathology, have been identified as risk factors for the development of internalising problems at age 11 years.[97] The finding suggests that when chronic illness, such as serious liver disease, affects younger children, it is important to

identify parent and family risk factors as possible targets of intervention to prevent later poor psychosocial outcomes for the children.

In summary, chronic illness in childhood is associated with poor psychosocial outcomes, particularly the development of internalising problems such as depression. The risk of psychological maladjustment is increased when the illness is severe, when there is CNS involvement, and in the presence of functional impairment. Parent stress, lack of family support, and family functioning problems are associated with an increased risk of psychosocial problems in the children. Illness that persists into adulthood is associated with poor adult adjustment. Meta-analyses have demonstrated large differences in outcomes for children depending on disease type, suggesting that studying individual illnesses is important in terms of intervention. Illness in very young children is associated with later internalising problems. Serious liver disease in childhood is associated with many of these risks for poor outcomes. Examining parent stress and family functioning in these children's families can provide further information about possible targets for intervention.

1.4.6 Adjustment and Coping in Parents and Families of Children with Chronic Illness

Parenting stress has been demonstrated to be higher amongst parents of children with chronic illness than in parents of healthy control children. Effect size varies according to disease, thus it is important to study parent distress in individual illnesses. A meta-analysis conducted on 13 studies published between January 1980 and June 2012 (total number of participants not reported) demonstrated an overall effect size of $d = 0.40$, 95% $CI = 0.19$ to 0.61, $P <= .0001$. The meta-analysis included studies of children with asthma, cancer, cystic fibrosis, diabetes, epilepsy, juvenile rheumatoid arthritis and sickle cell disease. Effect sizes ranged from -0.30 to 0.88 across the 13 studies that were included in the meta-analysis.[98]

The timing of research in the illness cycle is important. For example, in a meta-analysis of 29 studies published up to 2005 (total number of participants not reported), mothers and fathers of children with cancer were demonstrated to be significantly more distressed than parents of healthy children at the time of the child's diagnosis. However, there was no significant difference in levels of distress between the parents of children with cancer and the parents of healthy children one year after the diagnosis. The meta-analysis also demonstrated that mothers were significantly more distressed than fathers (mean effect size -0.23, 95% *CI* = -0.31 to -0.15, $P <$ .05), an effect that was maintained for a year after diagnosis.[99]

Family functioning and social support have been shown to interact with the level of disease activity to affect children's adjustment to illness,[100-103] suggesting that research into child and family characteristics may identify pathways to child adjustment.[80] However, uncertainty remains about the interaction between family functioning and social support in children's adjustment to chronic illness. For example, Robinson and colleagues failed to find a contribution of social support to children's adjustment to cancer despite finding that family functioning mediated the effects of paternal distress on the children's distress.[104]

Psychiatric disturbance in chronically ill children has been shown to be associated with high levels of family stress (such as marriage stress, stress due to looking after other children at home, and work stress) and low levels of family support, including support from the marriage, from work and from friends.[74]

In summary, there is research evidence that parents of children with chronic illness suffer high levels of stress, distress and psychological symptoms in comparison with healthy populations. Mothers are more severely affected than fathers. Parent psychological distress varies with disease type, demonstrating the importance of studying individual chronic illnesses. In addition, distress is greater at the time of

diagnosis, with resolution over a period of one year. There remains uncertainty about the interactional effects of family functioning and social support on children's adjustment to illness. Taken together, these findings suggest the importance of longitudinal research into individual chronic diseases in children.

1.4.7 Qualitative Approaches to Studying the Effects of Chronic Illness in Childhood

There has been increasing interest in studying the subjective experiences of children, their parents and families using qualitative research methods. Researchers have used interviews and mixed quantitative and qualitative methods to explore the effects of chronic illness from the perspective of the children themselves, finding that issues of normality, existential issues and the importance of family and other social supports are common.[105-108]

Qualitative research into the experiences of parents of children with chronic illness has revealed the importance of adjustment, loss and social support in addition to positive experiences flowing from the illness experience.[109-112] In addition, for preschool aged children with chronic illness, parent concerns centred around the child's long-term prognosis, future growth and development, aggressive behaviour and attention problems.[102]

1.5 Methodological Issues in Studying the Psychosocial Effects of Chronic Illness in Childhood

1.5.1 Measurement of Child Psychosocial Functioning

Measuring psychosocial outcomes and health-related quality of life (HRQOL) in children is more complex than studying these factors in adults. Developmental changes over time result in the need for measures to change according to the child's developmental level. Assessment of a child's psychosocial adjustment needs to take

account of childhood-specific functioning, such as school functioning, peer relationships, physical development and body image.[113]

Very young children are not able to provide self-report of their psychological or functional state, resulting in the need for parents to report on these aspects.[113-115] Given that HRQOL has both objective elements (for example, functional limitations due to the illness) and subjective elements (for example, the emotional impact of the illness on the child),[116] HRQOL is likely to be evaluated differently depending on the informant. The child, parents and clinician may all vary in their report of an individual child's HRQOL.[117] There is greater agreement between parent and child reports of a child's HRQOL on objective measures (such as physical functioning) than subjective measures (such as emotional functioning).[118] In addition, it has been shown that parents and clinicians underestimate emotional symptoms in children following solid organ transplant.[119] Measuring HRQOL in young children is even more challenging due to their developmental immaturity that may result in difficulties in comprehension, short attention span for completing lengthy interviews or measures, and limited life experience that may mean they do not realise that alternative experience exists.[120] For very young children, it is therefore necessary to focus on objective aspects of functioning, most appropriately obtained through parent report.[121]

In assessing child adjustment and HRQOL, it is often difficult to distinguish between illness variables and child characteristics.[121] For example, it may be difficult to judge whether a child's social difficulties are related to the illness resulting in lengthy hospitalisation and missed opportunities for socialisation, or whether the social difficulties reflect the child's temperament. Given that the impact of a chronic illness on child adjustment is likely to change as the child develops over time, prospective longitudinal research is likely to be of more value than cross-sectional studies in assessing the effects of the chronic illness.[122]

The context of the assessment of psychosocial functioning is also important. For example, Calinescu and colleagues found that both children and their parents report better psychosocial outcomes than expected during a personal interview with a familiar nurse, suggesting that to whom the child and/or family is reporting their psychosocial outcomes may influence the assessment.[123] Collecting data during a clinic visit rather than in the family home may also have an impact on the measures because children and parents may have an emotional response that colours their self-report data in the medical setting.[124]

In summary, assessing psychosocial outcomes for very young children is complex and relies on parent report. Such proxy reporting limits the assessment to objective measures of functioning rather than being able to assess the child's subjective experience. The assessment difficulty is compounded by the child's development over time, the child's temperamental characteristics and the context of the assessment. Longitudinal research will be important in determining the psychosocial impact of serious chronic illness in infancy.

1.5.2 Measurement of Family Functioning

The concept of family functioning comprises a number of interrelated elements such as the relationships between different family members, how family members communicate feelings, how they solve problems, and how behaviour is managed within the family. In addition, family functioning changes over time.[125,126] The complexity of the concept results in difficulties in measurement. Since reliability of a measure is assessed by how closely the individual items are correlated, validity may be compromised by not including items that measure different constructs within the overall measure.[127] In addition, measures need to perform in the same way in different settings, such as between different groups, across time and between different roles in the family (mother, father or child for example).[127] Family functioning

research, therefore, needs to take account of the range of aspects of family function while preserving instrument reliability. Many family functioning measures have dealt with these issues by assessing a range of family functions in addition to a general measure of functioning. This approach can provide a broad measure for comparison across groups.[125]

The method of assessment is also important. Self-report measures and family interviews provide different viewpoints on family functioning.[125] Combining assessment types and using multiple measures is likely to provide a broader understanding of the impact of serious childhood illness on family functioning than using one method alone.

Due to the variability between health conditions, individual or family measures may have limited applicability across types of illness, even if they have been validated in chronic illness groups.[125] Similarly, family functioning may differ in families of children with chronic illness compared with normative groups. Such altered functioning, however, may be adaptive in response to illness factors rather than pathological or dysfunctional.[125]

Although there is wide acceptance of systems theoretical viewpoints in both clinical practice and research settings, fathers continue to be poorly represented in family research.[125,128] Each family member may be affected by a child's illness in different ways and each member may offer a different perspective on the illness and its effects.[125] It is therefore important to include fathers as well as mothers in childhood chronic illness research to obtain a more comprehensive picture of the child and family's experience.

Timing of the research study in relation to the phase of illness is important in assessing adjustment of both child and family to illness. For example, researchers have found high rates of diagnosable psychiatric disorders in children soon after a

diagnosis of diabetes[83] or cancer,[84,85] but with a sustained resolution of distress symptoms within one year following diagnosis.[84,85] Similarly, differences in the developmental stage of the family may affect the response of the family to a diagnosis of chronic illness in the child. For example, illness in a child may affect a new family differently from an established family.[125]

In summary, family functioning is complex due to the multiple levels of relationships and activities engaged in by families. The complexity can be addressed through instrument design that takes account of the different facets of family functioning and the ways in which a family may be affected by illness in a child. Using multiple measures can be helpful in addressing these problems. Including fathers in research is important in gaining a more comprehensive assessment of the impact of childhood illness on the family. Finally, timing of research in the phase of illness and family cycle has implications for the study findings and should be considered during the design phase of the study.

1.6 Psychosocial Aspects of Serious Liver Disease in Infants

Research examining the psychosocial outcomes of serious liver disease in childhood has focussed on older children, with relatively little attention being paid to pre-schoolers and infants. Research studies typically include children with a wide range of ages. Much research has relied on reports of mothers and has not included fathers' reports about the child's functioning, or has not reported separately on mothers' and fathers' reports. The psychosocial outcomes research base has addressed two major areas, health-related quality of life and emotional and behavioural outcomes.

Emotional and behavioural outcomes have been the focus of an increasing number of studies, with a wide range of measures being used. There has been increasing

interest in specific outcomes including psychiatric disorders such as anxiety, depression and post-traumatic stress disorder, for both the children themselves and for their parents.

However, much of the research conducted to date has been cross-sectional in design and has included children and adolescents of varied age and developmental stage. There is a gap in the knowledge base about the impact on the infant or the family of a diagnosis of serious liver disease in the developmentally important time of infancy.

1.6.1 Health-Related Quality of Life in Infants and Children with Serious Liver Disease

The research focus on child adjustment, coupled with technological advances in the treatment of life threatening conditions that greatly improved survival rates, led to an interest in the quality of life of surviving children. Quality of life has been seen as an important factor in determining appropriateness of treatment and making informed decisions about treatment and use of medical resources.[117]

Attempts to assess and measure health-related quality of life (HRQOL) in children with chronic illnesses began in the mid-1980s and continued thereafter.[129,130] While different HRQOL instruments measure different aspects of quality of life, the concept usually encompasses a range of areas of functioning such as physical functioning, emotional functioning and social functioning.[115,131]

Few studies have examined HRQOL in infants or children who have serious liver disease who have not received a transplant. Most studies to date have been conducted with children who have biliary atresia, though one was undertaken in older children who have non-alcoholic fatty liver disease, a complication of obesity and overweight.[132] An overview of the main findings is presented below.

An early study from Japan examined the HRQOL of 25 children and young people with biliary atresia who had had the Kasai procedure 14 to 24 years previously but had not had a liver transplant. The Japanese group were compared with a group of 21 similar children and young people from the UK, using the same HRQOL measure that was available in both Japanese and English (the Short Form 36, SF-36). The Japanese group did not significantly differ from the UK group except on the domain of 'vitality' (a measure of well-being).[133] There were, however, some cultural differences apparent in the study; the UK group did not differ from the normal population in the UK, while the Japanese group had significantly lower HRQOL in a number of domains compared with the normal population in Japan.

A cross-sectional study examined HRQOL in 221 children and young adults aged 2 to 25 years who had biliary atresia but had not received a liver transplant, comparing them with a group of 151 matched children and young adults who had biliary atresia and who had had a liver transplant and a group of matched healthy controls. They found significantly poorer HRQOL in both groups of biliary atresia patients compared with the healthy controls, with no significant differences between the patients with biliary atresia who hadn't had a transplant and those who had been transplanted.[134]

Finally, a cross-sectional study examined HRQOL in 219 children who had biliary atresia and who had survived without a liver transplant for at least 5 years. While the primary purpose of this study was to examine the medical status of the children, a HRQOL measure was included. Poor HRQOL was defined as a score more than 1 SD below the population mean. By this measure, 46.6% of the children had poor HRQOL.[135]

1.6.2 Health-Related Quality of Life in Infants and Children Following Liver Transplant

There has been much more extensive research into HRQOL in children post-liver transplant, though a detailed description of the findings is beyond the scope of this thesis. A summary of the main findings from recent large studies, systematic reviews and a meta-analysis are presented here.

A multi-centre study compared HRQOL in 873 children who had had a liver transplant at least one year previously with a healthy control group and with a group of children actively receiving treatment for cancer. The liver transplant group reported significantly lower quality of life than healthy controls but similar levels of psychosocial and emotional health compared to the cancer group. Although the liver transplant children reported better physical quality of life than the children with cancer, this was to be expected since the liver transplant group were in the recovery phase of their illness while the cancer group were in active treatment.[136] In a later study, the same group of children was compared to children with other chronic illnesses (including Juvenile Rheumatoid Arthritis [JRA], type I diabetes, cancer in remission, cardiac disease, end-stage renal failure and inflammatory bowel disease) who had been assessed using the same HRQOL measure. The researchers reported HRQOL that was comparable to children who had had a renal transplant and children who were in remission from cancer. The children with a liver transplant had better functioning on most sub-scales compared with children who were on renal dialysis, and reported better physical health than children with JRA. School functioning was worse in children with liver transplant compared to that of children with JRA and diabetes, but similar to the other chronic illnesses included in this study. The findings indicate that while there are similarities across illness groups, HRQOL varies according to individual patient and disease characteristics.[137]

Research into the longer term psychosocial functioning of children who have received a solid organ transplant has attempted to assess changes in HRQOL over time and identify risk factors for poor outcomes. The research revealed that HRQOL improves after organ transplantation but remains significantly lower than in healthy controls, even after many years post-transplant. However, data assessing a group of 24 young adults who had had a liver transplant 20 years previously found HRQOL that was comparable with the healthy population.[138] Better HRQOL is associated with longer duration since time of transplant and younger age at transplant. Disease variables such as liver diagnosis, length of admission to hospital and physical complications of the illness, as well as demographic variables and family functioning have also been demonstrated to be associated with HRQOL outcomes.[139-142]

Pinquart and Teubert's meta-analysis of 954 studies of children with chronic illnesses included 45 studies examining kidney and liver disease as part of the analysis, specifically looking at physical and social functioning, as well as academic functioning, as detailed above. They found moderate effect sizes (ES) for these children in relation to physical functioning (*ES* of -0.72) and social functioning (*ES* of -0.59), indicating significantly reduced HRQOL in children with these chronic disorders.[31]

In summary, the HRQOL research base indicates that liver transplantation has the capacity to improve HRQOL. in individual children and that Younger age at transplantation is associated with better HRQOL outcomes than transplantation in older children. However, HRQOL remains poorer in transplanted children than in healthy populations, indicating levels similar to those found in children with other chronic illnesses. Some children are at greater risk of poor HRQOL outcomes, particularly those with more physical complications and family conflict, a situation that may be related to poor treatment adherence.

1.6.3 Emotional and Behavioural Outcomes in Infants and Children with Serious Liver Disease

There has been limited research into emotional and behavioural outcomes for infants and children with serious liver disease who have not received a transplant. Health related quality of life research, described above, includes psychosocial functioning and is not repeated in the discussion that follows.

Bradford undertook a descriptive study of 45 children with biliary atresia who had not had a transplant, using the Behaviour Checklist for the children aged under 5 years old (N = 24) and the Rutter A Scale (a measure of emotional and behavioural disorders in children and adolescents) for children aged over 5 years old. In total, 44% of the children scored above the clinical cut-off, indicating clinically significant problems with adjustment. The children's adjustment was not predicted by the severity of the children's illness, but was significantly predicted by the mothers' perceptions of their child's health.[143]

Hopkins and colleagues studied the temperamental characteristics of 14 infants with biliary atresia, aged 4 to 30 months, using the Infant Characteristics Questionnaire (ICQ). The infants were significantly less socially responsive when compared with normative data, and there was a significant correlation between the infants' scores on the ICQ and disease severity, with worse scores correlating with increasing disease severity.[17]

Mastroyannopoulou and colleagues undertook a descriptive study comparing 15 children with chronic liver disease, to a group of 10 children who had had a liver transplant and 15 healthy controls. They examined coping style, health locus of control and self-perception of health in addition to the children's understanding of illness. The children who had liver disease but had not had a transplant were significantly more likely to show an external locus of control. That is, they perceived

having less personal control over their health when compared with the liver transplant group and the healthy controls. The children who had not had a transplant were significantly less anxious about their health than the children who had had a transplant, but were less likely than the transplanted children to rate themselves as healthy.[144]

Ingerski and colleagues undertook a pilot study in a group of children aged 2 to 17 years with a number of chronic illnesses. The group included 23 children who were transplant candidates for solid organ or bone marrow transplants (the number of children who had liver disease amongst this group is not reported) in addition to 28 children with sickle cell disease and 13 children with HIV infection. They assessed post-traumatic stress symptoms in the children and their caregivers (predominantly mothers) and found significantly higher rates of post-traumatic stress symptoms in the caregivers and the children who were transplant candidates or had sickle cell disease compared with community norms.[145]

In summary, the evidence base regarding the emotional and behavioural outcomes of liver disease in infancy and childhood is extremely limited. The majority of studies that have been undertaken are small and often descriptive in nature, resulting in the need for caution in interpreting the results. However, the research suggests that children with chronic liver disease have problems with adjustment, perceive themselves as lacking control over their health, and suffer from post-traumatic stress reactions.

1.6.4 Emotional and Behavioural Outcomes in Infants and Children Following Liver Transplant

As in other areas of research in childhood chronic liver disease, there has been much more research conducted with children who have had a liver transplant. There have been several meta-analytic studies by Pinquart and colleagues that analyse the

emotional and behavioural outcomes of childhood chronic illnesses, including data from studies of paediatric liver transplantation.

1.6.4.1 Internalising and externalising symptoms

Pinquart and Shen conducted a meta-analysis of emotional and behavioural problems as measured by the Child Behavior Checklist (CBCL) in children with chronic illness. The meta-analysis included 569 studies (a total of 51,422 children) published up to May 2011. The analysis included 22 studies (a total of 937 children) that specifically looked at kidney and liver disease, six of which included children who had had a liver transplant (279 children). Unfortunately, the meta-analysis did not separate the data for liver transplant in children from the data related to kidney disease. Overall, there was a small ES for emotional and behavioural problems in chronically ill children across all studies combined, but children with kidney or liver disease had a moderate ES of 0.70 (95% *CI* 0.52 to 0.88) for CBCL Total Problems, a moderate ES of 0.67 (95% *CI* 0.50 to 0.85) for Internalizing Problems, and a small ES of 0.34 (95% *CI* 0.16 to 0.51) for Externalizing Problems. When the CBCL Total Problems and Internalizing Problems scales were modified to exclude somatic symptoms, the ES for Total Problems remained moderate (0.78, 95% *CI* 0.59 to 0.96)), as did Internalizing Problems (ES 0.59, 95% *CI 0.41 to 0.77*).[89] However, the meta-analysis also noted significant heterogeneity of effect size in all CBCL problem areas amongst kidney and liver disease studies. Examining the individual liver studies included in the meta-analysis revealed a range of findings as suggested by the heterogeneity reported in the meta-analysis. Despite the limitations of the meta-analysis in relation to separating liver disease from renal disease, the findings show significant emotional problems for children who have had a liver transplant. Liver transplant and chronic kidney disease are associated with greater emotional problems than many other chronic illnesses.

A study that was not included in the meta-analysis reported CBCL data on a subgroup of 54 patients from a larger sample. Although mean scores are not reported, there was a significant negative correlation between age at transplant and behavioural problems, particularly aggression. Children who had been transplanted at a younger age demonstrated more behavioural problems than those transplanted when older. The study included separate ratings for mothers and fathers. There was broad agreement between mothers and fathers on the difficulties being faced by the children.[146]

1.6.4.2 Depression and anxiety

Pinquart and Shen conducted a meta-analysis of 340 studies (including a total of 33,047 children), published up to September 2010, of depressive symptoms in children with chronic illness. While the meta-analysis included data from four studies on liver transplant (total of 105 individual patients), it also included analysis of this subgroup in an 'other illnesses' section with 79 other samples so it is not possible to ascertain the specific findings in relation to liver transplantation. Despite this, the meta-analysis found a small to very small effect size (ES 0.19, 95% *CI* 0.15 to 0.23) for depression in chronic illness.[87]

Examining the liver transplant studies included in the meta-analysis shows mixed findings. Windsorova and colleagues studied children aged 4 to 12 years, comparing 25 children who had had a liver transplant at least one year previously with a control group of 26 children with diabetes. They found no difference between the groups in anxiety or depression, and no difference in CBCL scores compared with normative data. The researchers did note lower depression scores compared with normative data on the Child Depression Inventory, and less anxiety compared with normative data measured by the State-Trait Anxiety Inventory for Children.[147]

Another study included in the meta-analysis, by Fredericks and colleagues, reported on 38 children aged 2 to 16 years who had had a liver transplant within the preceding 5 years. Using the CBCL, the investigators found that although mean scores were not elevated into the clinical range, Internalizing Problems scores were significantly higher in the study group compared with test norms.[148]

Another liver transplant study included in the meta-analysis investigated children aged 7 to 18 years, including 42 children who had had a liver transplant and 28 children who had had a renal transplant at least 6 months previously. The study investigated the associations between hope and uncertainty and anxiety, depression and treatment adherence. The parent reports were mostly completed by mothers, but included data from eight fathers. However, mothers' and fathers' responses are not reported separately. The investigators found a significant negative association between hope and anxiety and depression (less hope was predictive of higher anxiety and depression scores) and a positive association between uncertainty and anxiety and depression (more uncertainty was predictive of higher anxiety and depression scores). There was also a negative association between depression and treatment adherence (higher depression scores were predictive of less adherence).[149]

The final liver transplant study included in the meta-analysis examined the relationship between depression, anxiety and medication adherence in a group of children aged 7 to 18 years, including 32 children who had had a liver transplant and 23 children who had had a renal transplant at least six months previously. Interestingly, the researchers found psychosocial functioning, depression and anxiety scores comparable with normative data. Contrary to expectation they found that greater anxiety in the children was associated with better medication adherence.[150]

A third meta-analysis by Pinquart and Shen examined 332 studies (a total of 29,124 children), published up to November 2010 of anxiety in children with chronic illness. The meta-analysis included 19 studies (total number not reported) of children who had had a liver or renal transplant. Across all illnesses, there was a very small effect size for anxiety (ES 0.18, 95% *CI* 0.14 to 0.22), with liver and kidney disease also showing a very small effect size (ES 0.14, 95% *CI* -0.06 to 0.35).[88] Taken together with the preceding meta-analysis, it appears that findings of higher rates of internalising problems, measured by the CBCL, are likely accounted for by increases in rates of depression rather than anxiety in children with chronic illness and in those with chronic liver disease.

1.6.4.3 Post-traumatic stress disorder

Several researchers have focussed on post-traumatic stress disorder as a specific complication of paediatric solid organ transplantation. Each study has been cross-sectional in design, but used well-validated parent- and/or self-report instruments or clinician-rated assessments. PTSD is reported at rates of 11% to 16.3% in children who have had a transplant,[119,151,152] comparable with findings from PTSD research in children who have had an accident or have been diagnosed with another chronic illness such as cancer or diabetes.[153] Although two studies have differed in their findings directly comparing children who have had a liver transplant with children with other chronic illnesses,[119,152] the contradictory findings appear to be due to differences in the illnesses in the comparison groups. Walker and colleagues compared children who had had a transplant with children who had much less serious illnesses (asthma or a routine ear, nose and throat procedure) and reported significantly higher rates of PTSD in the transplant group.[152] Shemesh and colleagues' study included a comparison group of children with a range of chronic illnesses, the majority of which were associated with significant morbidity, such as

diabetes mellitus, chronic heart disease, joint disease and severe food allergy. The researchers found no significant difference between illness groups in this study.[119] Perception of threat, rather than objective disease severity, is important in the development of PTSD symptoms.[151] It is possible that the differences in morbidity between illnesses may have contributed to the different findings in these studies.

1.6.4.4 Risks associated with psychological distress

Much research relating to liver transplantation has been driven by the recognition that psychological distress is associated with poor treatment adherence and the risk of morbidity or mortality. A large number of studies and two meta-analyses have identified clear links between psychological symptoms, including depression, anxiety and PTSD, and poor adherence.

PTSD has been shown to be present at significantly higher rates in non-adherent compared with adherent children and adolescents,[154] and has also been found to be significantly correlated with non-adherence.[155,156] Similarly, a history of child abuse, frequently associated with the risk of PTSD, has also been shown to be a risk factor for non-adherence in children and adolescents.[157,158]

Psychiatric disorders in children and adolescents, including depression and anxiety, have also been identified as risk factors for poor treatment adherence following liver transplantation.[149,156,157,159-161] Interestingly, one study found anxiety to be predictive of better treatment adherence. Wu and colleagues prospectively investigated a group of children and adolescents post-transplant using an electronic medication adherence monitor (a micro-chipped medication bottle cap that records time and date each time the bottle is opened, hence inferring the time that medication is taken), arguably a more accurate source of information about medication adherence than self-report. They found that greater anxiety was associated with better medication adherence that was sustained over time.[150] However, it seems likely that

adherence is related to the interplay between a number of complex factors, including socio-demographic factors, child age, psychiatric disorders and family functioning.[148,157,160-162]

1.6.4.5 Body image and self-esteem

Pinquart conducted meta-analyses of body image[91] and self-esteem[92] in children with chronic illness, both of which found effect sizes for liver and kidney disease in the very small, non-significant range. The findings are somewhat surprising, given the physical morbidity that accompanies liver disease including abdominal distension from organomegaly and ascites, growth impairment, and Cushingoid appearance in some children due to steroid use. In addition, children with splenomegaly are often restricted from participating in contact sports due to the possibility of splenic rupture, potentially limiting opportunities for developing self-esteem.

1.6.4.6 Adaptive functioning

Since publication of the meta-analyses, there have been a number of research studies published in the area of HRQOL of paediatric liver transplant recipients, as summarised above. There has been only one further research study published investigating the emotional and behavioural outcomes of children with chronic liver disease. Shellmer and colleagues studied adaptive functioning in children aged 2 to 18 years, comparing 18 children who had had an intestinal transplant with 22 children who had had a liver transplant. Adaptive functioning refers to daily living skills such as communication, socialising, self-care, and everyday activities. The investigators found that the liver transplant group were not significantly impaired when compared with general population norms. The liver transplant group performed significantly better than the intestinal transplant group.[163]

1.6.4.7 Qualitative research

Several qualitative studies have been undertaken seeking children's and adolescents' perspectives on having had an organ transplant. Common themes include issues around the challenges of having a life-threatening illness, medication, feeling normal, peer relationships, the importance of family support and distress around physical limitations.[107,164-168]

1.6.4.8 Summary: Emotional and behavioural outcomes in infants and children following liver transplant

In summary, liver transplantation in childhood is associated with a heightened risk of internalising problems, which appears to be due to an increased risk of depressive symptoms rather than anxiety symptoms. Liver transplantation also appears to be associated with an increased risk of PTSD, at comparable rates to those found following road trauma or paediatric cancer. Emotional distress and psychiatric illness is associated with problems with treatment adherence in children who have had a liver transplant. Liver disease and transplantation does not appear to be related to problems with self-esteem or body image despite the visibility of the illness due to side effects of treatment or illness morbidity. Daily living skills in children who have had a liver transplant also appear to be unimpaired in comparison with healthy peers. Despite the lack of significant findings in relation to problems with self-esteem or body image, qualitative research of children and adolescents' experience of liver transplantation has revealed themes of normality, distress and the importance of peer relationships.

1.7 Parent Distress and Serious Liver Disease in Infants

Most research has concentrated on parent distress after liver transplantation has occurred rather than on parent distress when a child has liver disease. The research has also focussed on children of varying ages rather than focussing on specific age

groups. There is a gap in knowledge about parent distress when an infant has serious liver disease. The impact of living-related liver transplantation, in which a parent donates a portion of their liver to the child, is a specific situation that is beyond the scope of this thesis.

1.7.1 Distress in Parents

Four cross-sectional studies of children and adolescents who received a liver transplant up to 11 years previously included measures of parent emotional distress. The studies found ratings of emotional distress in the parents significantly higher than population norms.[142,148,169,170]

In contrast, a longitudinal study[171] in which mothers' psychological distress was measured both before and following their child's liver transplant found psychological distress scores within test norms at both time points.[171]

Gritti and colleagues examined the parents of children who had had a liver transplant compared with children who had chronic liver disease. They found no difference in the measures of parent distress between the two groups. The findings were not compared with normative data, limiting the generalizability of the results.[172]

Simons and colleagues studied 34 mothers and 22 fathers of children who were being evaluated for solid organ or bone marrow transplant, including 10 liver transplant candidates. The mothers, but not fathers, had significantly higher distress scores when compared with normative data.[173] However, studying a group of parents whose children were candidates for different types of transplantation may have affected the results of this study because the mothers of children who were being evaluated for bone marrow transplant had significantly higher distress scores than the mothers of children awaiting a solid organ (including liver) transplant.

As in other chronic illness research, PTSD is a risk for parents of children who have had a liver transplant. A cross-sectional study of 170 caregivers of children who had had a solid organ transplant (liver, kidney or heart) at least 10 months previously and found that 27.1% of the parents met DSM-IV criteria for PTSD.[174] The findings were reported to be comparable to reported rates in parents of children with cancer and higher than the expected community rate of 8%. In addition, 27.1% of the parents had at least mild depression measured by the Beck Depression Inventory, half of those scoring in the moderate to severe range, however the mean scores were within the normative range. Transplant variables (type of transplant and number of transplants), child health, negative attitudes towards health care, greater family and social impact of the child's illness, and less perceived benefits of the transplant were all significant predictors of the severity of parent PTSD.[174]

In summary, there is conflicting data about levels of parent distress in paediatric liver disease and following liver transplantation. Further investigation is required to assess whether parent psychological symptoms are increased in the context of liver disease, as occurs in other chronic illness.

1.7.2 Changes in Parent Distress over Time

Despite the lack of clarity regarding levels of distress in parents post-transplant, there is some evidence of changes in levels of parent distress over time. One prospective study found significant improvement in parent emotional impact 6 months after liver transplantation in children under the age of 5 years, compared with the pre-transplant period. The improvement was sustained for 12 months following transplantation. The authors did not compare their results with normative data.[175] LoBiondo-Wood and colleagues undertook a longitudinal study of 15 mothers of children from before liver transplant to 5 years post-transplant. The mothers' anxiety and depression were within test norms both pre- and post-transplant. The authors also reported significant

improvement in maternal anxiety and stress levels from the time of outpatient assessment for transplant to the period following discharge from hospital after the child's liver transplant.[171]

Another cross-sectional study examined parents whose children were awaiting liver and/or intestinal transplant, compared with parents of children who were 2 months post transplant. The researchers found no differences in parent distress between the pre- and post-transplant groups, but it is not possible to judge whether parent psychological symptoms were elevated because the findings were not compared with normative data.[176] Further, the periods of waiting for a transplant and 2 months post-transplant may be highly stressful for parents. The study results may therefore obscure differences that may occur between the pre- and post-transplant periods over a longer period.

However, another prospective study found ongoing high levels of distress in parents of children with transplants as compared to normative data. Devine and colleagues studied adolescents who had received a solid organ transplant (liver, heart or kidney) and their parents for 18 months following transplant and found significantly greater emotional distress in parents compared with normative data throughout the study period.[177]

1.7.3 Parent Distress and Child Outcomes

Greater parent distress is associated with poor treatment adherence in children who have had a solid organ transplant. A meta-analysis of 61 studies (total number of participants = 3834) of paediatric liver, kidney and heart transplant published prior to June 2008 found a very small but significant effect size of parent distress on child treatment adherence ($r = .13$, 95% $CI = .01$ to $.25$, $P < .05$).[162] Poor treatment adherence is associated with organ rejection. Stone and colleagues studied parents identified as 'at risk' on a range of psychosocial factors during the child's pre-

transplant assessment for heart transplant. They reported a relative risk of 3.14 (95% *CI* = 1.52 to 5.60, *P* < .001) for increased hospitalisations when the children were more than 6 months post-transplant. They also found a risk of organ rejection in this group, however the risk approached but did not achieve significance (RR = 3.40, 95% *CI* = 0.95 to 12.14, *P* = .06).[178]

Better emotional functioning in mothers has been associated with better psychomotor development in young children following liver transplant.[179]

1.7.4 Age at Transplant and Parent Distress

There is conflicting evidence about whether younger age at transplant is associated with differences in levels of parent distress. Alonso and colleagues undertook a cross-sectional study 20 to 28 months following liver transplant. The researchers found distress to be significantly higher in parents of children who were older than 5 years at the time of liver transplant compared with normative data. The parents of children who had had a transplant before the age of 5 years were compared with parents of children from a paediatric clinic who had an acute illness. There was no significant difference in distress in the parents of the younger children compared with the control group.[180] The differing results are likely to be due to the selection of comparison groups. The study was limited by the use of different measures of HRQOL for the two age groups, and normative data was not available for the measure used with the younger children. Hence, the researchers chose a comparison group of children without chronic illness (but who had an acute illness) for the younger children, while comparing the parents of the older children with normative data. Unfortunately the choice of comparison groups may have obscured differences between the younger children and the acutely ill comparison group. The same researchers had undertaken an earlier pilot study and had found significantly greater emotional distress in the parents of children transplanted at least 2 years

previously, including children aged under 5 years at transplant, in comparison with normative data.[170]

In contrast, Tarbell and Kosmach found that younger age of the child at liver and/or intestinal transplant was associated with greater parent distress.[176] A strength of the study is in its standardisation of timing of the research, suggesting that the finding of differences in the clinical groups may be robust. However, the sample size was small and included children with intestinal transplantation as well as children with liver transplantation, so the findings may not be generalizable.

In summary, there is continuing uncertainty about the relationship between the child's age at transplant and parent distress. Further investigation is required to determine the if there is an association between age at transplantation and parent distress.

1.7.5 Differential Effects of Child Liver Disease on Mothers and Fathers

The majority of research examining parent distress in paediatric liver disease has reported combined data from mothers, fathers and other caregivers. However, Posfay-Barbe and colleagues reported on mothers and fathers separately and found no significant differences in levels of distress between parents. Unfortunately the results were limited by a small sample size and high attrition rates for fathers.[179] Tarbell and Kosmach found significantly greater distress in fathers compared with mothers.[176] Rodrigue and colleagues studied 18 fathers of children who were being evaluated for a transplant (5 of whom had liver disease) and found the fathers to score significantly lower on a parenting stress scale (the Parenting Stress Index) compared with the normative data.[181] The same group of researchers separately investigated mothers of children being evaluated for transplantation. The mothers of 36 children (9 with liver disease) did not differ significantly from normative data in terms of parenting stress.[182] The researchers followed up the mothers after the children's transplant and found significantly higher levels of parenting stress in the

mothers post-transplant compared with pre-transplant stress, which was sustained for 6 months after the transplant.[183] Simons and colleagues also studied mothers and fathers separately in their group of 34 mothers and 22 fathers of children who were being evaluated for solid organ or bone marrow transplant, noted above. The mothers, but not fathers, were significantly more distressed when compared with normative data. The mothers' distress was mitigated by social support, while social support did not affect fathers' distress. The authors also found evidence that fathers and mothers used different coping strategies.[173]

1.7.6 Summary: Parent Distress and Serious Liver Disease in Infants

In summary, there is conflicting evidence about whether distress scores are elevated in parents of children who have liver disease or who have had a liver transplant, in comparison with normative data. However, distress post-transplantation has been found to continue long-term despite some improvement over time. Parent distress is associated with poor outcomes for the children through the mechanism of poor treatment adherence and the consequent increased risk of organ rejection. Uncertainty remains regarding the effects of the child's age at transplant on parent distress. There is also conflicting evidence regarding the differential effects of paediatric liver transplantation on mothers and fathers.

1.8 Family Functioning and Serious Liver Disease in Infants

Most research into family functioning in childhood liver disease has focussed on the post-transplant period. There is a gap in knowledge about family functioning when an infant has serious liver disease. One longitudinal study examined family functioning both pre- and post-transplantation and is discussed below.

1.8.1 Family Functioning

Three cross-sectional studies have shown family functioning to be within the healthy range following paediatric liver transplantation.[148,180,184] In addition, a longitudinal study of mothers of children being evaluated for liver transplantation, followed up five years later, found family functioning to be within the healthy range both pre- and post-transplant.[171]

However, other research has found significant disruption to family activities following paediatric liver transplantation in comparison to normative data,[142,148,169,170,177] though contradictory findings were reported in one study that found no impact on family activities or family cohesion compared with another chronic illness group or normative data.[185] The latter study was undertaken in South America, so it is possible that cultural differences may account for the variation in findings.

Pinquart conducted a meta-analysis of 325 studies (total number of participants = 31,288), published prior to February 2013, examining the parent-child relationship and parenting behaviour and styles of families with a child with chronic illness. The meta-analysis included 5 studies of children with liver or kidney disease (the two groups' data is reported together). The author found small effect sizes for differences in parenting behaviours, with more overprotection in the chronic illness group compared with families of healthy children or test norms (g = 0.39, 95% CI = 0.29 to 0.50). However, there was significant heterogeneity between studies and there were no significant differences in parenting behaviours in the liver or kidney studies in comparison with the healthy groups.[186]

Taken together, the findings suggest changes in aspects of family functioning, such as disruption of activities and parenting behaviours, following liver transplantation in children. There is limited research regarding the relationship between pre-transplantation paediatric liver disease and family functioning.

1.8.2 Mothers' and Fathers' Perceptions of Family Functioning

There is limited research that examines mothers' and fathers' perceptions of family functioning separately. Rodrigue and colleagues separately assessed mothers and fathers of children being evaluated for organ transplant including liver transplant. The researchers found that fathers[181] and mothers[182] each reported significantly less family conflict compared with normative data, while both parents also reported higher impact of the illness on the family compared with families who have children with other chronic illness.[181,182] The mothers were followed up and reported significantly greater impact on the family following transplantation compared with the pre-transplant period.[183]

1.8.3 Parent Distress and Family Functioning

Research has shown an association between parent psychological distress and family functioning in paediatric liver disease.

LoBiondo-Wood and colleagues examined a group of 29 mothers of children who were being evaluated for liver transplant. The researchers found significant correlations between family stress and family functioning ($r = .58$, $P < .01$) and maternal distress and family functioning ($r = .37$, $P < .05$).[187] Family conflict is also associated with greater psychological distress in parents of children who have had a liver and/or intestinal transplant.[176]

1.8.4 Family Functioning and Child Emotional Outcomes Following Liver Transplant

There are concerns about the effects of disrupted family functioning on the emotional outcomes for children who have had a liver transplant. An Australian study reported an association between greater disruption to family routines and poor HRQOL in the children who had had a transplant.[188] Parent report of perceived higher impact of the child's liver transplant on the family correlates with greater psychosocial difficulties in

the child.[189] Family conflict is associated with worse mental health outcomes in adolescents who have received a liver transplant[190] or solid organ transplant.[191,192] Poor family functioning has been identified as a risk factor for the development of PTSD in children who have experienced a traumatic event.[64] In a study of paediatric heart transplantation, family functioning was significantly correlated with child emotional functioning both before and after transplant,[193] a situation that was demonstrated to be sustained over time.[194]

Dew and colleagues conducted a meta-analysis of 61 studies (total number of participants = 3834), published prior to June 2008, examining treatment adherence in children who had had liver, kidney or heart transplant. Poor family cohesion was a significant predictor of poor child adherence, though the effect size was very small (r = .15, 95% CI = .02 to .28, P < .05).[162]

Healthy family functioning has been shown to be a significant predictor of social competence in children who have liver disease, although the effect size is small. Hoffmann and colleagues studied 30 children with chronic liver disease and found better family functioning to be predictive of better child social competence, contributing 23% to the variation in child CBCL Social Competence scores.[195]

While research assessing the impact of chronic paediatric liver disease on other family members is beyond the scope of this thesis, it should be noted that paediatric liver disease also has an emotional impact on siblings. Siblings of children awaiting liver transplant have been found to have mean scores on the social and behaviour scales of the Child Behavior Checklist within the normal range. However, large percentages of siblings score within the clinical range when compared with normative data.[196]

1.8.5 Family Demographics and Child Emotional and Behavioural Outcomes

There is conflicting evidence regarding the effects of family socio-economic status (SES) on psychosocial outcomes for children who have had a liver or other solid organ transplant. For example, parent education level has been associated with child psychosocial outcome after liver transplant.[142,163] However, other researchers have not found an effect of SES on psychosocial outcomes for children who have had a renal or liver transplant[197] or following heart transplantation.[193,194] Research examining family adaptation to a child's liver transplant found no effect of SES.[198]

Research in the general population and chronic illness groups suggests that SES would be expected to have an impact on psychosocial outcomes for children with liver disease and their families. Lower SES in the general population has been shown to be associated with the development of child emotional problems[97,199] and research in children with chronic illness has demonstrated an association between SES and problems in family functioning.[200] Maternal age has also been associated with psychosocial outcome for children with chronic illness, younger maternal age at childbirth predicting worse psychosocial outcome for the children.[201]

In summary, there is conflicting evidence about the effects of family demographic characteristics and the emotional outcomes of children who have liver disease. However, there is evidence from population research and from chronic illness research that suggests an interaction between family demographics, family functioning and child emotional functioning.

1.8.6 Summary: Family Functioning and Serious Liver Disease in Infants

There is a gap in knowledge about family functioning when an infant has been diagnosed with serious liver disease. Family functioning has been studied in children following liver transplantation and demonstrated to be in the healthy range compared with normative data. Despite this, there is evidence of disruption to family activities

and a correlation between measures of parent psychological distress and altered family functioning. There is conflicting evidence about differential perceptions between mothers and fathers in relation to family functioning and the impact of paediatric liver disease on the family. There is also continued uncertainty about the effects of family demographic variables such as socio-economic status and parent age on the emotional and behavioural outcomes of children with serious liver disease. Disrupted family functioning is associated with poor emotional outcomes and poor treatment adherence in the children. However, healthy family functioning is associated with improved social competence in children who have chronic liver disease.

1.9 Father Engagement and Serious Liver Disease in Infancy

Father engagement in paediatric liver disease or following liver transplantation has not been previously examined.

There is evidence that employment responsibilities, inconvenient health provider availability, and low confidence in parenting skills are barriers in fathers attending health-related appointments with their children.[202] Older fathers and younger child age are associated with greater father engagement in children's health care.[203] However, greater father engagement is associated with better HRQOL and treatment adherence in adolescents with chronic illness.[204] In addition, in families who have children with chronic illness greater father engagement is associated with fewer psychiatric symptoms in mothers and less perceived impact of the illness on the family.[205]

1.10 Limitations of Previous Research into Serious Liver Disease in Infancy

Although early onset of illness and younger age at transplantation appear to be associated with poorer emotional and behavioural outcomes in the children, research has typically focussed on children and families following liver transplantation. There has been no research to date examining the period immediately following diagnosis of serious liver disease in infancy.

Studies into serious liver disease in childhood have included children of a wide range of ages, limiting the ability to control for developmental stage of the child.

The majority of studies to date have been cross-sectional in nature. There are few prospective longitudinal studies, limiting the capacity to identify risk or protective factors for child emotional outcomes.

Much of the research undertaken to date has not provided separate data for mothers and fathers. There is continuing uncertainty about differences in emotional responses and perceptions of family functioning between parents. No prior research to date has examined the role of father engagement in the care of children with chronic liver disease or following liver transplantation.

There is evidence of persistent high levels of distress in parents following liver transplantation. However, to date there are no studies that examine distress in parents following the diagnosis of serious liver disease in infancy.

Family functioning appears to be within the healthy range following liver transplantation. However, there is evidence of disruption to family activities and a correlation between parent psychological distress and altered family functioning. There is continued uncertainty about mothers' and fathers' differential perceptions of family functioning and the impact of paediatric liver disease on the family.

The interactional effects of parent distress, family functioning and father engagement have not been studied in the context of serious liver disease presenting in infancy.

1.11 Aims of the Current Study

The principal aim of the study is to gain a deeper understanding of parent and family adjustment to the diagnosis of serious liver disease presenting in the developmentally sensitive time of infancy.

In addition, the study aims to examine the interaction between parent emotional reactions, family functioning and father engagement over time to identify predictive factors for the infants' emotional and behavioural outcomes.

Finally, the study aims to inform clinical best practice in the provision of mental health care for the families and their infants by identifying parents and infants who may benefit from early intervention.

1.12 Research Hypotheses

There are three research hypotheses.

1. Parents of infants with serious liver disease will have high levels of distress, demonstrated by the presence of psychological symptoms and alterations in family functioning
2. Fathers' perceived engagement in the infants' care will have an impact on parent distress and family functioning
3. Parent distress, family functioning and fathers' engagement will have predictive value for the emotional and behavioural outcomes of the infants

1.13 Conclusion

Serious liver disease presenting in infancy is associated with clinically significant morbidity, both physical and emotional. Improved patient survival rates have resulted in an increasing focus on the health-related quality of life and emotional outcomes for these children. Research into paediatric liver disease and transplantation reveals high rates of internalising symptoms that are, in turn, associated with poor treatment adherence and poor medical outcomes. However, research to date has used differing methodologies, leading to difficulties in interpreting and comparing studies with a variety of findings and has concentrated on the post-transplant experience. Studies have also typically included participants from a wide range of ages, limiting the ability to assess the impact of developmental stage on illness outcomes. Most studies have used a cross-sectional design, limiting the capacity to assess changes over time or to determine risk factors for later psychopathology. There is therefore a need to prospectively study serious liver disease from early in the illness course.

There is evidence of high levels of distress in parents of children who have had a liver transplant in comparison with normative data, which continues post-transplant. Although family functioning appears to be in the healthy range, there is evidence for disruption to family activities following transplantation and an association between parent distress and altered family functioning. Parent distress and problems in family functioning are associated with poor treatment adherence and increased risk of organ rejection. Uncertainty remains regarding the effects of the child's age at transplant on parent distress and family functioning, and whether there are differential effects on mothers and fathers of serious liver disease in infancy. Father engagement has not been studied in this context.

The interactions between parent and family factors and their predictive capacity for child outcomes have not been studied in children with serious liver disease.

2 Methods

2.1 Synopsis

This chapter explains the study methodology. The chapter begins with a description of the study design and justification for the choice of methodology used. The study has a mixed-methods design that utilises both quantitative and qualitative data collection and analysis. The methodology has the benefit of elucidating different aspects of the illness experience that cannot be obtained with either method alone. The study design therefore provides the opportunity for an enhanced understanding of the experience of parents who have an infant with serious liver disease.

Following a description of the setting and participants, the study instruments are presented. Next, the techniques used for data analysis are described, detailing the analytic approach to testing each of the study's hypotheses. The approach to analysing the qualitative data component and the final integration of quantitative and qualitative data are then presented. Validity and reliability of the qualitative data analysis were ensured through use of a journal, recording of field notes, use of an independent coder to assess the appropriateness of the coding, and triangulation of the quantitative and qualitative data.

2.2 Study Design

The current study is an exploratory one-year prospective cohort study. The study uses mixed methods with concurrent quantitative and qualitative data collection and analysis. In this approach data is collected in two relatively independent strands, one quantitative and the other qualitative. Each is analysed separately with synthesis of the results of each strand to form meta-inferences at the end of the study analysis.[206]

Qualitative research is increasingly being undertaken in families of children with chronic illness,[108,207,208] in liver and other solid organ transplantation [167,209-213] as well as with children and teenagers themselves after solid organ transplant[107,166,209] and liver transplant.[164,168]

Mixed methods research is especially useful when studying complex phenomena, such as patient experience of illness, because each method examines different aspects of the experience with the aim of broadening and enhancing overall understanding.[214] Mixed methods research has been used in paediatric acute illness,[215] paediatric chronic illness[216,217] and paediatric organ transplantation,[218,219] particularly in relation to medication adherence, coping and psychosocial functioning.

A mixed methods research design was chosen for this study for several reasons. Due to the rarity of liver disease in children, it was likely that the study sample size would be small. Infants were chosen so that the developmental stage of participants was standardised, though it was recognised that this would further limit the sample size. Obtaining parents' direct reports of their experiences via qualitative interview therefore resulted in a more complete understanding of the experiences of the parents than could have been obtained with the quantitative data alone.

2.3 Setting

There are three paediatric liver transplant units in Australia, located in Sydney (Children's Hospital at Westmead [CHW], the National Paediatric Liver Transplant Unit), Brisbane (Royal Children's Hospital Brisbane [RCHB]), and Melbourne (Royal Children's Hospital Melbourne [RCHM]). The study includes consecutive cases from all three centres and from the additional children's hospital in Sydney (Sydney Children's Hospital [SCH], located at Randwick) that does not offer transplants, but

whose Department of Gastroenterology treats infants and children who have serious liver disease.

2.4 Participants

Participants were enrolled between May 2009 and May 2013. Enrolment commenced at the two Sydney sites. Due to low numbers of infants presenting across the two sites, the author sought participation from the other two Australian Paediatric Liver Transplant units. Enrolment commenced at the Royal Children's Hospital Brisbane in January 2011 and at the Royal Children's Hospital Melbourne in July 2011.

2.4.1 Inclusion Criteria

- Parents of infants diagnosed with serious liver disease. Serious liver disease was defined as liver disease that may require transplantation in the future as diagnosed by the usual treating medical practitioner.
- Infants aged less than 2 years at diagnosis.
- The diagnosis of liver disease was made at least 3 months prior to study participation.

2.4.2 Exclusion Criteria

- Parental English that is inadequate to allow self-completion of the questionnaires.
- Families with separated parents. Two-parent families were sought in order to examine father engagement.

2.5 Ethics Approval

Ethics approval was obtained from the Human Research Ethics Committee (HREC) – Concord Repatriation General Hospital (a lead HREC), The University of Sydney HREC and each hospital's HREC. Written informed consent was obtained from all

participants. Copies of the HREC approval letters, Study Information Sheets and Consent Forms are included in Appendix A.

2.6 Study materials

2.6.1 Quantitative Measures

Each parent separately completed all self-report questionnaires used in the study.

2.6.1.1 Participant characteristics

Parents provided the author with the infant's date of birth, gender, birth order and number of children in the family, along with parent age, parent employment and highest level of education attained.

Each infant's diagnosis and date of diagnosis were obtained from the treating medical teams.

Family socio-economic status (SES) was calculated from the Australian Socioeconomic Index (AUSEI06, referred to in the current study as SEI).[220] The scale uses Australian labour force and census data to determine socio-economic status and also provides scores for those who are not in paid employment based on the highest level of education attained. It gives scores to a maximum of 100.0.

Birth order was dichotomised into infants born first and infants born second or later (referred to as 'first born child or later' in the data analysis), on the basis that adjustment to the birth of a first infant is likely to be a different stress for families compared with the birth of subsequent children.

2.6.1.2 Infant Illness Variables

2.6.1.2.1 Diagnosis

Due to the low numbers of infants in some of the diagnostic groups (for example, only one infant had a diagnosis of citrullinemia), infant diagnosis was dichotomised into

severe liver disease arising from Biliary Atresia and severe liver disease arising from other causes (referred to as 'Biliary Atresia or other severe liver disease' in the data analysis). Biliary Atresia is a non-genetic disorder. Corrective surgery for Biliary Atresia can postpone the need for liver transplantation by some years. Other severe liver diseases encompass rare autoimmune or genetic disorders for which liver transplant is the only surgical intervention available. In addition, previous research has shown better emotional outcomes in children who had a transplant for Biliary Atresia in comparison with other liver diseases.[142]

2.6.1.2.2 Severity of illness

There is no available standardized measure of severity in paediatric liver disease. Pediatric End-stage Liver Disease (known as PELD) scores are valid only for children who are in the end stage of liver disease[221] and are therefore not applicable to most infants in the present study. As proxy measures for illness severity the author calculated the ratio of days in hospital since onset of illness (the number of days in hospital divided by the number of days between onset of illness and the time of data collection) and the ratio of outpatient visits since onset of illness at each time period (the number of outpatient visits divided by the number of days between onset of illness and the time of data collection). Others have used a similar approach as a measure of disease severity. For example, DeMaso and colleagues[193,194] calculated the number of clinic visits and days of hospital per year as a measure of disease severity in a group of children who had heart transplantation.

Number of days in hospital and number of outpatient visits were obtained from a review of the medical records of each child and the ratios calculated for both the initial and follow up time points.

2.6.1.3 Parent psychological symptoms

The Depression Anxiety Stress Scales (DASS)[222] is a 42-item validated self-report scale that measures symptoms of depression, anxiety and stress experienced over the preceding one week. The scale provides scores for depression, anxiety, and stress symptoms in addition to a total symptom score. Higher scores indicate higher levels of psychological symptoms. It is a dimensional scale that indicates the likelihood that a mental disorder is present, but is not a diagnostic instrument. Each of the sub-scales has acceptable reliability with Cronbach's alphas ranging from .84 to .91.[222]

Given the large number of measures being used in the current study, the author decided to use the Total score of the DASS as a general measure of parent psychological symptoms. The Total score has a high Cronbach's alpha level of .97.[223]

2.6.1.4 Family functioning

The Family Assessment Device (FAD)[224] is a 60-item self-report questionnaire assessing family functioning. The FAD has been validated in healthy, clinical (psychiatric) and medically ill populations and provides healthy/unhealthy cut-off scores as well as mean scores for each of these groups.[225-227] It has good psychometric properties [225,227] and provides a measure of family functioning across seven sub-scales. Higher scores are associated with greater dysfunction in each sub-scale.

Although the complete FAD was administered to participants, given the large number of measures to be used in this study the author decided to use only the General Functioning sub-scale of the FAD to measure overall family functioning. The General Functioning sub-scale has been demonstrated by Byles and colleagues[228] to be a reliable and valid global assessment of family functioning independent of the other FAD sub-scales. The study reported good construct validity, demonstrated by

significant correlations between the General Functioning sub-scale and other family variables such as mental health of parents and marital disharmony of -.35 and .57 respectively. The authors also demonstrated good internal consistency with a Cronbach's alpha of .86,[228] similar to the findings of Kabacoff and colleagues who reported that the General Functioning sub-scale had the highest reliability of all the sub-scales.[227] The sub-scale is widely used in studies of children with chronic illness.[84,205,229] The 12 items used to calculate the General Functioning score are provided in Appendix C.

2.6.1.5 Impact of the infant's illness on the family

The Impact on Family Scale (IFS)[230,231] is a 27-item self-report questionnaire validated for use with families who have a child with a chronic illness. The scale provides a total score, with higher scores representing greater impact on the family of the child's illness. The first validation study of this instrument used a total score derived from 19 items, with internal reliability consistency of .88 (Cronbach's alpha). The 19-item total score was used in early studies that used the IFS.[232] In a later validation study[230] the developers revised the total score based on three separate studies in different patient populations. The later study found lower internal consistency alpha scores for two of the sub-scale items previously used to derive the total score, leading the authors to recommend using 15 items to derive the total score which had acceptable Cronbach's alpha scores of between .83 and .89. The study also demonstrated adequate construct validity through the scale's significant associations with maternal psychiatric symptoms, the child's poor general health and the child's poor psychological adjustment, with correlations ranging from .27 to .47.[230] An independent validity study supported the findings for the revised 15-item total score.[233]

The 15-item IFS total score is made up of items that have an impact on the family in three areas: practicalities of the illness; emotions; and relationship issues (Appendix D). The 19-item score includes four additional items concerning the financial impact of the illness on the family (Appendix D).

The present study uses the 15-item total score for the IFS. However the complete questionnaire was administered to parents, so it was possible to also calculate the 19-item total score for comparison with other research.

2.6.1.6 Fathers' engagement

The Dads' Active Disease Support scale (DADS)[234] is a 24-item validated self-report scale that asks about how much the father is engaged in tasks related to the sick child's illness and the helpfulness of his engagement. There is a separate form for mothers and fathers with scores for both the amount and the helpfulness of fathers' involvement. Higher scores reflect more involvement and greater perceived helpfulness. Cronbach's alphas for each parent and each aspect (amount and helpfulness) of the scores are high, ranging from .92 to .95, with test-retest reliability of .75 to .91 and good construct and convergent validity.[234]

2.6.1.7 Child emotional and behavioural outcome

The Child Behavior Checklist (CBCL)[235] is a 99-item parent-report questionnaire that has been used extensively both in research and clinical settings, including in populations of children and young people with chronic physical illness.[31,87,89] There are two versions: one for ages 1½ to 5 years (used in this study); and the second for ages 6 to 18 years. Each generates scores on a number of sub-scales that are classified into three factor scores: Internalising (emotional), Externalising (behavioural), and Total Problems scores. Raw scores for each of the three factors are converted into standardised T scores ranging from 50 to 100. T scores below 60

are regarded as 'non-clinical', scores 60 to 63 are 'borderline clinical' and scores of 64 and above are considered to indicate clinical psychopathology.[235] The Total Problems scale gives an overall measure of general problems and was the scale chosen for this study. The score is derived from all 99 items.

The CBCL 1½ to 5 years Total Problems scale has excellent test-retest reliability (Pearson correlation $r = .90$, $P < .01$) and inter-rater reliability (cross-informant correlation of .65, $P < .01$). Pre-schoolers who have been referred for behavioural and/or emotional problems have an Odds Ratio (OR) of 6 of scoring in the clinical range on the Total Problems scale when compared with non-referred preschool-aged children ($P < .01$). The scale has construct validity demonstrated by concurrent correlations ranging between .56 to .77 and predictive correlations between .56 and .75.[235] The Total Problems scale has also been validated in an international study, demonstrating only small differences in mean scores and internal consistency across multiple societies, with mean alpha scores of .94.[236]

It has been suggested that use of the CBCL in children with physical illness may result in an elevated Internalising Problems score because the Internalising Problems factor includes several items from a 'somatic complaints' sub-scale.[237] However, a meta-analysis[89] of studies that used the CBCL in children with chronic illness, including children with chronic liver disease, found no change in results on either the Internalising Problems or Total Problems scores after the somatic complaints sub-scale was removed, thus supporting the scale's use in this patient population.

The CBCL Total Problems score was used as the outcome measure in this study and therefore was only administered at the follow-up time period. As noted below, the infants were followed up 12 months after the initial assessment or when the infant turned 18 months of age, whichever was latest. All infants were therefore at least 18 months of age at the time that the CBCL was completed.

2.6.2 Qualitative Measures

2.6.2.1 Parent and family qualitative experience of illness

The author interviewed both parents together to ask them about their experience of their child's illness. The interviews were audio recorded and transcribed by the author.

The interview was semi-structured and based on four questions, with follow up questions asked as appropriate.

1. How has your child's illness affected your family?
    a. Emotionally
    b. Socially
    c. Relationships in the family
    d. Any other ways?
2. Other than your child's illness, have there been any other big stresses on your family?
    a. Please describe.
    b. How serious have these been?
    c. How have they affected the family?
3. What support do you have as a family?
    a. How would you describe this support?
4. Is there anything else you would like to add that will help me understand how things have been for you and your child?

2.7 Procedure

The treating medical practitioners and specialist nurses at each site identified and discussed the study with potential study participants. Parents were approached to participate approximately three months after the initial diagnosis, to allow early normative stress reactions to the diagnosis to resolve.[238,239]

Parents were given information about the study verbally and in writing during a routine hospital outpatient follow-up appointment or hospital admission. Parents who agreed to participate signed the study consent form and their contact details were given to the author.

The author contacted parents by phone to provide a detailed explanation of the research and to check ongoing consent.

Data was collected at two time periods, at approximately 3 months following the infant's diagnosis and about 12 months later, or when the infant was at least 18 months of age. See Table 2.1 for details of data collected at each time period.

Parents were sent the Time 1 questionnaires (DASS, FAD, IFS and DADS) with instructions for each parent to complete them individually prior to the parent interview. Parent interviews were arranged at a time and place preferred by participants. Parents chose to be interviewed at the hospital during the infant's admission, in the hospital outpatients department during a routine clinic visit, or at the family home. Interviews were conducted in a private room regardless of the setting.

The author checked that the questionnaires had been completed. If items had been unanswered, this was clarified with participants. Participants' reasons for not answering individual items were given as unintentional or not understanding the question. Clarification of the item allowed participants to answer the questions. No parent refused to answer any of the items.

About 12 months later the author contacted families again to participate in the follow-up phase of the study.

Parents who agreed to participate in the follow-up study completed all of the questionnaires again, with the addition of the CBCL. They were also interviewed again at a time and place preferred by participants. On this occasion, some participants were interviewed by phone, in which case parents returned the questionnaires to the author by mail.

The author conducted a review of each infant's medical chart to obtain accurate information about the diagnosis, date of diagnosis, number of clinic visits, and days of admission to hospital.

Table 2.1

Information Collected at Each Data Collection Period

Information collected	Time 1	Time 2
Infant's date of birth	✔	
Infant's diagnosis	✔	
Date of diagnosis	✔	
Family demographic information	✔	
DASS	✔	✔
FAD	✔	✔
IFS	✔	✔
DADS	✔	✔
CBCL		✔
Parent interview	✔	✔
Medical Chart review		✔

2.8 Quantitative Data Analysis

All quantitative data analyses were performed using the Statistical Package for the Social Sciences (SPSS, IBM corporation) version 22.0. Significance level was set at $P < .05$.

The qualitative data analysis was performed using NVivo (QSR International) version 10.

2.8.1 Sample Size Calculation

It was calculated that a sample size of 34 infants would allow demonstration of a mean within-subject change over time of 0.5 standard deviations, with significance at the 0.05 level and a power of 80%.

2.8.2 Score Distribution

The study measures (DASS, FAD, IFS and DADS) and continuous demographic and illness severity variables (SEI, parent age, ratio of outpatient visits, and ratio of days admitted to hospital) were examined for normal distribution. Skewness and kurtosis, histograms, box plots and normal and de-trended normal Q-Q plots were assessed. The Kolmogorov-Smirnov test was used to assess whether the study measures were significantly different from a normal distribution. If a measure was not normally distributed, attempts were made to transform the data to produce a normal distribution using log 10, square root or reciprocal transformations. Conversion to percentile scores was also undertaken if published percentile conversion charts were available.

Normally distributed data were analysed using one-sample T-tests for comparison with other published studies. The one-sample Wilcoxon signed rank test was used to analyse non-normally distributed data. The Wilcoxon signed rank test is the non-parametric equivalent of the one-sample T-test and is used to test whether the

median scores of the study data are significantly different from the median scores reported in other published studies.[240]

Data that were not normally distributed were assessed for skewness and outliers. If there was no significant skewness ($z < 2.0$) and no outliers on box plots, it was considered that the data could be used in regression analysis.[240]

2.8.3 Hypothesis 1

Parents of infants with serious liver disease will have high levels of distress, demonstrated by the presence of psychological symptoms and alterations in family functioning

To test the hypothesis, parent psychological symptom scores (measured by the DASS) were compared with published scores from the general population. Family functioning scores (measured by the FAD) were compared with published healthy/unhealthy cut-off scores. Parent ratings of the impact of the illness on the family (measured by the IFS) were compared with published scores.

2.8.3.1 Parent psychological symptoms

DASS raw scores were converted to percentile scores because the raw scores were not normally distributed (see Chapter 3, Quantitative Data Analysis Results). DASS percentile scores are not reported in the literature, except in one population study,[223] which provides median scores. To compare median scores, the one-sample Wilcoxon signed rank test was used for both parents' DASS scores at both time points.

2.8.3.2 Alterations in family functioning and impact of the illness on the family

The FAD General Functioning mean scores for mothers and fathers were compared with published healthy/unhealthy cut-off scores, using one-sample T-tests at both time points.

The IFS 19-item total mean scores for mothers and fathers were compared with published mean scores of families of children with chronic physical illness, using one-sample T-tests at both time points.

2.8.4 Hypothesis 2

Fathers' perceived engagement in the infants' care will have an impact on parent distress and family functioning

To test the hypothesis, analysis was undertaken of the relationship between fathers' engagement, parent psychological symptoms, and family functioning. Simple linear regression was used to analyse whether fathers' engagement (measured by the DADS) was predictive of parent psychological distress (measured by the DASS) and family functioning (measured by the FAD and IFS) for mothers and fathers at each time point.

2.8.5 Hypothesis 3

Parent distress, family functioning and fathers' engagement will have predictive value for the emotional and behavioural outcomes of the infants

To test the hypothesis, analysis was undertaken of the predictive value of the study measures for infant emotional and behavioural outcomes after controlling for significant demographic and illness variables. The demographic and illness variables were thus analysed first to determine which to include in the final analysis. The study measures were then analysed to determine which measures would be included in the development of the final best-fit hierarchical multiple regression (HMR) model for each parent.

2.8.5.1 Demographic and illness variables

The demographic variables of socio-economic status, parent age and the infant's birth order (first born child or later) were included in the analysis.

The disease variables (infant diagnosis, whether the infant had had a transplant or not, the ratio of outpatient visits and ratio of days admitted to hospital at each time point) were also included in the analysis.

The demographic and illness variables that significantly contributed to the model were then included in the final hierarchical multiple regression.

2.8.5.2 Building the hierarchical multiple regression model

Mothers' and fathers' data were analysed separately to build the best fit hierarchical multiple regression model for each parent.

Beta values were used to assess which measures to retain in the analysis. The beta value is a standardised value that indicates the magnitude of change (in standard deviations, *SD*) in the outcome measure for each one standard deviation of change in the predictor variable. For example, a beta value of 0.50 indicates that the outcome variable changes by 0.50 *SD* per 1 *SD* change in the predictor variable.[240] Any measure with a beta score significant at the $P < .10$ level was retained in the model.

If a variable made a significant contribution to the model at both time points, only the time point with the greatest contribution (highest beta value) was included in the analysis to minimise collinearity.

First, the demographic variables were entered as one block into a linear regression model, with CBCL Total Problems T scores as the dependent variable. The included demographic variables were Socio-Economic Index (SEI), parent age, and infant birth order (first born child or later).

Then, the infant illness variables were entered as one block into a separate linear regression model, with CBCL Total Problems T scores as the dependent variable. Time 1 and Time 2 illness variables were analysed separately. The included infant illness variables were diagnosis (Biliary Atresia or other severe liver disease),

whether the infant had had a liver transplant or not, ratio of outpatient visits, and ratio of days admitted to hospital.

Simple linear regression was performed for each of the study measures (DASS, FAD, IFS and DADS) separately at each time period, with CBCL Total Problems T scores as the dependent variable.

Then, the demographic and illness variables that contributed significantly to the model (variables with a beta value with a significance of < .10) were entered as the first block of a hierarchical multiple regression. Study measures (DASS, FAD, IFS and DADS) that contributed significantly to the model were entered individually as later blocks, thus analysing the study measures individually after controlling for the combined demographic and illness variables. The order of entry of each study measure was determined by the beta value. Measures with higher beta values were entered into the model before measures with lower beta values.

The model was refined after examining the analyses. The variables with significant contributions to the model, and which did not violate regression assumptions or produce unacceptable collinearity, were retained in the analysis and entered into the final best-fit models. Hierarchical multiple regression was then performed. Due to the number of variables included and the small sample size, in order to account for loss of predictive power the adjusted R^2 was used in reporting the overall contribution of variables to the model. Diagnostic analyses were conducted to ensure that assumptions of normality, linearity and homoscedasticity were not violated. Tests of collinearity were also performed.

2.9 Qualitative Data Analysis

2.9.1 Qualitative Data Collection

Participant interviews were conducted concurrently with the quantitative data collection to obtain additional data about parents' experiences of the infants' illness that could provide a deeper understanding of the quantitative data.

Initial qualitative analysis of the interview data comprised consideration of the themes evident across the cases during the collection and transcription of the interviews. Field notes were used to record particular observations or events that occurred during or around the interviews, as a method of aiding in the later analysis of the interview transcripts. A journal was kept to record the author's reflections on the qualitative data during the analysis, to assist with the development of ideas throughout the analysis.

2.9.2 Case Attributes

Assigning 'attributes' to cases, for example cases with high or low scores on the DASS, provides the opportunity to compare cases between and within groups. Continuous quantitative data, were transformed into categorical data before being imported into NVivo as case attributes. Parent scores on each of the study questionnaires (DASS, FAD, IFS, DADS and CBCL) were binned into two groups according to whether they were above or below the study group mean.

Parents were asked at the interview whether they had had any stressors in addition to the infant's illness. The number of stressors was recorded as a case attribute at each time point.

Parents were also asked at the interview about what social support was available to them and whether they felt that they had enough support. Parent perception of adequacy of support (yes or no) was recorded as a case attribute at each time point.

2.9.3 Selection of Cases for Qualitative Analysis

Cases were purposively selected for qualitative analysis, with the aim of ensuring inclusion of a wide range cases with different experiences and attributes.

Initial cases were selected from the author's recollection of the parent interviews, selecting a case whose parents described being highly stressed and a second case whose parents described coping well at the time of the interview. Further cases were then chosen for differing characteristics across the study period, such as parents who had to make major changes to their plans as a result of the infant's illness, a family with good social support and a family whose infant had a prolonged hospital admission.

Cases were then selected according to case attributes, based on the parent responses to the questionnaires, as described above. The analysis therefore included representation of cases with parent study questionnaire scores above and below the mean. Cases were also selected based on stability or change in parent measures over time, and difference or agreement between parent scores within families.

2.9.4 Coding the Transcripts

The interview transcripts were coded using the NVivo software (QSR International), version 10. The codes and categories were developed from an inductive perspective, based on the contents of the interviews rather than on *a priori* theory.[241]

The codes used for analysis were developed directly from the data, based on relevance of individual passages to the study hypotheses. Codes generated were grouped into categories of related items, and individual codes were later examined to ensure consistency of use and ongoing relevance to the project. Some codes were therefore discarded or merged with other codes that covered the same idea, or were

moved into a different category. The codes were re-examined in this way on several occasions as the analysis proceeded and the author's understanding of the overarching themes became more refined. Finally, codes that were different facets of the same essential experience were aggregated together. The process was enhanced by discussion with peers and supervisors.

Coding of the interview transcripts continued until no new codes were identified in the data and it was determined that saturation was reached.

The attributes of cases already coded were then examined to assess whether a full range of attributes were included and that there was adequate representation of infant diagnoses, infant gender and recruitment hospital. Additional cases were then selected for inclusion in the analysis based on this assessment.

The author then checked whether the remaining interviews contained any new concepts to ensure that saturation had been reached.

2.9.5 Thematic Analysis

The selected cases were analysed to identify the overarching themes in the parents' discussions of their experiences. Parents' discussion of the different phases and aspects of the infants' illness, their emotional responses to the illness, and discussion of relationships were used as a framework to identify the main themes. Consideration of the overall narrative contained in the qualitative data served to organise the themes to give a comprehensive understanding of parent experiences.

2.9.6 Integrated Quantitative and Qualitative Analytic Approach

The integrated analysis examined parent interview transcripts according to parent scores, as described above.

Firstly, the parents' interview discussion of their emotional reactions to the infants' illness was analysed, comparing families in which both parents reported fewer

psychological symptoms (DASS scores below the group mean) with those who reported more psychological symptoms (DASS scores above the group mean).

Secondly, the parents' discussion of family relationships and the effects of the illness on the family were analysed, comparing families in which both parents reported fewer problems in family functioning (FAD scores below the group mean) with those who reported more problems in family functioning (FAD scores above the group mean).

Thirdly, the parents' discussion of emotions, relationships and practicalities associated with the illness were analysed, comparing families in which both parents reported lower impact of the illness (IFS score below the group mean) with those who reported higher impact (IFS score above the group mean).

Fourthly, parent scores on the DASS, FAD and IFS were assessed in relation to parent reports of additional life stressors and the adequacy of social support obtained during the interviews.

Fifthly, parent discussion of parent emotional reactions to the infants' illness, family relationships and family response to the illness were analysed, comparing families with less father engagement (DADS scores below the group mean) with those who reported more father engagement (DADS scores above the group mean).

Finally, parent discussion of parent emotions, family relationships and discussion of fathers were analysed, comparing families who reported fewer infant emotional and behavioural problems (CBCL scores below the group mean) with those who reported more infant emotional and behavioural problems (CBCL scores above the group mean).

The integrated analysis therefore examined the quantitative data analytic results in light of the parents' direct expression of their experiences, providing an enhanced understanding of the meaning of the quantitative data.

2.9.7 Validity and Reliability of the Qualitative Analysis

The author personally conducted the parent interviews, transcribed the interviews, and checked each transcription against the original recording. This process ensured the accuracy of the transcripts and assisted with interpreting the participants' meaning and tone during interviews. The author reviewed all of the interviews and transcripts that were not included in the final data analysis to ensure that no information or themes had been missed.

The purposive selection of cases ensured adequate representation across the study sites and appropriate infant gender balance in line with that of the overall sample.

Reliability of the qualitative data was improved by the mixed methods design, which resulted in triangulation of information.[241] The author kept an audit trail journal about analytic decisions that were made during the study. To ensure reliable and valid interpretation of the transcripts, an independent coder (a trained research psychologist) was employed to review several of the coded transcripts at random, ensuring that the coding was sensible and meaningful and that the codes were appropriately fitted to the data.[242]

2.10 Conclusion

The current study has a mixed methods design. It collects both quantitative and qualitative data to enhance understanding of the illness experience in families who have an infant with serious liver disease. Mothers and fathers each completed a series of self-report questionnaires and parent interviews at two time points, at least three months after the infant's diagnosis and one year later.

The quantitative data analysis examines demographic and infant illness variables in addition to measures of parent psychological distress, family functioning, impact of

the illness on the family, fathers' engagement in the care of the infants, and infant emotional and behavioural outcomes.

The qualitative data consist of interviews with both parents together at each time point, asking parents about the family's experiences of the infant's illness, additional stressors and social supports.

The following chapter presents the results of the quantitative data analysis. The subsequent chapters present the results of the qualitative data thematic analysis and the integrated data analysis.

3 Quantitative Data Analysis Results

3.1 Synopsis

This chapter presents the results of the quantitative data analysis. Participant characteristics are presented, including demographic and infant illness variables. Following this are the results of the data analysis for each of the research hypotheses in turn. Summary tables of relevant statistics are included in the chapter. SPSS output tables for the major analyses are provided in the Appendices.

Mothers' median Depression Anxiety Stress Scale (DASS) total scores at Time 2 were significantly higher than the general population median ($P < .05$). Mothers' DASS scores at Time 1 and fathers' scores at both points were not significantly different from the general population.

Both parents' mean Family Assessment Device (FAD) General Functioning scores were significantly lower than the published healthy/unhealthy cut-off scores at both Time 1 and Time 2 ($P < .001$).

Fathers' mean Impact on Family Scale (IFS) scores were significantly lower than the published mean scores of parents of children with chronic illness at both time points ($P < .05$). Mothers' mean IFS scores did not significantly differ from the published mean scores of parents of children with chronic illness at either time point.

Fathers' mean Dads Active Disease Support (DADS) Amount scores were predictive of fathers' FAD scores at Time 2 ($P < .05$). That is, fathers who reported that they spent less time helping also reported greater problems in family functioning. Fathers' scores on the DADS Amount or Helpfulness scales were not significant predictors of the remaining study measures at either time point. Mothers' scores on the DADS scale were not significant predictors of the other study measures at either time point.

Hierarchical multiple regression analysis revealed different best fit models for mothers and fathers. For mothers, infant diagnosis other than biliary atresia, a higher ratio of outpatient visits at Time 2 and mothers' IFS score at Time 1 resulted in an overall significant model (*Adj* R^2 = .32, $F(3,33) = 6.62$, $P = .001$). For fathers, lower socioeconomic status, infant diagnosis other than biliary atresia, the infant having had a liver transplant at Time 1 and fathers' IFS score at Time 1 resulted in an overall significant model (*Adj* R^2 = .44, $F(4,32) = 8.12$, $P < .001$).

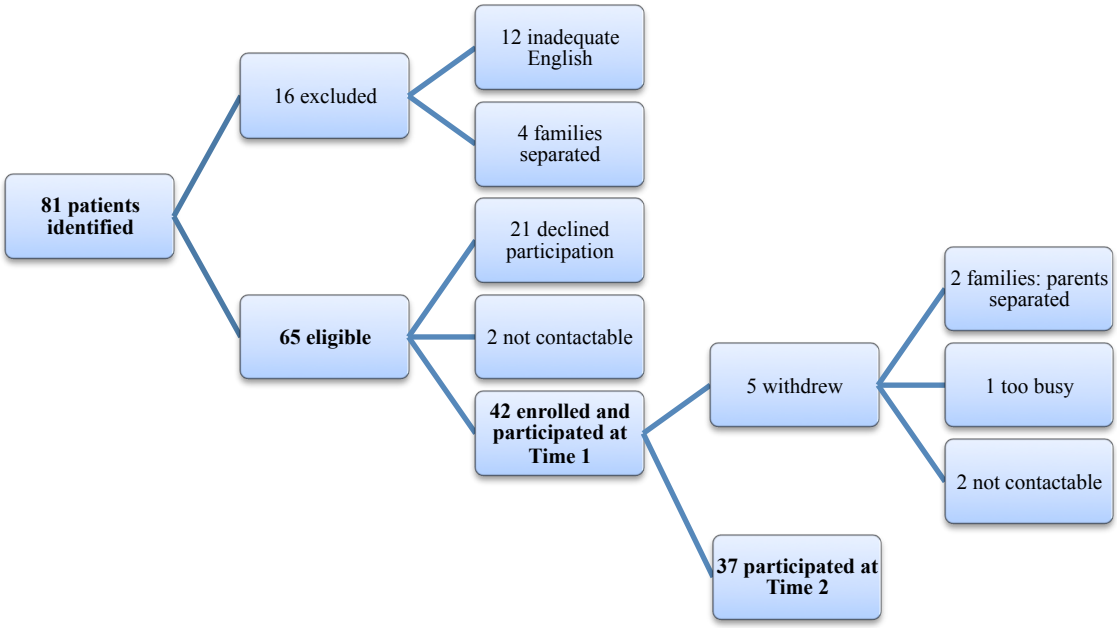
3.2 Participants

3.2.1 Study Recruitment and Participation

Eighty-one infants with serious liver disease were identified. Sixteen cases were excluded, 12 because the parents had inadequate English to be able to complete the study and four cases excluded because the parents were separated. Sixty-five families were eligible and were approached to participate in the study. Two eligible families could not be contacted. Of the 21 contactable families who declined to participate, eight declined citing time constraint, one declined as their child was too ill, one declined as the mother was suffering from postpartum depression and the remainder either did not give a reason or stated that they were not interested in the study. Forty-two eligible families agreed to participate in the study (participation rate 65%). Of these, 37 (88%) completed follow up (Figure 3.1).

Figure 3.1

Patients Screened for Study Enrolment



In two families, the parents had separated in the period between Time 1 and Time 2 and therefore did not participate in the follow up. Of the 37 families who completed follow up, one mother completed all measures except for the Time 2 IFS scale (no reason given) and three families completed the questionnaires but did not complete the follow up interview (one because of pregnancy and imminent delivery, two because a suitable time for the interview could not be found). Therefore, complete Time 1 and Time 2 questionnaires were available for all fathers and 36 mothers. All except the Time 2 IFS questionnaire were available for the remaining mother. There were interviews completed for all participants at Time 1 and for 34 participants at Time 2.

3.2.2 Participant Characteristics

Participant characteristics tables can be found in Appendix E.

There were 15 male infants (36%). The most common diagnosis was biliary atresia (60%). A quarter of the infants had already received a liver transplant at the time of study participation, and half had received a transplant by the time of follow up. Time from infant diagnosis to study participation varied widely due to difficulties contacting participants to arrange completion of study measures (Table 3.1).

Table 3.1

Participant Characteristics (N = 42)

Characteristic	***N* (%)**
Infant gender male	15 (36%)
Recruitment Hospital	
Children's Hospital Westmead	18 (43%)
Sydney Children's Hospital	5 (12%)
Royal Children's Hospital Brisbane	8 (19%)
Royal Children's Hospital Melbourne	11 (26%)
Diagnosis[a]	
Biliary Atresia	25 (60%)
Alpha-1 Antitrypsin Deficiency	6 (14%)
Alagille Syndrome	3 (7%)
Autoimmune Hepatitis	3 (7%)
Cryptogenic Hepatitis	3 (7%)
Citrullinemia	1 (2%)
Hepatoblastoma	1 (2%)
Liver Transplant at Time 1 (N = 42)	11 (26%)
Liver Transplant at Time 2 (N = 37)	19 (51%)
	Median (range)
Child age at diagnosis (days)	61 (2 – 700)
Mother's age (years)	34 (19 – 46)
Father's age (years)	36 (20 – 53)
Time from diagnosis to study participation (days)	206 (80 – 485)
Ratio of outpatient visits from illness onset to Time 1	.04 (.00 - .18)
Ratio of outpatient visits from illness onset to Time 2	.03 (.01 - .10)
Ratio of days in hospital from illness onset to Time 1	.18 (.00 - .98)
Ratio of days in hospital from illness onset to Time 2	.12 (.00 - .49)

Note. Ratio of outpatient visits is calculated by the number of outpatient visits divided by the number of days between onset of illness and time of data collection
Ratio of days in hospital is calculated by the number of days in hospital divided by the number of days between onset of illness and time of data collection

[a] Totals do not add to 100% due to rounding

3.3 Score Distribution

See Appendix F for the descriptive statistics, normality tests and normality plots for the family demographic and infant illness variables.

See Appendix G for the descriptive statistics, normality tests and normality plots for the study measures (DASS, FAD, IFS, DADS).

3.3.1 Family Demographics Distribution

Continuous demographic and illness severity variables were examined for normality of distribution. Parent age was normally distributed. Socioeconomic Index was not normally distributed and did not become so with transformation. However, SEI was not significantly skewed (skewness = -.12, *SE* = .37, *z* = 0.32) and there were no outliers identified, so it was considered that it could be used in regression analysis (Appendix F).

3.3.2 Infant Illness Severity Distribution

The ratio of outpatient appointments and ratio of days of admission at each time point were not normally distributed. Square root transformation resulted in normal distribution and the transformed variables were therefore used in the data analysis (Appendix F).

3.3.3 DASS Score Distribution

Initial examination of the DASS total scores showed positively skewed data at both time periods, which was also identifiable on histograms. Normal and de-trended Q-Q plots also indicated non-normally distributed data, and there were outliers on box plots in the Time 1 data. The Kolmogorov-Smirnov test was significant, indicating non-normally distributed data. Transformations of the data (log 10, square root and reciprocal transformations) did not normalise the data.

Crawford and Henry provide a table to convert raw scores to percentiles.[223] Conversion of the data to percentiles improved the skewness and kurtosis *z* scores to non-significant levels (that is, $z < 2.0$) with the exception of the kurtosis *z* score for fathers' DASS total percentile scores at Time 2 (kurtosis = -1.59, $SE = 0.76$, $z = 2.1$). The normality plots also improved and there were no longer outliers present on the box plots. The Kolmogorov-Smirnov test was no longer significant for mothers' scores at both time points and for fathers' scores at Time 1. However, fathers' scores at Time 2 remained non-normally distributed (Kolmogorov-Smirnov test $P < .001$), though there were not outliers on the box plot (Appendix G). The DASS percentile scores were therefore chosen for analysis.

Fathers' DASS scores at Time 2 were considered suitable for regression analysis because there was no significant skewness (skewness = 0.20, $SE = 0.39$, $z = 0.51$) and there were no outliers on the box plot.[240]

3.3.4 Family Assessment Device Score Distribution

The Kolmogorov-Smirnov test approached significance for mothers' FAD General Functioning at Time 2 ($P = .05$) and was significant for fathers' FAD General Functioning scores at Time 2 ($P = .01$). However, calculation of the skewness and kurtosis *z* scores for each of these did not reveal significant skewness or kurtosis. Further, the histograms, normal Q-Q plots and de-trended normal Q-Q plots did not appear to be significantly non-normal and there were no outliers on box plots (Appendix G). Therefore parametric tests were used for the FAD, and non-parametric tests were used to check any results that had a significance level between 0.05 and 0.01.

3.3.5 Impact on Family Scale Score Distribution

Both parents' IFS scores at both time points were normally distributed (Appendix G).

3.3.6 Dads Active Disease Support Scale Score Distribution

Mothers' scores on the DADS Amount and Helpfulness scales were normally distributed at both time periods.

Fathers' scores on the DADS Amount and Helpfulness scales at Time 1, and DADS Amount scale at Time 2 were normally distributed. Although the Kolmogorov-Smirnov test was significant for fathers' DADS Helpfulness scores at Time 2 ($P = .002$), calculation of the skewness and kurtosis z scores did not reveal significant skewness or kurtosis. Further, the histogram, normal Q-Q plot and de-trended normal Q-Q plot did not appear to be significantly non-normal and there were no outliers on box plots (Appendix G). Therefore DADS Helpfulness scores were used in regression analysis.

3.4 Hypothesis 1

Parents of infants with serious liver disease will have high levels of distress, demonstrated by the presence of psychological symptoms and alterations in family functioning

Parents' psychological symptoms scores (measured by the DASS) were compared with scores from the general population. Family functioning scores (measured by the FAD) were compared with published healthy/unhealthy cut-off scores. Parent ratings of the impact of the illness on the family (measured by the IFS) were compared with published scores. See Appendix H for statistics tables.

The analysis demonstrated that the hypothesis was partially supported. Mothers' psychological symptom scores were significantly higher (more symptoms) at Time 2 when compared with scores from the general population, but fathers' scores were not significantly different from the general population at either time point. Family functioning scores were significantly lower (more healthy) when compared with the published healthy/unhealthy cut-off scores for mothers and fathers at both time

points. However, mothers' ratings of the impact of the illness on the family were comparable with published research in families of children with chronic illness. Fathers' ratings of the impact of the illness on the family were significantly lower than findings from the published research.

3.4.1 Parent Psychological Symptoms

Descriptive statistics tables for the DASS raw and percentile scores are presented in Appendix H.

DASS percentile scores are not reported in the literature, with the exception of Crawford and Henry's normative population data[223] which provide a median percentile score of 50. The one-sample Wilcoxon signed rank test was therefore used to compare both parents' DASS total median scores with the normative data to assess whether parent scores are elevated compared to the general population. Mothers' median DASS scores were not significantly different from the population median at Time 1, but were significantly higher than the population median at Time 2. Fathers' DASS scores did not differ from the population median at either time point (Table 3.2).

Table 3.2

One-Sample Wilcoxon Signed Rank Test of Parents' DASS Percentile Scores Compared with the Population Median

	Population median = 50.0					
	Time 1			Time 2		
	N	Median	*Z*	*N*	Median	*Z*
Mothers' DASS Score	42	60.0	1.67	37	60.0	1.97*
Fathers' DASS Score	42	47.5	-0.83	37	40.0	-1.78

*$P < .05$

3.4.2 Alterations in Family Functioning

3.4.2.1 Family Assessment Device

Descriptive statistics tables for parent FAD scores are presented in Appendix H.

One-sample T-tests were used to compare mean scores with the published healthy/unhealthy cut-off score (cut-off = 2.00).[226]

Mothers' and fathers' scores on the FAD at both time points were significantly lower (that is, within the healthy range) than the published healthy/unhealthy cut-off score (Table 3.3). Fathers' scores at Time 2 were not checked with non-parametric tests because they were significantly lower than the cut-off score at a significance level of 0.001.

Table 3.3

One-Sample T-Test of Mean Differences in Parents' FAD General Functioning Mean Scores Compared with the Healthy/Unhealthy Cut-Off Score

	Healthy/unhealthy cut-off = 2.00							
	Time 1				Time 2			
FAD Score	*t*	*df*	Mean diff.	95% CI	*t*	*df*	Mean diff.	95% CI
Mothers	-7.06*	41	-0.41	[-0.52, -0.29]	-5.44*	36	-0.39	[-0.54, -0.25]
Fathers	-6.13*	41	-0.39	[-0.52, -0.26]	-5.41*	36	-0.35	[-0.48, -0.22]

Note. $N = 42$ (Time 1), $N = 37$ (Time 2).
Mean diff. = Mean difference, 95% CI = 95% Confidence Interval
* $P < .001$

3.4.2.2 Impact on Family Scale

Impact on Family Scale (IFS) scores for mothers and fathers were calculated for the 15-item and 19-item totals to allow comparisons with other research (Appendix H). As noted above, one mother who participated at Time 2 completed all measures except for the IFS at Time 2. Therefore the Time 2 IFS data for mothers is based on a sample size of 36.

The developers of the IFS used the 19-item total and report a mean score of 48.03 (*SD* = 8.20) in their group of 209 families of children with a range of chronic illnesses.[243] Mothers' scores in the present study were not significantly different from the published mean scores, while fathers' scores were significantly lower (that is, indicating less impact of the illness on the family) than the published scores at both time points (Table 3.4).

Table 3.4

One-sample T-test of Mean Differences in Parents' IFS 19-item Total Mean Scores Compared with the Published Mean Score for Families of Children with Chronic Illness

	Mean score for families of children with chronic illness = 48.03							
	Time 1				Time 2			
IFS Score	*t*	*df*	Mean diff.	95% CI	*t*	*df*	Mean diff.	95% CI
Mothers	-0.17	41	-0.34	[-4.34, 3.66]	-0.97	35	-2.42	[-7.49, 2.65]
Fathers	-2.31*	41	-3.96	[-7.43, -0.49]	-2.28*	36	-5.35	[-10.12, -0.59]

Note. *N* = 42 (Time 1), *N* = 36 (mothers, Time 2), *N* = 37 (fathers, Time 2)
Mean diff. = Mean difference, 95% CI = 95% Confidence Interval
* *P* < .05

3.5 Hypothesis 2

Fathers' perceived engagement in the infants' care will have an impact on parent distress and family functioning

Simple linear regression was used to analyse whether fathers' engagement in the care of the infants (measured by the DADS) was predictive of parent psychological symptoms (measured by the DASS) and family functioning (measured by the FAD and IFS). See Appendix I for statistics tables.

The hypothesis was partially supported. Fathers who reported helping less also reported more problematic family functioning at Time 2, but otherwise fathers' DADS scores were not predictive of the other study measures. Mothers' ratings of father engagement were not predictive of other study measures.

3.5.1 Fathers' Engagement

Descriptive statistics tables for parents' DADS scores are presented in Appendix I.

3.5.1.1 Mothers' DADS scores

Mothers' scores on the DADS were not significant predictors of mothers' scores on the other study measures at either time point (Appendix I).

3.5.1.2 Fathers' DADS scores

Fathers' DADS Amount scores were predictive of fathers' FAD scores at Time 2 (R^2 = .14, $F(1,35) = 5.45$, ß = -.37, $P < .05$). Fathers who reported spending less time helping (lower Amount scores) also reported more problematic family functioning (higher FAD scores). Fathers' scores on the DADS were not significant predictors of any of the other study measures (Appendix I).

3.6 Hypothesis 3

Parent distress, family functioning and fathers' engagement will have predictive value for the emotional and behavioural outcomes of the infants

Family demographic and infant illness variables were analysed in a linear regression model to determine which variables to include in the development of the hierarchical multiple regression model.

The study measures were also analysed in individual simple linear regression to determine which variables to include in the development of the hierarchical multiple regression model.

Beta values were used to assess which measures to retain in the analysis. Any measure with a beta score significant at the $P < .10$ level was retained in the model.

The hypothesis was then tested using hierarchical multiple regression, entering any study measure that was a significant predictor of infant outcome after controlling for any demographic or illness variables that were also associated with infant outcome. See Appendix J for statistics tables.

3.6.1 Demographic Variables as Predictors of Infant Outcome

The demographic variables SEI, parent age, and infant birth order (first born or not first born) were entered as a single block into a linear regression analysis, with CBCL Total Problems T score as the outcome variable. Any measure with a beta score significant at the P < .10 level was retained in the model.

3.6.1.1 Mothers' demographic variables

The analysis resulted in an overall significant model ($R^2 = .21$, $F(3,33) = 2.97$, $P <$ .05). Of the three variables entered, only SEI made a significant contribution to the model (ß = -.30, $P = .07$) and was therefore retained in the regression analysis.

3.6.1.2 Fathers' demographic variables

The analysis resulted in an overall significant model (R^2 = .21, F(3,33) = 2.98, P < .05). Of the three variables entered, only SEI made a significant contribution to the model (ß = -.40, P < .05) and was therefore retained in the regression analysis.

3.6.2 Illness Severity Variables as Predictors of Infant Outcome

The illness severity variables diagnosis (biliary atresia or other severe liver disease), whether the infant had had a liver transplant or not, ratio of outpatient visits, and ratio of days admitted to hospital were entered as a single block into a linear regression analysis, with CBCL Total Problems T score as the outcome variable. Any measure with a beta score significant at the P < .10 level was retained in the model.

3.6.2.1 Mothers' illness severity variables

Time 1 was analysed first. None of the Time 1 variables made a significant contribution to mothers' CBCL scores and so were not retained in the regression analysis.

Time 2 was then analysed. The analysis resulted in an overall significant model (R^2 = .30, F(4,32) = 3.35, P < .05). Diagnosis (ß = -.33, P = .05) and ratio of outpatient visits at Time 2 (ß = .72, P = .006) both made a significant contribution to the model and were therefore retained in the analysis.

3.6.2.2 Fathers' illness severity variables

Time 1 was analysed first. The analysis resulted in an overall significant model (R^2 = .28, F(4,32) = 3.05, P < .05). Diagnosis (ß = -.37, P = .03) and whether the infant had had a liver transplant or not at Time 1 (ß = .31, P = .09) both made a significant contribution to the model and were therefore retained in the regression analysis.

Time 2 was then analysed. The analysis resulted in an overall significant model (R^2 = .29, F(4,32) = 3.30, P < .05). Diagnosis (ß = -.48, P = .007) was the only variable that made a significant contribution to the model.

Infant diagnosis (biliary atresia or other severe liver disease), and whether the infant had had a liver transplant or not at Time 1 were therefore retained in the regression analysis.

3.6.3 Study Measures as Predictors of Infant Outcome

Simple linear regression analysis of each study measure was undertaken to assess whether the measures DASS, FAD, IFS and DADS were significant predictors of the infants' outcome measured by the CBCL at Time 2.

3.6.3.1 Mothers' study measures

Mothers' DASS scores at Time 2 and IFS scores at both time points were significant predictors of mothers' CBCL scores (Table 3.5). Mothers' IFS scores at Time 1 had a higher beta value than mothers' IFS scores at Time 2. Therefore mothers' IFS scores at Time 1, as well as mothers' DASS scores at Time 2, were entered into the analysis.

Mothers' DASS scores at Time 1, and FAD scores and DADS scores at both time points were not significant predictors of mothers' CBCL scores (Appendix J).

Table 3.5

Mothers' Study Measures: Predictors of Mothers' CBCL Scores

	Infant emotional and behavioural outcomes at Time 2 (CBCL)						
Predictor	*N*	B	*SE*	β	*t*	Sig.	95% CI for B
Mother DASS Total Percentile Time 2	37	0.12	.066	.293	1.81	.079	[-0.01, 0.25]
Mother IFS 15-item total Time 1	37	0.48	.160	.450	2.98	.005	[0.15, 0.80]
Mother IFS 15-item total Time 2	36	0.41	.146	.432	2.80	.008	[0.11, 0.71]

Note. SE = Standard Error. 95% CI = 95% Confidence Interval

3.6.3.2 Fathers' study measures

Fathers' DASS scores at Time 1 and IFS scores at both time points were significant predictors of fathers' CBCL scores (Table 3.6). Fathers' IFS scores at Time 1 had a higher beta value than fathers' IFS scores at Time 2. Therefore fathers' IFS scores at Time 1, as well as fathers' DASS scores at Time 1, were entered into the analysis. Fathers' DASS scores at Time 2, and FAD scores and DADS scores at both time points were not significant predictors of fathers' CBCL scores (Appendix J).

Table 3.6

Fathers' Study Measures: Predictors of Fathers' CBCL Scores

	Infant emotional and behavioural outcomes at Time 2 (CBCL)						
Predictor	*N*	B	*SE*	β	*t*	Sig.	95% CI for B
Father DASS Total Percentile Time 1	37	.12	.068	.280	1.73	.093	[-0.02, .026]
Father IFS 15-item total Time 1	37	.58	.201	.435	2.86	.007	[0.17, 0.99]
Father IFS 15-item total Time 2	37	.43	.153	.429	2.81	.008	[0.12, 0.74]

Note. *SE* = Standard Error. 95% CI = 95% Confidence Interval

3.6.4 Hierarchical Multiple Regression Results

Tables and graphs of the SPSS output for the Hierarchical Multiple Regression analysis can be found in Appendix J.

3.6.4.1 Mothers' hierarchical multiple regression

Development of the model for mothers identified SEI, infant diagnosis (biliary atresia or not), and ratio of outpatient visits at Time 2 as significant predictors of mothers'

CBCL Total Problems T score. The three variables were entered as Block 1 into the regression analysis.

Mothers' IFS score at Time 1 had a higher beta value (ß = .45, *P* = .005) than mothers' DASS score at Time 2 (ß = .29, *P* = .079). Therefore, mothers' IFS score at Time 1 was entered as Block 2 and mothers' DASS score at Time 2 was entered as Block 3 into the regression analysis.

The analysis resulted in an overall significant model, but SEI was no longer a significant contributor to the model (ß = -.11, *P* = .49) and there was multi-collinearity between IFS and SEI (Appendix J). SEI was therefore removed from the analysis.

Mothers' IFS score was a significant contributor to the model at Block 2 (ß = .32, P = .08) and it was therefore retained for further analysis.

Mothers' DASS score was not a significant contributor to the model at Block 3 (ß = .21, *P* = .16).

Removing SEI and mothers' DASS score at Time 2 from the model resulted in a significant model with no violation of assumptions of multiple regression and no multi-collinearity.

For mothers, the best-fit HMR model therefore included the infant diagnosis (biliary atresia or other severe liver disease), ratio of outpatient visits at Time 2 and mothers' IFS score at Time 1. The overall model was significant (*Adj* R^2 = .32, $F(3,33) = 6.62$, *P* = .001). Mothers' IFS score contributed 10.7% to the variation in the CBCL score (ΔR^2 = .107, $\Delta F(1,33) = 5.68$, *P* < .05). The Durbin-Watson test was close to a score of 2, indicating that the assumption of independence of errors was not violated. All three predictors had ß scores with significance of < .10 and therefore were significant contributors to the model (Table 3.7).

Table 3.7

Hierarchical Multiple Regression Analysis Predicting Infant Emotional and Behavioural Outcomes from Infant Illness Variables and Mothers' IFS Score

	Infant emotional and behavioural outcomes					
Predictor	ΔR^2	B	*SE*	β	*t*	95% CI for B
Step 1	.27**					
(Constant)		32.71	5.568		5.87	[21.39, 44.02]
Biliary atresia or other severe liver disease		-8.56	3.633	-.372*	-2.36	[-15.94, -1.18]
Ratio of outpatient visits Time 2		101.87	30.731	.523**	3.32	[39.42, 164.32]
Step 2	.11*					
(Constant)		24.86	6.174		4.03	[12.29, 37.42]
Biliary atresia or other severe liver disease		-9.34	3.421	-.405*	-2.73	[-16.30, -2.37]
Ratio of outpatient visits Time 2		65.68	32.574	.337‡	2.02	[-.59, 131.95]
Mother IFS 15-item total Time 1		0.41	0.171	.384*	2.38	[0.06, 0.75]
Total R^2	.38**					

Note. Outcome variable: Mothers' CBCL Total Problems T Score
Step 1 $R^2 = .27$, $\Delta F(2,34) = 6.24$
Step 2 $R^2 = .38$, $\Delta F(1,33) = 5.68$
Biliary atresia coded 1, other severe liver disease coded 0
Ratio of outpatient visits is calculated by the number of outpatient visits divided by the number of days between onset of illness and time of data collection
‡ $P < .10$. * $P < .05$. ** $P < .01$

Tolerance statistics were all close to 1.0 (well above 0.2) and VIF statistics were all close to 1.0 (well below 10), indicating that collinearity was not a problem in the model. Collinearity diagnostics indicated that the predictors were spread across different eigenvalues, indicating no problems of multi-collinearity. There were no cases outside the standardised residuals limit of 3. Normality of residuals was demonstrated by the normal distribution on histogram and little deviation from

normality on the residuals normal probability plot. The scatterplot of standardised residuals and standardised predicted values showed random distribution around 0, demonstrating normally distributed residuals. The partial regression plots for outpatient visits at Time 2 (sqrt transformation ratio OPD Time 2) and IFS scores at Time 1 were also randomly distributed around the value of 0, demonstrating normally distributed residuals (Appendix J).

3.6.4.2 Fathers' hierarchical multiple regression

Development of the model for fathers identified SEI, infant diagnosis (biliary atresia or not), and whether the infant had had a transplant at Time 1 as significant predictors of fathers' CBCL Total Problems T scores. The three variables were entered as Block 1 into the regression analysis.

Fathers' IFS score at Time 1 had a higher beta value (ß = .44, $P = .007$) than fathers' DASS score at Time 1 (ß = .28, $P = .093$). Therefore, fathers' IFS score at Time 1 was entered as Block 2 and fathers' DASS score at Time 1 was entered as Block 3 into the regression analysis.

The analysis resulted in an overall significant model, but fathers' DASS score at Time 1 was not a significant contributor to the model (ß = .01, $P = .952$) and it was therefore removed from the analysis (Appendix J).

Removing fathers' DASS score at Time 1 from the model resulted in a significant model with no violation of assumptions of multiple regression and no multi-collinearity.

For fathers, the best-fit HMR model therefore included socio-economic status, the infant diagnosis (biliary atresia or other severe liver disease), whether the infant had had a liver transplant at Time 1 or not, and fathers' IFS score at Time 1. The overall model was significant ($Adj\ R^2 = .44$, $F(4,32) = 8.12$, $P < .001$). Fathers' IFS score

contributed 14.8% to the variation in CBCL score (ΔR^2 = .148, $\Delta F(1,32) = 9.55$, $P <$ .01). The Durbin-Watson test was close to a score of 2 and therefore the assumption of independent errors was not violated. Each of the predictors significantly contributed to the model (Table 3.8).

Table 3.8

Hierarchical Multiple Regression Analysis Predicting Infant Emotional and Behavioural Outcomes from Family Demographics, Infant Illness Variables and Fathers' IFS Score

	Infant emotional and behavioural outcomes					
Predictor	ΔR^2	B	*SE*	β	*t*	95% CI for B
Step 1	.36**					
(Constant)		57.47	4.768		12.05	[47.77, 67.17]
SES		-0.16	0.070	-.336*	-2.33	[-0.30, -0.02]
Biliary atresia or other severe liver disease		-5.43	3.382	-.231	-1.61	[-12.31, 1.45]
Liver transplant at Time 1 or not		9.85	3.971	.348*	2.48	[1.77, 17.93]
Step 2	.15**					
(Constant)		39.02	7.328		5.32	[24.09, 53.95]
SES		-0.13	0.063	-.277*	-2.13	[-0.26, -0.01]
Biliary atresia or other severe liver disease		-7.37	3.079	-.314*	-2.39	[-13.64, -1.1]
Liver transplant at Time 1 or not		7.72	3.605	.273*	2.14	[0.37, 15.06]
Father IFS 15 item total Time 1		0.53	0.172	.402**	3.09	[0.18, .88]
Total R^2	.50***					

Note. Outcome variable: Fathers' CBCL Total Problems T Score
Step 1 R^2 = .36, $\Delta F(3,33) = 6.07$
Step 2 R^2 = .50, $\Delta F(1,32) = 9.55$
SES = Socio-Economic Status (measured by SEI)
Biliary atresia coded 1, other severe liver disease coded 0
Liver transplant coded 1, no liver transplant coded 0
* $P < .05$. ** $P < .01$. *** $P < .001$

Tolerance statistics were all close to 1.0 (well above 0.2) and VIF statistics were all close to 1.0 (well below 10), indicating that collinearity was not a problem in the model. Collinearity diagnostics indicated that the predictors were spread across different eigenvalues, indicating no problems of multi-collinearity. There were no cases outside the standardised residuals limit of 3. Normality of residuals was demonstrated by the normal distribution on histogram and little deviation from normality on the residuals normal probability plot. The scatterplot of standardised residuals and standardised predicted values showed random distribution around 0, demonstrating normally distributed residuals. The partial regression plots for SEI and IFS scores at Time 1 were also randomly distributed around the value of 0, demonstrating normally distributed residuals (Appendix J).

3.7 Conclusion

This chapter reported the quantitative data analysis results for the study. At the follow up period one year after the infants' diagnosis, the mothers (but not the fathers) of infants with serious liver disease reported high levels of distress on a psychological symptoms measure, when compared with the general population. However, both mothers and fathers reported family functioning to be within the normal healthy range at both time periods. Mothers' reports of the impact of the infant's illness on the family were in line with other research, while fathers reported significantly lower impact on the family when compared with previously published work.

Fathers' engagement in the medical care of the infants was not a predictor of the mothers' psychological symptoms, mothers' perceptions of family functioning or mothers' ratings of the impact of the illness on the family. At the follow up time period, fathers who reported spending less time helping in the care of the infant also reported greater problems in family functioning.

Results of hierarchical multiple regression analysis are presented. There were commonalities and differences between the parents. For both parents, infant diagnosis, illness severity and perception of the impact of the illness on the family significantly contributed to the final best-fit model. For mothers, the best-fit model included serious liver disease other than biliary atresia, a greater rate of outpatient visits at the follow up period and mothers' scores on the Impact on Family Scale at the initial time period. For fathers, the best-fit model included lower socioeconomic status, serious liver disease other than biliary atresia, the child having had a liver transplant at the initial time period and fathers' scores on the Impact on Family Scale at the initial time period.

The following chapter will present the results of the qualitative data analysis and the subsequent chapter will present the final integrated mixed methods analysis.

4 Qualitative Data Thematic Analysis Results

4.1 Synopsis

The previous chapter presented the results of the quantitative data analysis. This chapter presents the results of thematic analysis of the parent interviews. The following chapter will present the results of the integrated quantitative and qualitative data analysis.

Cases were selected purposively to ensure broad representation of issues across the analysis, resulting in 25 families being included in the qualitative and integrated analyses.

Thematic analysis of the selected interview transcripts identified six themes of importance discussed by parents: uncertainty; infant vulnerability; isolation; dealing with other aspects of life; shared experience; and adjustment.

The chapter begins with an outline of the results of the case selection, case coding and identification of themes. Each theme is then described in full, with examples from the interview data demonstrating the different aspects of the themes.

Overall, the thematic analysis demonstrated that the experience of parents of infants newly diagnosed with serious liver disease is marked by uncertainty throughout the course of the infants' illness during the study period. Uncertainty commenced at the time of realisation that the infant was ill, and continued throughout the period of physical investigations, diagnosis, and treatment including surgery and hospital admissions. The ongoing management of the illness after discharge from hospital was also characterised by uncertainty for parents. The infants' vulnerability was emphasised by the uncertainties of the illness, as well as by the distressing investigations and treatments that the infants underwent. Parent awareness of infant

vulnerability was further added to by the reality of the illness and its complications including the risk of infection and mortality. Parent distress related to uncertainty and infant vulnerability, in combination with periods of time in hospital, added to a sense of isolation for many parents. Other life pressures compounded parent distress, adding further stress to parents' already stressful situation. The combination of uncertainty, infant vulnerability, isolation and dealing with other aspects of life emphasised the protective capacity of social relationships for parents. A sense of shared experience for parents cemented relationships and provided support through understanding. Adjustment was a necessity as parents tried to cope in order to be able to continue to care for their infant. Parents coped and adjusted in different ways, often depending on their infant's progress. As time passed many parents were able to feel more positive and developed a new sense of perspective.

4.2 Case Selection and Generation of Codes

As described in Chapter 2, Methods, cases were purposefully selected for qualitative analysis to ensure that a broad range of parent experiences was included. Initial cases were selected based on the author's recollection of the interviews and review of the transcripts, with families chosen for their differing experiences. The interview transcripts were coded for content. Further cases were then selected to ensure inclusion of cases with different case attributes (developed from parent results of the self-report questionnaires). Three families scored below the group mean on the DASS, FAD and IFS at both time points and on the CBCL at Time 2, but each of these families' parents differed in their responses on the DADS. One of these families was included in the analysis, chosen to ensure adequate representation of infant diagnosis and recruitment hospital in the group as a whole. No family scored above the group mean on all questionnaires at both time points. Saturation was reached when 16 cases had been coded. An additional eight cases were then selected to

ensure appropriate infant gender balance (in line with the total sample), and also to ensure that each study site was represented adequately, based on the overall numbers of cases from each site. A final review of the remaining transcripts resulted in the identification of an additional code in one case transcript, leading to the inclusion of that case in the qualitative analysis. The purposeful selection of cases for analysis therefore resulted in 25 families being chosen for the qualitative and integrated (mixed methods) analyses. Appendix K shows summary information for each case included in the analysis and the reasons for inclusion. All identifying data have been removed to protect individuals' privacy and pseudonyms are used throughout this thesis and in Appendix K.

The interview transcripts were coded as described in Chapter 2, Methods. The codes were organised into eight categories of related items (See Appendix L). The eight categories arose directly from the parents' discussion and are listed below:

1. Emotions
2. External circumstances
3. The hospital experience
4. The infant
5. Physical aspects of the infant's illness
6. Relationships
7. The response of the family to the infant's illness
8. Social support

Thematic analysis was undertaken using the selected cases. Six overarching themes were identified: uncertainty; infant vulnerability; isolation; dealing with other aspects of life; shared experience; and adjustment.

4.3 Uncertainty

Uncertainty was a feature of all aspects of the parents' experience of the infants' illness that was associated with significant distress for parents and contributed to parents' sense of lack of control over what was happening for their infants. Uncertainty resulted in problems with planning daily activities and created worry for parents about the future for their infant and the family. The infants' unpredictable physical state restricted family activity and added to parent distress.

4.3.1 Uncertainty during the period of diagnosis

Uncertainty was important during the period of diagnosis as parents first realised their infant was ill and waited for confirmation of the illness. The time to definitive diagnosis varied for each infant, leading to varying periods of uncertainty for each family while investigations proceeded.

> Peter: We were very happy when the day, you know, he was born. But this was noticed after the second day or third day, when he, after his birth. So from the time we, till such time we didn't feel the seriousness of it. Till such time, yeah. The third day, when we discussed with the doctor, he said you know, 'he has to go for some scans, make sure that there's no problem in his liver'. And we just went for the scan and they said 'it's ok, but there's some small blockage might be, should be ok if he has to undergo one surgery and then should be ok'. Till such time, we didn't feel the seriousness of it. And once we got all the information from the doctors and we get some information from the internet and then we realised the, you know, seriousness of it. But even now we can't digest what's happening … We can't digest, we can't, we can't accept what's happening now.

Following diagnosis, parents worried about the implications for the infant's future health and possible mortality.

> Alan: I suppose one thing that has been throughout has been the difficulty with this diagnosis and the unknowns around the diagnosis. On the one hand we were lucky because we got the diagnosis extremely quickly and we've got a friend who we met in the hospital who still doesn't have a diagnosis for her daughter, you know and that was back in April when we met her so we are very conscious of that. But the diagnosis is kind of this, 'you could have a perfectly healthy baby who will never, you will never experience anything in that child's life' or you could need a transplant within 12 months and at which point it was never specified but we always inferred, rightly or wrongly, there was a serious risk of not surviving and all that sort of stuff … But it was difficult I guess, and that was particularly around that period where it looked like it wasn't working
>
> Alicia: Yeah
>
> Alan: and we probably still didn't know at that point, though we were thinking about it, we still didn't actually know what not working meant.

4.3.2 Uncertainty and surgery

Surgery offered hope, but was associated with uncertainty as a positive outcome was not guaranteed. Parents felt very distressed when the infant required surgery, given the infant's young age and vulnerability. The Kasai procedure was performed before the age of 10 weeks for all infants who had Biliary Atresia, but all parents worried that their infant may need a liver transplant at some stage regardless of the diagnosis.

> Darren: The first operation, the Kasai procedure was like when she was five weeks, that was the best, like the hard part for us because like she's really

tiny and they have to perform that operation and she has to stay like, I think 12 days in the hospital?

Donna: Yeah

Darren: And then she got out. And after that, you know and that's the main thing that the doctor said, it will work 75 to 25, so 75 is it didn't work, so that's the hard part. From there you have to wait for like, and the doctor's always like updating us, like how the liver is deteriorating so quickly, so that's, that's, in between that time it's really hard for us. Yeah. Mmm.

Parents were extremely stressed if their infant had a liver transplant, due to the seriousness of the surgery and the ensuing experience of the Intensive Care Unit.

Winona: I think that that 12 hours of um, oh, hang on, how long was it? It was longer than 12 hours. Twenty hours from getting the phone call to say that 'we have a liver for Whitney' was the longest 20 hours of our life.

Warren: I found that the worst thing for me was more to do, you had the operation, I understood the risks of what the outcome was, the liver could fail, but I thought the worst part was the three days after because basically they weren't going to feed her and it was up to her where she was going to go from there. There was nothing we could do but she had to take her medication. They couldn't feed her because of the bowel movements. She didn't have any bowel movements.

For some infants, liver transplant was required urgently and the lack of an available donor organ resulted in a parent donating a portion of their own liver. Living-related donation meant that the other parent was distressed about the risks for their partner as well as for the infant.

Lucy: Well, before the transplant I was extremely upset and worried because Leonard and Luke were both having operations. That was a pretty awful time, before the transplant and on the day of the transplant. And then just

Luke: Yeah, likewise

Lucy: And then after, Luke, because his recovery was so bad, because of the surgery, and just worrying about Leonard pulling through because he had a few high blood pressures and they were giving him a lot of bad medicine to try and make him better and that was a bit scary in ICU, but yeah. We still worry now, but each day it's a little bit less because he's getting a bit stronger, but you still worry because he doesn't have an immune system so we're always, that's always there.

4.3.3 Uncertainty and infant illness morbidity

The infants suffered various physical symptoms or complications of the liver disease. Parents were sometimes unsure of the cause of the infant's distress and worried that the infant might be in pain.

Abigail: I think she's, I think she's uncomfortable. Her tummy's definitely getting bigger

I: Is it?

Abigail: So she's very, she can't find a comfortable position. She she can't sit up yet, so she's lying down all the time so, and her tummy's big so I don't think she can get comfortable. She's itchy. And she just scratches herself, like her ears are just full of blood all the time. Scratching.

Adam: And we wonder if she's in any pain, you know

Abigail: And we don't know whether she's in pain or not

Adam: We we don't know if there's, if it's painful for her or

Abigail: Yeah

Waiting for results of investigations was a difficult time for parents, who worried about abnormalities.

Gretel: Because I mean I know when they did the ultrasound there was a problem because they kept focussing on one bit and they called a doctor in and then they called someone else in and they wouldn't tell me anything because they can't, I know that, but I knew there was something wrong, so I've been sitting there for 2 months thinking, 'what the hell is going on, what's the problem, what, what did they find?' You know, because they wouldn't say anything and we had to wait till our appointment.

Gary: And they still don't know.

Gretel: And they still don't know, so I'm still going to sit there for another month probably thinking, 'what is going on', you know. I just pray it's not cancer, you know, that he doesn't have a little growth or something on his liver.

4.3.4 Uncertainty and hospitalisation

The experience of being in hospital resulted in feelings of loss of control for some parents. Parents were often uncertain of when procedures would occur or even how long the infant would need to stay in hospital.

Ben: It's like they say on that show, Your Life in Their Hands, there is no truer statement because everything in our lives is now in, we're both people who are used to being completely the masters of our own destiny and here, now we're just cogs in a machine. We get told, we don't even get advanced warning of when things are happening, you know, they'll walk into the ward

and say, ‘ah, you’re having a blood test now’, ‘oh, right’, ‘ah, you’re going down for a scan now’, ‘oh, right’. Do you know what I mean? So everything is just like, no future, no plan, no future, you know? [laughs] So that’s hard to deal with but it’s the way it has to be because it’s the only way the system can work

Brooke: You have to accept that that’s the way it is.

For some families, the uncertainty about the infant’s progress and changing plans for discharge from the hospital was difficult.

Ewan: It seems to take so long. You’re looking for this light at the end of the tunnel. And it just sort of seems to keep growing as you find more things wrong, or seem to have some kind of date or day, ‘you’re going home on this day’ and then she’d get a virus, or she’d get sick or there’s blood in the poos or there always seems to be something. So that day keeps getting, you can’t reach it.

4.3.5 Uncertainty on returning home after hospital

Many parents felt isolated when they returned home after a period of time in the hospital and were uncertain about managing the infant’s illness.

Winona: Initially it was quite stressful. I used to make phone calls to the gastroenterology people at the hospital. When we first came home we had to administer about 14 medicines to Whitney each day. There were times when like when she’d spit these medicines out, like how do we go about readministering these medicines, do we have to give them again or do we wait for the night session? We got so worried because almost on a daily basis Whitney would um, wouldn’t consume those medicines.

However, as time passed after the transplant the medication regimen was gradually simplified and parents became more confident.

> Adam: At first it was really hard
>
> Abigail: Yeah, it was
>
> Adam: because there was a list with about, you know, half a dozen, seven or eight, you know, this there and that
>
> Abigail: We had 10 a day when she came home, yeah.
>
> Adam: And now it's
>
> Abigail: Two, yeah. So it's not as, it's not as painstaking as what it, as what it was, which relieves a stress.

4.3.6 Uncertainty about the infant's ongoing health

The infants' health status was often unpredictable and could deteriorate quickly, resulting in families needing to remain close to a hospital in case the infant needed urgent treatment. Visiting relatives outside of major cities or going on holidays was therefore difficult. Donna and Darren were from an Asian country and worried that they wouldn't be able to visit their relatives there.

> Donna: It's just, it's just limited everything that we need to consider Debbie for everything that we do. Like
>
> Darren: Like going for holidays
>
> Donna: Like going for holidays, yeah
>
> Darren: So we can't really, like, straight on go to [their home country] or somewhere else because of her situation, like she might be needing in that time to be in hospital or something so.

Travel was a particular issue for parents whose infant was on the transplant waiting list, because they had to be close enough to the hospital at all times to be able to attend quickly if an organ became available.

> Abigail: We can't make any plans because we don't know. We're on the transplant list now, so we don't know when we're going to get the phone call. So we're hesitant to make plans … Well, you know, we might have liked to have gone up, we've got family up at [country town] or [another country town]
>
> I: Oh, I see
>
> Abigail: at Christmas time, so, yeah, we've put, we've canned that. Because that's too far, you know, to get home. We don't always, we can't always guarantee we're going to have phone service up there. You know, we'll certainly go and do day trips and that sort of thing, but you know, it would've been, might've been nice to have a couple of weeks away, sort of thing, and especially being her first Christmas.

Uncertainty about the infant's health led to changes in plans for Alicia and Alan, related to whether Alicia would return to work full-time and uncertainty for both parents about their future plans including whether they could move away from the city.

> Alicia: Yeah, where we think about living, like I think we'd like to move out of [city] but I don't think that's possible
>
> I: I see
>
> Alicia: And it affects, it kind of affects … I mean I don't think I would have been really wanting to be full time at work, but I don't think I can be now, so I don't know how much that's had an effect, but yeah. And I guess it affects how I think about the future. Just the uncertainty.

I: Uncertainty?

Alicia: At some point he'll need a transplant and that … yeah, I mean, it colours life really.

Alan: Yeah. And certainly, living outside [city] was a point I was going to raise.

4.3.7 Uncertainty and the future

The infant's health also led to worry for parents about their infant's future wellbeing. Ben talked about his worries for his daughter.

Ben: It's sad because you worry about the immediate future, the medium term future and the long term future for her, you know, like I always, I'm 6 foot 4, I was always very healthy my whole life and I always expected to have tall, healthy children and I worry about her growth being stunted. I mean, we're not, now that we've seen how she's turning out we're not too worried about her intellectual development. She's clearly very smart, but you just worry that it may, whether it be, whether it be the scar impacting on her as a teenage girl, you know, teenage girls can be mean, you know, will it cause her to have all sorts of body issues or something, you know, things like that.

Worrying about the future also encompassed concerns about other children in the family. Simone spoke about her worry for all of her children. Her infant had been well until the sudden onset of Autoimmune Hepatitis at the age of 21 months.

Simone: Yeah, I'm always making sure I look out for the signs and everything with her. If she's ever feeling unwell I sort of, oh, now she's old enough to say what it is and what's wrong and everything but I always have it in the back of my mind, or, with all the girls that if it, if it could have happened to her it could happen to them as well.

Charlotte and Clarke's infant had alpha-1 antitrypsin deficiency, a genetic illness. Charlotte was pregnant at the time of the second interview and spoke about her reasons to have the new baby tested after he was born.

> Charlotte: Obviously to know, so we can provide the, get the right support for the baby [inaudible] but also for my, I think, our sanity, so I'm not looking at the baby each day just thinking, 'are they getting yellower and yellower', because that was how it happened. Her eyes were yellow and yeah. I want to enjoy having a baby.

4.4 Infant Vulnerability

Infant vulnerability was foremost in parents' minds. Repeated medical and surgical procedures emphasised the infant's vulnerability, as did infant distress. The risk of infection was a reminder of infant vulnerability, especially if the infant was immunosuppressed post-transplant. Awareness of the infant's vulnerability led to feelings of helplessness if parents were unsure of the cause of infant distress. Some parents feared losing the infant and one family described mourning the loss of their healthy infant. Infant vulnerability raised feelings of guilt for some parents, especially when the cause of illness was genetic or unknown. Some infants became anxious or 'clingy', further reinforcing parental awareness of the infant's vulnerability. Seeing other sick children in the hospital similarly raised fears for parents about their own infant's vulnerability. Trust in the treating team lessened parent distress about infant vulnerability.

4.4.1 Infant vulnerability and medical procedures

Painful physical investigations and procedures were common throughout the infant's illness, and raised parent awareness of the infant's vulnerability. Infants often

became increasingly aware of when procedures were going to occur and were more distressed over time.

> Trish: She copes with, well, with blood tests she doesn't cope really good. She knows now … she's more aware. She watches everything, she knows. She knows them from the gloves, from everything, from the smell of the cream they put on her hand, she knows everything now. She cries.
>
> I: That's hard, too, for you as a parent
>
> Trish: Yes, she breaks my heart when I see her going through all this pain, yeah.

4.4.2 Infant vulnerability: symptoms of illness

Symptoms of the infant's illness were a sign of the infant's vulnerability. Parents were often unsure about what was causing their infant's distress and felt helpless in knowing how to soothe the infant.

> Abigail: Yeah, just her restlessness, it's very, you feel so helpless.
>
> Adam: She kind of looks at you like, 'help me'
>
> Abigail: 'Help me, help me'
>
> Adam: You know? These little eyes and that, that's pretty hard when you can't sort of, you know, you try for six hours to calm her down and nothing works.

The risk of infection was a reminder of the infant's vulnerability. Luke and Lucy discussed this issue.

> Luke: Yeah. Like, the show was here, not last weekend, the weekend before, or whenever it was, but we usually do that as a family thing with the kids but we couldn't this year because
>
> Lucy: We're just worried about Leonard

Luke: We're just worried about Leonard getting sick or someone coughing on him or him touching something and then him ending up back in hospital and yeah.

Some infants had severe symptoms of chronic liver failure, such as persistent itching, which was difficult for parents because they empathised with the infant's distress but were unable to soothe the infant.

Gretel: I just feel like sitting there and crying all the time, but you just can't

Gary: You can't cry because that defeats, you've got to keep going.

I: You're often feeling tearful, are you, Gretel?

Gretel: Yeah because it's so hard seeing him in pain and suffering and not being able to sleep properly because he can't, because he's itching or, I don't know what's sore on him, he really, you know. So it is. It's very, very hard.

4.4.3 Infant vulnerability: fear of loss

The infants' vulnerability raised fears of losing their child for some parents. The parents in six families spoke about fearing the loss of their infant.

Fiona: Emotionally, didn't think anyone could be as emotional as, as what we are.

I: What do you mean, Fiona?

Fiona: Just drained, like wondering each night are you going to wake up, whether the kid's going to wake up in the morning, yeah [cries]

Diane and David also discussed their fear of losing their infant.

Diane: Oh, you know, the reality is it's not a cure. We know that and we're grateful for where she is at this stage. We know that we just have to be on top of things and hope that her body accepts what's been done and the meds that

are helping her live. But emotionally, yeah, we sit there sometimes and think, 'will she be around forever?' you know, it is, you can't not think about those things but it's not a good feeling, so, yeah.

David: Mm. Oh, I don't think, I dunno, I don't think about, I don't think about it too much, but I just, there's those quiet moments where, where you're just by yourself or whatever and you just think to yourself, you know that it's not, as much as we, you get on with what we're doing and everything seems like it's okay, there's quieter moments where you're a little bit retrospective or, you know, and then you still know, it brings you back and says, 'you could lose her at any time'. So.

One mother, Hollie, also spoke of mourning the loss of her healthy child.

Hollie: I think also that um, it might be different for fathers, but I think um, you know, things, when you're pregnant and you have um a healthy pregnancy and you have, have all these hopes and dreams for your child and then you go through a period of mourning because [Hayley crying] you mourn the loss of a healthy child I think. So it's quite difficult. I find it difficult to look at pictures of myself pregnant with her. I don't want to look at stuff because it sort of, it sort of, you know, um, um, oh, excuse me [tearful], it's er and even now if friends are pregnant, going to baby showers for example, things like that are very difficult for me.

4.4.4 Infant vulnerability and parent guilt feelings

The infant's vulnerability also raised concerns for some parents who wondered whether they did anything to cause the illness. Guilt could be a particular issue for parents whose infant had a genetic disorder such as alpha-1 antitrypsin deficiency, or when the cause of the illness was uncertain. For Neil and Nicole, whose infant had

Biliary Atresia, the guilt feeling was related to the uncertainty of causation of this illness.

> Neil: You blame yourself, you know. You think what might we have done to cause this or, obviously, you know, you think about that. But I think knowing the, knowing the facts it makes it easier to deal with I guess, otherwise you just guess and [inaudible]. So I think that was very helpful. And here they did go into a little bit of the cause, but they didn't really, you know, when they were explaining it, all that discussion, but they didn't really
>
> Nicole: And there's still a lot unknown as well, there's still a lot of research, they don't really know how
>
> Neil: Sure
>
> Nicole: How exactly, you know there's an unknown [inaudible]
>
> Neil: Sure

Isaac suffered from alpha-1 antitrypsin deficiency, a genetic illness. His parents discussed their guilt feelings in relation to his illness.

> Imogen: Oh, I think, I think because it's sort of something we, we, I mean we understand that we, we're carriers and we didn't realise and sometimes we joke [laughs] and say, you know, we were either …
>
> Ian: I know I feel guilty about it
>
> Imogen: Do you?
>
> Ian: Yeah
>
> Imogen: I thought you were joking
>
> Ian: No

Imogen: I don't feel guilty because it's not something that I can control, but I didn't realise you felt guilty.

Ian: Oh, I would, I would generally feel it's my fault he's got it because, you know, I've got a defective gene, so, yeah. But there's not much that I can do about it.

4.4.5 Infant vulnerability and infant anxiety

Infant anxiety was a marker of the infant's vulnerability and parents worried about possible long-term effects of repeated painful experiences.

Abigail: I'm hoping that she's too young to, to remember it. Um, str-, look, strangers, I will say, emotionally, strangers come near her and she gets very, um, very clingy with Adam or I, very much so, or she'll, or she'll cry if she sees, you know, a strange person.

Adam: I'm sure a lot of babies, kids are like that

Abigail: Yeah

Adam: who've been through a traumatic event, are like that, aren't they?

I: Yeah. How old is she now?

Abigail: Nineteen months, yeah, nineteen months, yeah. So mostly I don't know. She's just too young. I'm hoping she's too young. We're both hoping that. Mm.

Jason spoke of the emotional impact on his infant of repeated painful procedures.

Jason: Yeah we think it may have made Jasmine more clingy, being, I mean for weeks, for most of her life up to a certain point she'd been surrounded by strangers who would hurt her, you know, with the cannulas and stuff. She had terrible times getting cannulated and scream and scream. We don't know, but

we think that that's made her less inclined to want to be held by other people and so on, so.

One infant developed more serious psychological issues.

Trish: Tina was so upset and stressed out that she started to vomit to get, to get everyone's attention. I'd say nearly six months with vomiting. Carrying the bucket all over the house, even vomiting in her sleep. Like when she was sleeping, she would vomit. And we ended up with gastroscopy for her, and nothing. And [the doctor] said, 'believe me, lots of kids vomit, 50% of kids vomit just to get your attention.' And I still didn't believe her till it happened to my daughter and they taught me how to deal with her and what to do to her. I went to the Speech Therapist again, like after the Psychiatric Clinic, and then she told me how to talk to her and how to deal with her as a normal, normal kid. She had too much attention, like more than the others.

I: So, has that vomiting settled down now?

Trish: It stopped completely.

4.4.6 Infant vulnerability: seeing other sick children

Seeing other sick children in the hospital or being aware of other children dying was often a reminder for parents of their infant's vulnerability.

Heath: What I was exposed to in ICU I think was a bit of a turning point for me. I know that liver transplant's severe, but I've seen how bad things can get. And having seen a couple of kids, one kid in particular passed me, they were in the cubicle across from us in ICU and he'd passed away. He would've been 9 or 10 and the nurses wheeled him out. He looked peaceful and asleep in his bed, but I just, I don't know, I don't know it's just

Hollie: You're exposed to a lot of things you don't want to see.

Heath: just the fragility of life I guess. And you think, 'what's next? Is it, is it, is it me? Or is it somebody else I know? Or?' Because all this stuff's fairly random. Biliary Atresia itself doesn't seem to have any pattern, it's just, it's a congenital defect and 'wham, bam, here you are, liver transplant'. And we kind of took the hard road to that.

Adam also talked about his distress at seeing other sick children in the hospital.

Adam: I don't feel comfortable in this environment at all. [child cries]

I: What is it about the environment here do you think?

Adam: Just the sickness here. Like I walk around and I see those little kids with leukaemia. It really affects me, I get, I just get really sad for them, just it's, it's, it's just horrible. I've never had exposure to this sort of thing before. Yeah, so the whole hospital thing's a real freak out for me.

4.4.7 Infant vulnerability: trusting the treating team

Surrendering control to the treating team required parents to trust the team with the infant's care. Interestingly, parent experiences of the team were extremely positive and many parents talked about feeling supported by the team. Being able to trust the team helped parents to deal with the infant's vulnerability.

Max: Sometimes I say to my family if we not stay here and we don't have, you know, [the hospital] and everything, maybe we cannot save Madeline, so that is, we thank … very much for the support from the hospital, from government and

Marie: We feel very appreciative for what the hospital

Max: Did for us

> Marie: Did for us, for Madeline. Even like [name], [the social worker] and [the psychologist], very supportive
>
> Max: We're very appreciative for that.

Parents turned to members of the team for advice about the infant's care.

> Elizabeth: I know, like, this week I was worried about this platelet thing, so I'd ring [name], you know, the transplant coordinator. So, you know, she, [name] and [name, other transplant coordinator] are a big support and the nurses here are absolutely amazing. I know that if I had a concern I could ring them up with no worries at all, you know.

Nicole and Neil also talked about the importance of having confidence in the treating team.

> Nicole: And there were other things as well. I mean, the staff here are very good, you know,
>
> Neil: Yeah, yeah
>
> Nicole: Everyone involved. You felt really that they, obviously they knew what they were doing and that gives you a bit of reassurance. And the nurses as well, they were all really friendly, very good, very helpful, so that, so obviously you don't feel too, too on your own.

4.5 Isolation

Isolation was another theme common to many parents' discussion of their experience. Lengthy periods of time in hospital resulted in parental separation from usual supports, especially for families who lived interstate or in rural areas. Returning home from the hospital was also associated with isolation as parents took on responsibility for the infant's medical care. Caring for the infant's medical needs in

addition to usual parenting responsibilities left little time for social contact. If the infant was at risk of infection, social contact had to be limited, leading to further isolation. Lack of understanding from other people added to parents' sense of isolation. Isolation was a source of distress, but was mitigated by the availability of social support.

4.5.1 Isolation during the hospital admission

Periods of hospitalisation were often isolating for families when parents were separated from family and other supports. Abigail and Adam spoke of the difficulties of separation for the family.

> I: Are there any other ways it's affected you as a family?
>
> Abigail: Just being separated all the time with the hospital visits, don't you think?
>
> Adam: Mm
>
> Abigail: They're really tough. We're really good when we're together, 'cause we draw strength from each other. But when we're separated it's really tough.
>
> I: Yeah.
>
> Abigail: Because he's trying to go to work and trying to keep some sort of normality for [older sister] and I'm in hospital going through everything with Amelia. And there's only so much of the blood tests and the tests and everything that I can see her take before, you know, I get to cracking point.

Some mothers talked about receiving distressing or complex information and having to relay this to fathers later.

> Winona: Sometimes I'll have meetings with the doctors and then I'll call Warren straight after that meeting and tell him what, exactly what's happened

and he'll direct questions at me, 'oh, have you asked about this?' and I, it's just gone straight past my head, about asking a specific question and he's, you know, thought of something and then I'm like, 'okay'

I: So what's that like?

Winona: So then the next time I see those doctors I'll ask them those questions, but it's hard. It'd be great if both of us were there, but it, obviously with him working it's difficult.

Isolation was particularly difficult for families who lived in rural areas as they became dislocated from their usual supports. Ewan spoke of the problems of separation for his family. He and Elizabeth decided to stay at the hospital with their two younger sons and their infant. However, their eldest son was close to finishing High School and remained in the family home.

Ewan: Well, I suppose the two younger boys, they've been taken out of their environment, well, we've all been taken out, us four, five have been taken out of our normal environment. And so they're not playing with their friends and stuff neither, or going to their own school. Fortunately they've got a school in this hospital that they've been able to go to and forge new friendships and stuff. And for the older boy, he's, I suppose he's just got on with what he does, but he hasn't had that input, that parent input as much. Contact on the phone and stuff, but yeah. And so it's a huge upheaval.

4.5.2 Isolation after returning home from hospital admission

Many infants spent lengthy periods of time in hospital. While parents were keen to go home from the hospital, adjusting to being away from the hospital was a difficult experience for some, often associated with feelings of isolation. Ewan and Elizabeth described their ongoing distress at the second interview.

I: So, that adjustment to moving back home after such a long period of time, what was the emotional impact of all of that?

Ewan: I'd say it was quite overwhelming, I guess it still is some days

Elizabeth: For me, I would say I pretty much had a breakdown because going from being here and having such a support base, you know, from nurses, doctors, cleaners, the staff in the cafe that you'd see all the time, other mums and everything, to all of a sudden, and Ewan being at work and me being, the kids being at school, and me being there on my own with the two kids. So all of a sudden, I'd gone from being here sixteen months straight to all of a sudden being home and then deciding whether I'd go back to work and things like that, I found. And socially I didn't want people over because I didn't want them to bring the germs into the house, but I was so lonely as well, so it was such a worrying time that I, yeah, I pretty much had a breakdown.

Ewan's perspective was different to Elizabeth's, which he related to being able to work.

Ewan: I was definitely looking forward to moving back home. I probably didn't have as quite, I didn't have as, the trouble that Elizabeth had because I managed to continue to go to work … So for me going home, returning home wasn't a great, it was definitely a plus and I didn't struggle with that. I did struggle, like the overwhelming feeling of so much to do at home and you probably had the, seeing Elizabeth struggle with it and struggling with getting enough sleep with her sleeping patterns, being out of the hospital, where before you, we could go to the lodge and sleep and if she [their infant] was up all night playing up, well the nurses were there so that that definitely helped. To go home, to go home and not have that, we definitely noticed that difference. Yeah, so that's. But yeah, it's fairly isolating I suppose in respect

to, you didn't have the support of the doctors and nurses, so if she played up all night, well you just have to cope with that, so I guess that just added to the workload.

Abigail also spoke about returning home post-transplant and her anxiety about managing the infant's medications. She suggested one way that the treating team could help to lessen parents' isolation when they returned home.

Abigail: I was very nervous to have her at home after the transplant. Um, maybe a weekly, maybe a weekly phone call from a nurse to see how you're going, how she's settling in, how's everybody else going, sort of thing, because I guess I felt I walked around on eggshells for just a couple of weeks afterwards, just while we got the medicines and knowing she, especially because she was so highly suppressed we weren't allowed to leave home for six weeks, we had to keep her at home for six weeks, so maybe, and apart from the clinic visit, which is all just the health and blood tests and that in the visit, an after care phone call or something like that probably would have done the world of good, I think … Because you do feel very isolated.

4.5.3 Isolation due to time-consuming illness management

Many parents reported that they had little time for social contact, given the demands of looking after the infant's medical needs in addition to usual parenting responsibilities. For example, Richard talked about how the infant's illness had affected the family's social relationships:

Richard: Socially, well it's limited us socially to what we, how we used to live our life to how we are now, because it's full time looking after him.

I: Full time looking after him?

Richard: Mm, so. You don't get a lot of social activities and when we do, it's more like family orientated. So, yeah.

Charlie also spoke of how time-consuming managing his infant's illness was, reducing his opportunities to socialise.

Charlie: Yeah. Really in the last 12 months it's every, I've got a group of friends that I used to see three or four times a week, I see maybe once or twice a month

I: Right, quite a big difference

Christine: Mm

Charlie: Because it would be, you know, when [brother] went to bed at 7, 7.30, there was all that time afterwards. Whereas with her, with getting things ready for day care, getting meds done, getting her settled it's 9 o'clock most nights before we're ready to sit down and it's just too late to go out again.

4.5.4 Isolation due to the infant's risk of infection

Infants who had had a transplant were immunosuppressed and therefore at high risk of infection. Some parents became isolated as they avoided going out due to the fear of infection.

Elizabeth: Socially, socially I find that I tend not to want to go anywhere because of illnesses and other people being sick. If you go to the shops, you know, you've got all the germs there and I'll tend not to, if I'm going to go somewhere it'll be times when I'm not going to take the kids so I'll go or Ewan will go. It won't be all of us go unless it's to immediate family, or friends that really understand, you know. And I'll make sure that I find out if anyone's been sick or anything first, where before I wouldn't have worried about that sort of thing.

Fear of infection meant that many parents were also careful about spending time with other people either at home or outside.

> Shannon: With her immune system being down
>
> Simone: Swimming lessons she wasn't allowed to do because germs again, so we sort of stayed home and whoever we did let come into the house to visit
>
> Shannon: They needed to
>
> Simone: we needed to make sure that they couldn't be sick or, to risk anything getting worse.
>
> Shannon: Simone and the kids didn't come to a football game for ages after, to watch me play.

4.5.5 Isolation: friendships changing

Some families spoke about friendships changing, leaving parents feeling isolated. Friendships changed due to lack of time or because friends didn't understand the experiences that parents were going through. Victor and Valerie were young parents whose friends did not have children. They noted that their experience with their infant's illness had brought them closer together as a couple, but that they had stopped seeing their friends partly due to lack of time.

> Victor: Yes, we're definitely closer and [sigh] sort of don't have time for, for people who are selfish. Like, we've had quite a few friends which we don't have anything to do with any more, just because they are all about themselves. And I think going through what we're going through, I think we find it hard because people our age don't usually have kids, so again they don't know what we've been going through having kids and

I: I forget how old you both are

Victor: 21

Valerie: 21, yeah.

Victor: So, but it's sort of aged us a bit and we don't sort of get along with people our own age and also we're sort of [Vivienne crying, Valerie takes Vivienne out of the room]. Yeah, we've sort of learnt to rely on ourselves rather than depend on friends and even family … We still hung out with all of our friends when Vivienne was first born. Once she started getting sick it was harder and then when she got really bad we sort of didn't have time for anyone really.

Elizabeth and Ewan also spoke of how their friendships had changed over the period of their infant's illness.

Elizabeth: Yeah, I think at the start of your, when your child is first ill, she was first born and then she got sick. I think people were just in your face and they just, everyone wanted to know, the phone would be ringing constantly, you'd get texts day in, day out, and everyone wanted to know how everything was and that was all a bit overbearing and now I think it's down the track, sort of 12 months down the track and it's died off. People aren't so interested in it any more and yeah.

I: What's your perspective on it all, Ewan?

Ewan: People, I think people just think 'that's not fixed yet', they had this expectation that it's been enough time, that it should've been fixed, but they have no understanding of what, of what you're going through and how long it takes. It seems to take so long … But socially, well you, there's not a lot socially because this takes so much time. This takes almost all your time.

Lack of understanding from other people was also important for some parents.

> Barbara: I don't know. I guess at the end of the day no one, a lot of people don't really understand what I'm going through or what he goes through. Yeah. Sometimes at the end of the day I just say 'yeah, he's been good', just to keep everyone happy. Because they don't understand. I go, 'yep, he's been good, he's been sleeping', but, yeah. Because even when you tell them the bad, we don't get help anyway.

One family spoke about friends not understanding that smoking around their infant could have serious health implications for her due to her alpha-1 antitrypsin deficiency, which can have lung complications. Charlotte and Clarke had changed their friendships as a result.

> Charlotte: But we don't, a lot of our other friends, it has changed, you know we won't go round to see people's houses because they smoke cigarettes
>
> Clarke: Yeah
>
> Charlotte: So that socially we've had to make some decisions.
>
> Clarke: And we've also been disappointed with those friends that they haven't really taken an interest in Caitlyn's condition, so in that sense it's affected the friendships that we had, and it's brought us closer to people who have shown an interest and have shown consideration, and maybe driven us away from people who have perhaps not been so considerate.

4.5.6 Isolation and social support

Lack of social support created feelings of isolation for some families. For some parents, extended family members lived at a distance, were facing their own health problems, or were emotionally unavailable or unreliable.

Elizabeth: I don't think Ewan's mum really understands what we go through and my mum and dad are really just too far away because they live on the south side of [city] and dad works two jobs and mum doesn't drive so it's not like we can catch up and stuff, so. Yeah.

I: And no brothers or sisters?

Elizabeth: No. No, well Ewan's got a brother

Ewan: I've got a brother, five years younger. He's got Schizophrenia, so he's no real, he's no support.

I: Is he in [city]?

Ewan: He's in [country town]. So he's not that far from us, but we don't have that, hardly any contact with him.

I: But you haven't got any family members who you can rely on?

Elizabeth: No

Ewan: No, which definitely makes it harder.

One family preferred not to talk with the extended family about the infant's illness, reducing the opportunity for extended family members to offer support.

I: Okay. But now, what about supports for the family? Just kind of normal supports. You mentioned your parents live close by to here, Karen.

Karen: Yeah, yeah. Well, I don't tell them a lot of things about

Kane: Kristy

Karen: how Kristy's, well I tell them about her development and growth like. The problem, I don't actually tell them because I just don't want these things to spread around the family.

I: No? Okay

Karen: Because when you tell them something else, it probably spreads to another person in a different story. It just makes things worse and I take it very serious when, when words come back to my ear. So I better not to make things worse, so I better not to say anything, like any bad things or problems.

Having good family support, however, helped parents to feel less isolated. Max spoke about grandparents visiting from overseas, helping him to feel more connected to his family.

Max: Yeah. We get a lot of support from family because when we, when Madeline has a problem my mother-in-law and my father-in-law and my parents come to support us all the time and we feel very happy when we have their support and we feel more confident, we don't feel isolated from you know, our family in [country] so that's the good things.

Likewise, Janice spoke of the importance of family and friends.

Janice: And the extended family, I have to say our family and friends were really supportive, which I think makes a difference. You know, I'd see like, we saw some people here in the hospital, single mothers perhaps who may not have close family around and it would be really really difficult. I feel very grateful to have that support.

4.6 Dealing With Other Aspects of Life

Dealing with other aspects of life was also important for parents. Financial burden was a concern due to the cost of managing the infant's illness. Hospital admission also added costs, such as car parking at the hospital or having to buy meals instead of cooking. Work pressure resulted from fathers taking time off work or from their own business, and mothers being unexpectedly unable to return to work after maternity

leave. If there were other children in the family, parents were often split between the needs of the other children and the infant. Dealing with other life pressures was helped by the availability of social support, particularly from extended family members.

4.6.1 Dealing with other aspects of life: finances

The illness added financial pressures for several reasons. Parents often reported high costs of transportation as they made more trips than usual to attend the hospital or outpatient clinics. Being at the hospital also meant that meals had to be purchased, which was more expensive than cooking.

> Winona: I think the main impact is financial stress. Um, I've just been on m-, I'm still on maternity leave, but that's the, okay, we're spending time in hospital it just makes it hard to, being in hospital and if I have to get out and about, just the life here can be expensive and just relying on one income as well while you're trying to look after a child, it's very hard.
>
> Warren: Yeah, I agree with that because um, you know, normally our pattern, even when we were both working, you'd only really go out once, on a Saturday or once for breakfast in the morning, or you know, once, once a week during the night or something. Here, you've got the expense of parking always, even though it's discounted it does build up. I think I've exceeded over $3000 in parking now, just from coming in every night, even though it's discounted, it's at a discounted rate. And you look at the other things, like food, you've got to go to [fast food outlet], the selection isn't that great here, but it's, you've got, for me I've got to go to [fast food outlet]. All of a sudden you're spending 20 or 30 dollars that you wouldn't spend every single night buying food. You know, things like that.

Winona: You don't have time to go home, time for cooking.

Warren: All of these things add up. All of a sudden you've got all these little expenses, um, I take, for instance, you know, to save myself going home, I take the toll charge to go home, only because if I don't it takes 40 minutes. I'm already tired, and I think 'well I may as well go the fast way home' but then there's the toll charges and all of a sudden I'm building up toll charges ... The other choice is I drive home normally and then I'm half asleep already and I'm more exhausted. I'm trying to time manage my sleep as well.

Some parents were unaware of available government support. For example, Hollie and Heath talked about having found out from other parents about financial supports that were available to them.

Heath: And they [other parents] tell you where to get all these refunds and rebates, more so than

I: So that's the kind of network that helps you out

Heath: You get some packages and things that give you some avenues, but it's difficult enough to, to navigate yourself through Centrelink websites and government bureaucracy to get these things done because they make you jump, jump through all these hoops. If somebody can at least tell you where to go, then that's half the battle.

I: Yeah.

Heath: Um, so. And we probably missed out on, on um saving ourselves some money and some funds

Hollie: Yeah, we have, I was so annoyed when I found out because we could've done that last year when we had the pump and you can, apparently there's a medical rebate that you can get, which we just found out and we

were like, 'damn! Wish we'd known that last year', you know. So, I'm just annoyed. I think, why doesn't someone tell you this stuff? But I guess people don't know until they go through it.

4.6.2 Dealing with other aspects of life: work

Some fathers noted that they had to keep working to cope with the increased financial pressures of the illness or because they feared losing their employment if they took long periods of time off work. For families who had self-employed fathers, family finances suffered if they took time off work due to the infant's illness.

I: Have there been any other ways in which having a sick child has affected your family?

Lucy: Well, just financially it was for a while, because of Luke being off work. Having to take time off, and lose business and travelling down to [city] all the time, just costs like that.

Adam worried that his employer would think he wasn't needed at work if he took too much time off.

Adam: And I don't want to kind of do myself out of a job. Like, you know, if they're pulling the weight then maybe the big bosses will go, 'well, do we need this guy?' If the other guys can do the workload, like, you know, [Amelia cries] 'should we just get rid of him?'

Some fathers, however, reported having very supportive workplaces with bosses who facilitated them taking as much time off as they needed. Gary reported that his boss had been very supportive and that taking time off work was not a problem.

I: So, have there been any other effects on your family, having a sick child, would you say?

Gretel: Stress, financial, what else is there?

Gary: Yeah, I don't let that get down to me, get me too bad, I try as hard as I can to support the needs.

I: What's the cause of that? Are you having to take a lot of time off, Gary?

Gary: No I'm lucky the boss has been really good about that sort of stuff and all these visits are taken out of my compassionate leave, rather than my holidays, so I have been accruing holidays.

I: That's good

Gary: But if it comes to the point where he [Gordon] has to go into hospital ... So yeah, he's been really really really supportive about all that sort of stuff

Work was positive for some fathers in terms of the distraction it gave them from thinking about their infant's illness.

I: Okay. You were saying that it's helpful for you to be out at work, in some ways.

Peter: Yeah, because I have to, I get to mingle with people, that's what I was trying to tell you. I get to mingle with, because I'm working. I have to move with people. So, as long as I'm in the work I couldn't think much about what is happening here. So, that way I'm getting rid of things.

I: Yeah, it takes your stress away, you mean?

Peter: That's it. Yes, so.

By the time of the follow up study period, many mothers had not been able to return to work as they had planned prior to the infant's birth, due to the infant continuing to be unwell and requiring a high level of care. The parents therefore had to adjust to

having a lower income for a longer period than anticipated. For example Elizabeth spoke about financial difficulties as a result of her not being able to return to work.

> Elizabeth: I find, I'm finding it a bit overwhelming at the moment because we seem to have so many bills that keep coming and we spent so much time in here without me b-, like I was only meant to have 3 months off work, and then ended up being, having 2 years, almost, off work. And, so that money we didn't have. And you know, we've got, had to use credit cards while we were in here and stuff so now we've got those bills to pay as well as everything else. So I'm finding that, like I have to go to work. You know, it's, and I'm only, next week because I'm older than all the other people there, then you know, you don't work Anzac day because it costs them too much [laughs] so I miss out on those things and stuff like that but yeah. But then I guess on the other hand I think, 'oh, well, money's not everything'. Because what can you do? If you can't pay it you can't pay it [laughs]. Yeah, that's right. But it's a bit of a struggle with, you know, with mortgage and bills and they just keep coming.

4.6.3 Dealing with other aspects of life: keeping the family going

The infant's illness resulted in parents spending a lot of time looking after the infant, making it harder to keep the family going. Lucy spoke of the difficulties keeping everything going for the family.

> Lucy: Oh, um, just Luke and I spending time together, we don't do as much. I feel bad because I'm trying to be 3 people. Trying to be a wife and friend to Luke and trying to be a mother to the girls and Leonard and I find it hard to split myself 3 ways. So I'm trying to do that and I find that that sort of gets me down sometimes. It gets me upset that I can't also just do things like dusting the house and getting the washing up done, just those sort of things.

Penny described the impact on her older daughter of having to spend time looking after her sick infant.

> I: Are there any other ways in which having a sick baby has affected your family?
>
> Penny: Yeah, especially her [sister]. I couldn't give much time to her because. There is a 10 years gap between them, so all along this nine and a half years it was taking up most care to her. Now, but she's growing up now, so she's doing her work on her own nowadays, slowly she's started doing it. So, but anyway when her studies is considered, I'm not giving much time to her. She suffers a lot because of that. Yeah.
>
> Peter: Yeah.
>
> Penny: I couldn't sit for half an hour together with her. More than half hour I couldn't sit with her. Because I have to, I tell her that 'you have to learn everything at school and try to learn on your own'.
>
> I: How do you think that's affected [sister]?
>
> Penny: She is now a bit poor in her studies.

Extended family members were important for providing support to the family. Parents relied on grandparents to look after the infant's siblings and to provide practical support such as making sure that bills were paid or the gardening was done.

> Simone: But it showed you a lot more, probably appreciation for my parents for how much they
>
> Shannon: They do
>
> Simone: they do and helped out with anything, like mail, or the little things that you don't realise they were doing and we weren't even realising it until

you're home, you're back doing it again and thinking 'oh, they've been doing all this' and

I: So they took care of everything?

Simone: They did everything so we knew when we were up there we didn't have to worry about

Shannon: Worry about it

Simone: anything being done, like if there was a bill that was due, it was paid and we didn't know any different till afterwards and it was good to have that stress of home life taken off us and could concentrate on Suzie, which was good.

David and Diane also spoke about the helpfulness of having grandparents look after their older son.

David: So what we'll do is my parents live next door to where we work

I: Oh, okay

David: So I'll get him up at six in the morning, I'll take him, take him there, mum'll give him some breakfast and I'm always around

Diane: It gives me a bit of relief

David: because I can just go next door, in and out, but he's cool.

I: That's great

David: He's cool and then I bring him home at 6.30, 7. Which gives her a day.

Diane: It gives me time, just to clean or vacuum, you know

4.7 Shared Experience

Shared experience was an important protective factor for parents. Most parents reported that they were closer as a family due to experiencing the infant's illness together. Meeting other families who also had sick children was reassuring for parents and offered hope in addition to the opportunity to 'give something back'. Shared experience therefore reduced the sense of helplessness and assisted parent resilience. However, shared experience also sometimes contributed to parental distress when other families' children had adverse health outcomes.

4.7.1 Shared experience in the family

Many parents said that the illness had brought them closer as a family as a result of facing the infant's illness together and providing emotional support to each other. Lengthy hospital admissions, or distance from the hospital, often resulted in parents being separated from each other or from the infant's siblings, which, for some families, emphasised the value of their relationship.

> Abigail: And look I think our relationship, Adam and my relationship, has strengthened too, with what we've been through. I, it was horrible in hospital all the time without him and without [sister] and he felt the same, you know, being at home with [sister] and not being with us together. And I think we just realised how much, how much we do need each other to, to keep each other grounded and supported and that sort of thing, well that's how I, I don't know if you, but that's how I feel.
>
> Adam: Definitely, it's a good friendship
>
> Abigail: Yeah
>
> Adam: Which is other than, it's something that's evolved other than the romantic kind of thing, isn't it?

Abigail: Yeah. Definitely.

Adam: We've been through this thing together.

Neil and Nicole also spoke about the infant's illness bringing them closer together as a couple.

Neil: Yeah, I mean it brought us together and we learnt to talk about things a lot more

Nicole: We talk about it, and been worried, but I guess initially we were both just there worried, because, no one knows what's happening to you. Nothing you're expecting. But as we understood more

Neil: I think we worked really well as a team.

Extended family members often also became closer as a result of the infant's illness.

Alan: I think we mentioned this last time, I suspect we did, but certainly in those early, you know, the first 6 months or so, probably the big change was Alicia and my mum got a lot closer

Alicia: Yeah

Alan: Because they really, a few times when we needed them they really came, literally came up a week at a time and you know I remember one time when we asked, we were at, pretty much at crisis point, you know I'd just burst into tears on the phone and mum said 'alright' and

Alicia: She'd be on the next plane

Alan: She actually got on a plane

Alicia: That night

Alan: That night or the next morning and flew over, like she was on the plane the next day or something, and that was, that was a, like Alicia got on fine with my parents but it really changed that relationship

Alicia: Yeah

One infant's cousin had been diagnosed with a serious physical illness at the same time as the infant was diagnosed with liver disease. Ben talked about how this shared experience had brought him and his sister closer together.

Ben: Well, look, with me, my, this year it's been a really bad year for us because my sister's, my nephew [name], who is 15 was diagnosed with something called multiple endocrine neoplasia this year, which, do you know what that is? Yeah … and now he's got cancer, multiple cancers through his endocrine system and probably won't last for much longer. And I think having sick children has definitely brought my sister and I closer together and their family, because they live in [city] and so since we've come back there, we've seen them a lot more than we had previously, as I say, simply because we've both got very very sick children and that's been, you know, that's made it a lot harder too.

4.7.2 Shared experience with other families with sick children

Shared experience also created a bond with other families who were going through or had gone through a similar experience, either with a child with serious liver disease or transplantation, or with other families who had sick children. Parents took hope from others who had had similar experiences.

Diane: And then you hear good stories

David: Mm

Diane: The longest liver is still going at 26 years … so that's great, so

David: Mm

Diane: And we've spoken to one mother in particular

David: Mm

Diane: that we befriended through the friend of a friend and her son's thirteen and is like any other normal kid, getting in trouble at school, you know [laughs] the normal things. I'm like, 'great! That's good!' yeah.

Talking with other parents who had children with serious liver disease was supportive for some parents because of the sense of shared experience.

Winona: [The team] has introduced us to other parents of children with Biliary Atresia who have had liver transplants so we can hear stories and help each other out and I found that helps me, being the ultimate carer for Whitney most of the time, spending most of my time looking after Whitney while she's in hospital, while Warren's at work, um, finding it a little bit easier knowing I'm not the only one going through this by myself, you know, knowing what I'm doing other parents are doing the same, of a similar nature, and we're going through this together.

Some parents were able to provide support to other parents in the hospital and took value from this experience.

Odette: But I found that I was able to help other people in the hospital by talking about it. Like all the other new parents who came in with the babies, they tended to put them in the same room as me, because of the problems that I had with Olivia, what Olivia had been through. They put those mums and babies in with us and I spoke to them and I was helping, apparently I was helping them, so I've been told, you know. It was good for me to be able to talk to somebody that had a similar problem, or a baby with a liver problem,

> not the same problem but a liver problem, because it, I found a few of my friends didn't get it, and unless you've got that sick baby, they don't understand, they're like, 'oh yeah, yeah, they'll be ok', but unless you talk to another parent with the same problem, that's when they get it, and you get it and you feel like you have that bond, which I have with another mother.

However, Elizabeth talked of the problems when children with the same illness died.

> Elizabeth: And in that time we'd also watched other kids die from the same disease Ebony had and we were struggling with that as well, so.
>
> I: Did you know many other kids who died?
>
> Elizabeth: We ended up I think we knew
>
> Ewan: We knew four
>
> Elizabeth: Four kids that died of the same thing. One family we became really really close to, so we spent a lot of time here with them, so yeah, it was pretty hard.

4.8 Adjustment

Adjustment was also a feature of parent experience throughout the period of the infant's illness. The birth of a new child is a period of adjustment for all families. Diagnosis of serious illness in infancy required further adjustment, particularly because the illness was usually unexpected. Some parents experienced emotional shock in response to the infant's diagnosis or as a result of other illness experiences. Anniversaries and other reminders of the illness sometimes threatened parent adjustment. Some parents described their infant's adjustment in terms of the infant's distress as well as infant resilience. Many parents' adjustment improved over time with developing confidence in managing the infant's illness. Parents spoke of the

need to accept what was happening to their infant and of having a different perspective on life as a result of the infant's illness.

4.8.1 Adjustment in family relationships

Some parents talked about family relationships already having been in a period of change due to the birth of the new baby. The infant's illness added complexity to the adjustment and it was sometimes difficult for parents to know how much of the adjustment was due to the illness and how much was due to adjusting to the infant's birth.

> Brooke: Yeah, I think the interesting thing for us is because she was diagnosed when she was only five days old, so when you talk about a fam-, as a family, we weren't even a family, really, so for us her being ill is, is being the family
>
> Ben: Yeah
>
> Brooke: and that's the norm.
>
> I: Yes
>
> Brooke: So, you know, it's difficult to kind of say what was it like before versus what it was like after because it's always been
>
> Ben: There wasn't really a before
>
> Brooke: Yeah.

Luke spoke about the benefits of spending more time looking after the other children because Lucy had to look after their sick infant.

> Luke: I probably didn't spend enough time with my girls that I would like to because of my workload but now it's sort of, the advantage of me having to be there, I'm closer to the girls and doing more with them and seeing them do

different things, which normally I wouldn't have because I would've been working or not doing that because Lucy would've been.

Lucy: Yeah

Luke: So I was. Yeah, that's the only advantage I see out of this whole thing is I sort of spend a bit more time with my girls and they, I'm closer to them that way, because of what's happened and I'm doing more with them and dropping them to stuff, dropping them places, doing things, getting them ready.

Fathers were often engaged emotionally and practically in the care of the infant and the rest of the family. Abigail and Adam talked about Adam helping with the infant's medications.

Abigail: He does a lot

Adam: I do the meds and all that stuff

Abigail: He does a lot

Adam: Not as efficiently as you do

Abigail: That's alright, nobody does it like that, darling.

Other fathers saw their role as more supportive of the mother.

Jason: And we've implemented some practices at home, where I'll take the baby in the morning to allow Janice to have a couple of hours extra sleep and that kind of thing, so that helps, doesn't it?

Janice: Mm.

Jason: Even just that extra hour or two makes the difference.

Sibling relationships can be strained due to the extra attention that parents need to give to the sick infant. For example, some siblings complained to their parents that they didn't feel loved any more.

> Barry: Yeah, I think sometimes he doesn't understand. His brother gets a bit more attention than what he does. But he is good about it. He has his little hissy fit and then he gets over it. I'm sure we have to spend a little bit more time with Blake than we'd like but I think someone understands.
>
> Brother: Yeah.
>
> Barbara: We had one issue a while ago, you know, 'do you love me any more?' Blake was going through a really bad stage.

4.8.2 Adjustment to the new diagnosis

The diagnosis and its implications came as a shock to many parents, particularly because the illness was usually unexpected. Rhonda described her distress after her infant was first diagnosed.

> Rhonda: Yeah, it's been really hard for me
>
> Richard: Especially for Rhonda, yeah, especially in the first 2 months I suppose.
>
> Rhonda: I think I cried for the whole first 2 months, virtually. Anyone came near me. The poor ladies who come around to give the babies cuddles while you go off for a shower, I screamed at them, told them 'don't bother, don't come back, we'll be here for him thank you' [laughs]

For some parents, the level of distress appeared to be particularly severe. For example, Odette talked about being unable to speak, being tearful and not being able to take in information after the baby was diagnosed.

Odette: For the first few weeks I couldn't speak about it with anybody. I could send a text to my friends, but I couldn't talk.

I: Why was that?

Odette: Why I couldn't? I couldn't stop crying. Because I'd go into the hospital and I'd stand near her cot and I'd cry. That was when she was in ICU, I just, cause you just didn't know what was going to go on during the day, or, you know, the blood tests that they put her through, and they couldn't find veins and she'd be screaming and crying and you know, just to look at her, all the tubes coming out of her, and not knowing whether she had this disease that was going to, that they wouldn't be able to help her with, all of that. It was hard, it was heartbreaking to see, you know, your little baby go through that. And especially when it was a problem that you didn't know she was going to have, that just sort of, on day three … Yeah, I couldn't, because for the first few weeks I didn't get any sleep [inaudible]. I didn't understand anything that they were saying, I didn't get it. They had to tell me several times for me to understand it. Orlando understood absolutely everything the doctors said, and he got it.

Adjusting to the infant's diagnosis was sometimes made harder by telling the story to other people repeatedly.

Gary: It's like when we found out that Gordon was crook, and just telling people over and over and over and over. And like everyone, like a lot of customers at work found out because I was gone for so long, and it was just, well it was starting to get me down, like having to repeat it over and over and over and over and over and over and over.

I: Yeah

Gretel: You don't want to just keep reliving it, really, and that's what you do.

Gary: You just want to move on, just take it every day, one at a time.

4.8.3 Adjustment over time

At the second interview, some parents reported more distress than previously and reminders about the illness or of distressing events were particularly difficult. Abigail was interviewed 6 months after the infant's diagnosis and described the emotional effects on her.

> Abigail: I know that I could just hear about somebody talking about somebody being sick, or have a sick child or, or hear, hear a song, or have a conversation or something like that and I can feel my eyes well up and, you know, get the feeling in your throat and that sort of thing, which never used to happen to me before. So emotionally, that way, that's how it's affected me.

Ben described how he felt he had coped well during the infant's illness, but now things had deteriorated and he described symptoms of depression. He related his distress to the anniversary of a procedure which had a serious complication for his child, which was also linked with the anniversary of another child's death at the hospital.

> Ben: … but also, excuse me, around [clears throat] around the same time is the anniversary of when, after Becky's transplant she was doing quite well and down in the ward she um, oh, really for no reason, and that's the thing that really bugged me, is that she'd been on intravenous antibiotics for a long time and they, they were worried about the veins, you know, about the veins giving out because she was constantly on the antibiotics, although they hadn't given out, but because that was a concern they decided to put in a PICC line, like a direct line in. And um, they, the, the, the doctor bungled it and um he

pierced her chest cavity um, but it happened, even though they do it under, you know, in, under X-ray, um, the, her clavicle was hiding the tube as they were feeding it up the vein and it pierced out, it went out of the vein, it pierced the sheathing of the vein and got into her chest cavity and for two days they were pumping her chest cavity full of antibiotics and um

Brooke: And fluids, IV fluids

Ben: and IV fluids and she was screaming and I, and we knew something was wrong and we knew that, and I knew that it had gone wrong when they did this PICC line, yeah, and anyway they kept telling me nothing was wrong, nothing was wrong and then finally it got to a critical point and it was, it's the anniversary this week of that as well, isn't it? Or last?

Brooke: Er, in about two weeks.

Ben: In about, yeah, so, you know what I mean, I just remember that it happened immediately after [another liver transplant patient's name] died and um, in the same hospital, during the period of the hospital stay that I was here, and she nearly died then as well, so you know what I mean, there's a lot of, a lot of pretty awful anniversaries that come staggered two or three weeks apart, so.

Parents used various strategies to adjust over time. Seeking information was one way of coping with the infant's illness, even if the knowledge gained was frightening.

Nicole: And then for us it was I think you, you know, trying to understand what the problem was at the time

Neil: Yeah, the physiology of it, the biochemistry, all the things that were happening, so we tried to focus on that as a way of understanding it, you know, because otherwise it can be a real scary, scary, nasty thing, you know,

you, you, you read this description and you read some internet forums and you can, you can, you read about the worst, but I think … but still the statistics are, are, you can't argue with them, you know, and they are quite grim, to be suddenly faced with those sorts of statistics when you've got a new baby, you know. So that was hard.

Some parents coped in different ways from each other.

I: So, Imogen, when you're looking up on the internet and so on to do research, is that a helpful thing?

Imogen: Yeah

I: Yeah

Imogen: Yeah, I think so there's a, because there's a lot of resources out there to look at and so I find that helpful. I'm that sort of person anyway, I like to have, get the information and everything, so

I: It helps you deal with it?

Imogen: Yeah. Yeah.

I: And do you do similar things Ian?

Ian: No. No, I know enough really and I don't want to go researching and, you know, just, that would worry me more, I think.

Parents' distress can be pervasive and long-lasting. Darren described ongoing tearfulness at the second interview, 17 months after his infant's diagnosis and nine months after she had had a liver transplant.

Darren: Sometimes, actually, it's just like it happens like out of the blue, I was crying and all that but suddenly I'm just like, sort of like, think of like, I'm really down, because of my daughter I don't know what to expect and that's for

> myself … so the people around me are a good support to like keep up, yeah, my emotions, because sometimes I'm just like, like crying and I don't know, it just like comes out.

Other parents became irritable. Marie developed feelings of irritability despite her infant doing well physically.

> Marie: Even now we're very happy and haven't got any stressful, but I'm easily irritable you know, very easy to upset, very easy to angry, even with Madeline, with my Mum, with Max. And people say to me like, 'why you like, why you reacting like this?' like overreacting you know, but I don't know.

8770910 also spoke about being irritable.

> I: So, how do you think it's affected the family emotionally, all of this?
>
> Gary: Oh, I know I get short-tempered a lot.
>
> Gretel: Yeah, I think I get short-tempered and stressed and
>
> Gary: And there's not much you can do for anyone, just be there for them. Sort of, yeah.
>
> I: It's hard to know what to do.
>
> Gary: You don't know what to do.

Anxiety about infection risk due to immunosuppression was prominent for parents whose infant had had a transplant. However, with time many parents became more confident and less worried. Abigail and Adam acknowledged that the risk of infection was much lower a year after the infant's transplant. They also spoke of weighing the risk of infection against the importance of providing normal developmental experiences for their infant.

> Abigail: We're a lot more relaxed now than what we were. Definitely.

I: And have her medications been reduced?

Abigail: Yeah. Quite a few of the medicines have been off, but she's still on a couple of immunosuppressants, so I think the level is still there, but not as, not as critically as it was.

I: You don't need to be quite as careful as you were?

Abigail: No. Oh, look, I don't think so. And she's had quite a few colds and everything that she's just brushed off, so she's starting to get stronger in that sense, so we're not as, we're not as worried about, I'm not as worried about that anymore, I should say, not Adam, me! [laughs]

Adam: She can go out in the back yard and play in dirt and

I: You're not too worried

Adam: I am worried, I'm not reckless

I: Yeah, yeah

Adam: But I do think she needs to have, you know

Abigail: Some exposure, yeah I know, yeah [laugh]

Adam: to get to know her environment, you know, picking things up and putting them in her mouth, you know, you can't, we can't, we can't

Abigail: No, we can't

Adam: shield her from that forever.

How well the infant was progressing had an effect on parent adjustment. For example, Marie and Max spoke about how their emotions had changed over time.

Marie: Yep. First when she was sick, everything, we worry about her, we did not enjoy, we don't know what was going on outside just always thinking

about her, and her, that was, therefore we were really down this time, yeah. And now when she's grown up she's back to the normal and we go to the check up to the doctor and everything was fine, we're much happier. I think so, yeah. It's like a rollercoaster! [laughs] Yeah, alright, and now we're at the high point! [laughs] Not the low one, like in the hospital, yeah.

Max agreed, particularly emphasising how the family's life had returned to normal.

Max: Yeah, I think, we have the first time, been, last year she was in hospital for a long time and, yeah, but now I feel very happy and she's back to normal and yeah, I feel like, when I'm back from work I see her, you know, say 'papa' or something like that and I'm very happy and I think all the, all the thing we do for the payoff you know. And I think it's amazing because you see her last year and now she's here it's very different and she's back to normal kid, so I'm really happy you know. And yeah, so we are, she's alright now and we, our family is back to normal we enjoy, you know, the activities, normal social and [inaudible] social and I can work and Marie can take her to shopping and I'm very happy.

4.8.4 Adjustment of infants

The illness resulted in some infants being anxious and having difficulty with separation from parents.

Odette: Yeah, because she's very clingy, Olivia, she's, you know, if I, with both of them actually, if I need to leave here, they either have to be at the other end of the room and I have to sneak out, like I can't explain to them, 'Mum's going out for a bit' because then the tears start and [sister] has actually had a meltdown one time because I tried to explain to her that I was going out without her and she couldn't cope. But she's OK when I drop her off

at school, but if I go anywhere, like to pick [brother] up from soccer she has to come with me.

Some of the infants seemed to be able to cope well with the hospital experience. Ben and Brooke talked about how well their infant was coping with procedures:

Ben: She's so tol-, she's a lesson to us actually

Brooke: Yeah

Ben: Because she's had more blood tests in six months than I've had in my entire life, she's been pricked with needles, she's had blood drawn out, she's had horrible, horrible things happen to her and she, she puts up with it all and she'll be smiling two minutes later so we kind of have to do the same thing, you know [laughs]

Brooke: Yes, we follow her example. She's a really brave little soul.

4.8.5 Adjustment: developing a different perspective

Fourteen of the 25 families included in the qualitative analysis expressed positive emotions at some point in the study. Parents talked about the need to accept what is happening for their child, illustrated by Donna, whose infant had Biliary Atresia.

Donna: But we're trying to support each other, we're trying to deal with it, trying to accept it and trying to look at the positive points, you know, and that's what we're doing, me and my husband need to be positive and to be supportive of each other, there's no, we don't have a choice. We just have to deal with it and accept the situation. And, you know, Debbie's illness is not permanent, it's temporary and it's going to, and she's going to get better. And that is the positive point that we look at. The future, yes.

Other families spoke about feeling lucky to have been able to receive high quality treatment and for other aspects of their lives.

> Gretel: I mean I sometimes feel a bit angry, why, why, why us? I mean, but then I think well it could be a lot worse, so we should be grateful, in a way
>
> Gary: Yeah
>
> Gretel: And we live in a good country with good medical support, so
>
> Gary: And we live in a nice part of [city]

Jason also spoke of feeling fortunate for the treatment of his daughter.

> Jason: But the whole thing's been tempered by a sense of gratitude really and an awareness of how lucky we are to have this system in place. I mean, I just know for a fact that at the moment around the world there are fathers holding their baby with biliary atresia in their arms who is going to die in the next year or so. You know, and I just can't forget that, that we're actually extremely fortunate that we've got now, to all intents, a healthy, healthy little baby so, you know, I feel lucky.

Parents also reported other stresses now seemed less significant, with the suggestion that they felt they could cope with anything compared with the stress of the illness.

> I: So, have there been any other big stresses on you as a family, other than his illness?
>
> Peter: Not really big stresses
>
> Penny: We never think, you know, if there, if we didn't have this stress, this problem, we'll think something, silly thing is a big issue, like that. Now,

whatever comes is nothing to us, yeah, so like that. God has given us a lesson.

Peter: Nothing's going to be close to that one, or more than that. Whatever it's going to be, it's going to be a small thing for us because we've gone through a big thing.

Brooke talked about how the infant's illness had changed her perspective on other people's suffering:

Brooke: Yeah, I think it's probably made me think about other people a lot more. Like be a bit more compassionate about others whereas previously I'd be a bit more nyi [noise to indicate dismissal] you know, with other people's issues and problems and so forth. I think this has made me appreciate, yeah, society and so forth, you know, issues that you do have to deal with a little bit more compassionately.

4.9 Conclusion

This chapter presented the results of the thematic analysis of the parent interviews. Six overarching themes were identified that give a comprehensive understanding of parent and family experience of infant illness. Uncertainty was a characteristic of all stages of the infants' illness, from recognition that the infant was ill right through to the ongoing management of the illness after transplantation and parents' expectations for the future. Uncertainty created distress for parents that made coping difficult as expectations changed or unpredictable events occurred. Parents were acutely aware of their infant's vulnerability in the face of the illness, physically and emotionally, raising fears of loss and further adding to the sense of uncertainty. The experience engendered a sense of isolation for parents as they attempted to manage the illness. Dealing with the infant's illness in the face of also keeping other aspects

of life going, including the needs of other children or the practicalities of everyday life, was exhausting for some parents. However, their shared experience of adversity led to couples and families becoming closer in their relationships as they supported each other. Although sometimes friends and families were not able to fully comprehend the parents' experience, which was isolating for parents, parents took hope from the experiences of other families who had sick children. Such shared experience was also distressing at times, however, when other families lost children to illness. Families had to adjust to the diagnosis of the illness, resulting in changes over time in their relationships and their coping mechanisms. The infants themselves adjusted to their illness, many becoming anxious but some developing resilience. Overall, families recognised that they had developed a different perspective on life as a result of having experienced the illness, feeling that they were better able to cope with adversity.

5 Integrated Quantitative and Qualitative Data Analysis Results

5.1 Synopsis

The previous two chapters presented the findings from the quantitative data analysis and the qualitative data thematic analysis. This chapter reports the results of the integrated mixed methods analysis.

Parents' qualitative interview transcripts were analysed for content according to their scores on each of the scaled variables from the study questionnaires, comparing the transcripts of parents who scored either above or below the study group mean for each scale.

The main results of interest are reported, including important negative findings.

Parent distress is common, with all parents describing significant distress at some point. However, some parents were no longer reporting distress at the time of the interview. The majority of families in which at least one parent scored above the group mean on the DASS described current distress during their interviews.

Lack of extended family support is an important risk factor for mothers' distress, for more problems in family functioning, and for greater impact of the illness on the family. All mothers who reported not having enough extended family support scored above the group mean on the DASS. All families in which both parents scored below the mean on the FAD reported that they had enough extended family support. There was only one family in which both parents scored below the mean on the IFS who reported not having enough extended family support. However, the family also noted that their infant's illness was not severe and the impact on them was not great.

The parents' discussion of the effects of the infant's illness on the family revealed that although disruption to the family is common, families who have greater difficulty adjusting to the disruption have higher IFS scores. In addition, financial stress is a specific risk for parents reporting a greater impact of the illness on the family; all of the parents who reported worrying about financial problems scored above the group mean on the IFS. Parents who struggled to adjust to the family disruption caused by the infant's illness rated the infant as having more problems on the CBCL, compared with parents who reported that they were able to adjust to the disruption.

The integrated analysis did not reveal a relationship between father engagement, parent distress or family functioning, a confirming the quantitative analysis results.

The integrated analysis therefore supported the findings of the quantitative analysis and the qualitative thematic analysis.

5.2 Introduction

All three of the study hypotheses were partially supported by the analysis of the quantitative data. The first hypothesis that the parents of infants with serious liver disease would have high levels of distress was supported in the case of mothers at Time 2, but not for fathers. However, the result for mothers may reflect a Type I error (see Chapter 6, Discussion). The quantitative analysis did not demonstrate alterations in family functioning, but did show that mothers' perceptions of the impact of the illness on the family were comparable with published data of other families of children with chronic illness. The quantitative analysis also partially supported the second hypothesis that fathers' engagement in the care of the infants would have an impact on parent distress and family functioning. Fathers' own ratings of their engagement were predictive of their ratings of family functioning, however the finding is not likely to be clinically significant (see Chapter 6, Discussion). Mothers' ratings of

father engagement were not predictive of the other study measures. The quantitative analysis also partially supported the third hypothesis that parent distress, family functioning and fathers' engagement will have predictive value for the emotional and behavioural outcome for the infants. Mothers' and fathers' reports of the impact of the illness on the family were significant predictors of infant emotional outcomes. The quantitative data analysis also showed an effect of family demographics and illness characteristics on infant outcomes.

The results of the quantitative data analysis were therefore discordant with clinical experience of high levels of distress in parents of infants with serious liver disease. The findings were also contrary to expectations that father engagement would have an impact on parent and family responses to the illness.

The qualitative data thematic analysis revealed important themes in the parents' discussion of their experience of their infants' illness. All parents reported distress at some stage during their infant's illness, often related to the uncertainties associated with the illness, the infant's vulnerability or parent isolation. Parents reported changes in their relationships and noted the importance of shared experience with others who faced similar situations. Parent adjustment to the illness was influenced by each of these factors and changed over time, with many parents reporting an altered perspective on life as a result of their experience. The thematic analysis also demonstrated the importance for parents of dealing with other life pressures while also dealing with the infant's illness.

The qualitative thematic analysis therefore suggested support for the hypothesis that parents of infants with serious liver disease will have high levels of distress, despite the quantitative data demonstrating no increase in psychological symptoms in parents compared with the normal population. The qualitative analysis also suggested that the illness had resulted in changes in family relationships, one

element of family functioning. The qualitative analysis did not reveal any additional information about the effects of father engagement. The thematic analysis had mixed findings in relation to the outcomes for the infants. While not specifically included in the parent interview, some parents discussed their infant's progress during the interviews. Awareness of the infants' vulnerability was important to many parents. Some infants developed emotional problems such as anxiety, while other infants were reported by their parents to be coping well. The analysis also drew attention to the importance of social support and additional stressors for families, two areas that were the focus of specific questions in the parent interviews.

Several issues therefore remained after the quantitative and qualitative analyses. Firstly, conflicting evidence in the data regarding parent distress and family functioning requires clarification. Secondly, the effects of other life stressors and adequacy of social support on parent distress and family functioning have not been assessed in the quantitative or qualitative analyses. Thirdly, the lack of a demonstrated effect of father engagement in the quantitative data analysis has not been further assessed using the qualitative data. Finally, the relationship between parent psychological symptoms, family functioning, fathers' engagement and infant emotional and behavioural outcomes will be explored to confirm the quantitative analysis findings. The integrated data analysis will therefore examine the relationship between parent scores on the study measures and the qualitative data obtained during the parent interviews.

The chapter will describe the overall approach to the integrated analysis and will then provide a summary of the main findings.

5.2.1 Analytic Approach to the Integrated Analysis

The integrated quantitative and qualitative analysis focuses on how parents discuss their psychological distress and family functioning, the influence of father

engagement on parent reports of their experience, and the outcomes for the infants. As described in Chapter 2, Methods, parents were divided into two groups according to whether their scores on the questionnaires (DASS, FAD, IFS, DADS and CBCL) were below the group mean, or above the group mean. The continuous quantitative data were thus transformed into categorical data and then imported into NVivo as case attributes. The process allowed comparison of the qualitative data between the groups who scored below or above the mean on each scale to assess differences between the groups. In this way, parent discussion of emotions, family relationships and the effects of the illness on the family were analysed, comparing the group of parents who scored below the mean with the parents who scored above the mean on each scale.

A summary of the characteristics of the total study population and of the families chosen for the qualitative and integrated mixed methods analyses are presented in Appendix M.

5.3 Parent Distress: Past or Present

The integrated analysis demonstrates that parent distress in relation to the infants' illness is complex. All parents discussed distress in relation to their infant's illness, but parents who scored above the group mean on the DASS (more psychological symptoms) reported current distress, while those who scored below the mean (fewer psychological symptoms) reported previous distress. (Table 5.1). The pattern was most apparent at Time 1 and suggests that although all parents had experienced distress about their infant's illness at some stage, many parents were no longer distressed at the time of interview. The finding is in line with the instructions for completing the DASS, which asks parents to report on their psychological symptoms within the preceding week.

The pattern holds for families in which parents' DASS scores differed (that is, one parent scored above the group mean and the other parent scored below the mean). In most of these families, if at least one of the parents had a DASS score above the group mean, both parents continued to talk of distress due to their infant's illness in the present tense. The finding suggests that when one parent remains distressed, the emotional distress continues to be a current concern for the couple (Table 5.1).

Three cases are exceptions to the pattern.

1. Charlie and Christine both scored below the DASS group mean at Time 1, but still spoke in the present tense about psychological symptoms. However, their symptoms were mild, potentially accounting for the low DASS scores. The parents were interviewed in the hospital because their infant was an inpatient, potentially influencing their expression of distress as a current concern despite their reports that they were generally coping well.
2. Victor and Valerie both scored below the group mean on the DASS at Time 2 but still spoke in the present about psychological symptoms, specifically worrying that their infant, Vivienne, may be in discomfort. However, they reported that they had improved from previously and didn't think about Vivienne's illness when they were at home. Their discussion of negative emotions in the present tense may have reflected the fact that they were interviewed at the hospital outpatients' clinic.
3. Simone scored below the group mean on the DASS while Stephen scored above the group mean at both time points, but they spoke in the past tense about negative emotions. Their infant, Sam, was doing well physically at both time points and they said that the illness wasn't currently affecting their lives. However, at Time 1 Simone had back problems that limited her ability to perform routine household chores. As a result, Stephen was providing more

support in looking after the family in addition to having returned to work, potentially leading to his higher DASS score. At Time 2 Sam had recently suffered from influenza, which had come as a shock to both parents and a reminder that things can go wrong. It appears that Stephen had had an increase in psychological symptoms as a result of Sam's recent influenza, leading to Stephen's higher scores on the DASS. However, Sam's liver disease had improved and the parents therefore discussed their emotional distress due to Sam's liver disease in the past tense. In this case, it appears that Stephen's scores on the DASS were due to factors other than Sam's liver disease.

There was no difference in the types of emotions discussed by families in which both parents reported fewer psychological symptoms compared with those reporting more symptoms. All parents discussed negative emotions such as shock, tearfulness, guilt, depression and worry. Although not all parents talked about positive emotions, similar numbers of parents in each group discussed positive emotions such as hope, gratitude, acceptance and happiness (Table 5.1).

Stressors in addition to the infants' illness were common, regardless of parents' DASS scores. Having experienced additional stressors did not alter the way parents spoke about their emotional responses to the infant's illness. Whether or not parents had experienced additional stressors, they spoke about worrying, being tearful, or feeling emotional or stressed. Parents from each group also spoke of positive emotions such as happiness, pride in the infant's development or gratitude.

Table 5.1

Parent DASS Scores in Relation to Discussion of Emotions at Interview

	Time 1 (*N* = 25)		
	Both parents below the DASS group mean (*N* = 8)	One parent above the DASS group mean (*N* = 8)	Both parents above the DASS group mean (*N* = 9)
Negative emotions (*N* = 25)	8	8	9
Positive emotions (*N* = 14)	5	4	5
Current distress (*N* = 17)	1	7	9
Past distress (*N* = 8)	7	1	0
	Time 2 (*N* = 25)		
	Both parents below the DASS group mean (*N* = 4)	At least one parent above the DASS group mean (*N* = 13)	Both parents above the DASS group mean (*N* = 8)
Negative emotions (*N* = 25)	4	13	8
Positive emotions (*N* = 14)	4	5	5
Current distress (*N* = 21)	1	12	8
Past distress (*N* = 4)	3	1	0

5.4 Extended Family Support

Parents were asked at interview whether they felt they had enough support, and to discuss the nature of the support available to them. Analysis of the interview data revealed that all families spoke of having supportive friendships, but families differed in relation to the availability of extended family support. Extended family support

consisted mostly of grandparents, but some parents relied on their own brothers and sisters or reported that they had no extended family support.

For mothers, not having enough extended family support appears to be a risk factor for distress, reflected in mothers' DASS scores. All of the mothers who scored below the study group mean on the DASS reported that they had enough extended family support. Extended family support was less important for fathers and was not reflected in their DASS scores.

Extended family support was an important risk factor for more problems in family functioning and parent perceptions of a higher impact of the illness on the family, reflected in parents' scores on the FAD and the IFS. Parents who reported that they did not have enough support also scored above the study group mean on the FAD and the IFS.

5.4.1 Extended Family Support and Parent Psychological Symptoms

There were mothers and fathers who scored above the group mean on the DASS despite reporting that they had enough extended family support. For fathers there was no pattern between adequacy of extended family support and fathers' DASS scores. However, there were no mothers at either time point who scored below the group mean on the DASS and who also said that they did not have enough extended family support (Table 5.2). The finding suggests that extended family support is not sufficient to protect mothers from developing psychological symptoms, but that lack of extended family support is a risk factor for mothers reporting more psychological symptoms.

Table 5.2

Adequacy of Social Support in Relation to Parent DASS Scores

	Time 1 (*N* = 25)	
	Enough Support (*N* = 19)	Not Enough Support (*N* = 6)
Mother DASS below the mean (*N* = 12)	12	0
Mother DASS above the mean (*N* = 13)	7	6
Father DASS below the mean (*N* = 12)	10	2
Father DASS above the mean (*N* = 13)	9	4
	Time 2 (N = 25)	
	Enough Support (N = 19)	Not Enough Support (N = 6)
Mother DASS below the mean (*N* = 9)	9	0
Mother DASS above the mean (*N* = 16)	10	6
Father DASS below the mean (*N* = 12)	10	2
Father DASS above the mean (*N* = 13)	9	4

Adequacy of support was not reflected in the nature of parents' emotional reactions. All families discussed a range of negative emotional reactions, regardless of whether they had adequate support. Although not all families discussed positive emotional responses, there were parents in each group who spoke about positive emotions such as hope, gratitude and acceptance.

Parents who reported fewer psychological symptoms were no different from the parents who reported more symptoms in how they discussed their experiences of

support. Parents in both groups described positive and negative experiences with support from family, friends and other supports.

5.4.2 Extended Family Support and Family Functioning

Although some families who reported that they had enough support scored above the mean on the FAD, almost all families in which both parents scored below the group mean on the FAD (fewer problems in family functioning) reported that they had enough social support overall (Table 5.3).

Some parents who scored below the group mean on the FAD reported that grandparents were not available or were not supportive. However, the parents who scored below the mean on the FAD who spoke of unsupportive grandparents also reported that other relatives supported them. Only one family scored below the group mean on the FAD and also reported that they didn't have enough social support overall. Adam and Abigail both scored below the mean on the FAD at Time 1. Abigail reported that her family didn't offer enough support. However, both parents reported that Adam's parents were very supportive. Although the parents reported that they didn't have enough support overall, the extended family support that was available to them appears to be reflected in their FAD score, which is below the study group mean (Table 5.3).

At Time 2 there were six families in which both parents scored below the mean on the FAD, all of whom reported that they had adequate support from extended family members (Table 5.3).

The findings suggest that extended family support is not sufficient to protect families from problems in family functioning, but that lack of extended family support is a risk factor for more family functioning problems.

Table 5.3

Adequacy of Social Support in Relation to Parent FAD Scores

	Time 1 (*N* = 25)	
	Enough support (*N* = 19)	Not enough support (*N* = 6)
Both parents below the FAD group mean (*N* = 9)	8	1
One parent above the FAD group mean (*N* = 6)	4	2
Both parents above the FAD group mean (*N* = 10)	7	3
	Time 2 (*N* = 25)	
	Enough support (*N* = 19)	Not enough support (*N* = 6)
Both parents below the FAD group mean (*N* = 6)	6	0
One parent above the FAD group mean (*N* = 8)	6	2
Both parents above the FAD group mean (*N* = 11)	7	4

5.4.3 Extended Family Support and Impact of the Illness on the Family

Lack of extended family support is also a risk factor for parents reporting higher impact of the infant's illness on the family.

Discussion of extended family relationships showed a similar pattern to the earlier findings in relation to family functioning. Parents in each group reported having good extended family support, but lack of extended family support was associated with parent reports of higher impact of the illness on the family.

Only one family with both parents who scored below the mean on the IFS at Time 1 reported not having enough extended family support. Neil and Nicole reported that extended family members lived at a distance from them and were not available to provide support. In addition, they had recently moved some distance from the city

and had few social contacts in their local area. However, they reported that their infant had few day-to-day problems and so the impact of the illness on the family had not been great (Table 5.4).

At Time 2, there were no families who reported inadequate support and also scored below the mean on the IFS (Table 5.4).

The findings again suggest that extended family support is not sufficient to protect families from experiencing a high impact of the infants' illness on the family, but that lack of extended family support is a risk factor for parents perceiving a greater impact.

Table 5.4

Adequacy of Social Support in Relation to Parent IFS Scores

	Time 1 (*N* = 25)	
	Enough support (*N* = 19)	Not enough support (*N* = 6)
Both parents below the IFS group mean (*N* = 7)	6	1
One parent above the IFS group mean (*N* = 7)	6	1
Both parents above the IFS group mean (*N* = 11)	7	4
	Time 2 (*N* = 24[a])	
	Enough support (*N* = 18)	Not enough support (*N* = 6)
Both parents below the IFS group mean (*N* = 9)	9	0
One parent above the IFS group mean (*N* = 6)	5	1
Both parents above the IFS group mean (*N* = 9)	4	5

[a] *N* = 24: One mother did not complete the IFS at Time 2

5.5 Family Adjustment

The integrated analysis shows that parent reports of greater disruption to the family as a result of the infant's illness, such as time constraint and problems with planning, are reflected in parent scores on the IFS.

Financial stress is a risk factor for parent perceptions of greater impact of the infant's illness on the family. Parents who reported worrying about financial problems also scored above the group mean on the IFS. Other stressors in addition to the infant's illness are not related to parent scores on the IFS.

Parents who reported difficulty adjusting to the family disruption caused by the infant's illness rated the infant as having more problems on the CBCL compared with parents who reported that they were able to adjust to the disruption.

5.5.1 Family Adjustment and the Impact of the Illness on the Family

Many families talked of their immediate family relationships being closer, regardless of their score on the IFS. However, parents who talked about being separated from each other while the infant was in hospital scored above the mean on the IFS. The finding suggests that although the parental relationship was often experienced as being closer, disruption to the family resulting in separation was reflected in parent perceptions of a higher impact of the illness on the family.

Parents who scored above the mean on the IFS reported more disruption to the family than parents who scored below the mean on the IFS. Families with higher scores on the IFS reported changing routines, not having enough time to do things, and having to change future plans. They spoke of the illness as life changing and an upheaval for the family, or reported feeling that their lives had been turned upside down. Those who scored below the group mean reported less disruption, even if

future disruption was anticipated. These parents spoke of taking a day at a time and appeared to cope with the changes that resulted from the infants' illness.

Taken together, the findings suggest that parent scores on the IFS are reflective of parent experiences of the infant's illness. The qualitative data reveals that parental separation due to the infant's illness, altered family routines, lack of time, and changing future plans are important factors that result in parent perceptions of a greater impact of the illness on the family.

5.5.2 Family Adjustment and Financial Stress

Stressors in addition to the infant's illness were common regardless of parent scores on the IFS (Table 5.5).

However, money was important for families and was discussed by 16 families. Nine of these families, however, said they were able to manage and finances were not a stress. All families who spoke of the financial impact of the infant's illness as being stressful had at least one parent who scored above the mean on the IFS (Table 5.5).

Table 5.5

Additional Stressors and Financial Stress in Relation to Parent IFS Scores

	Time 1 (*N* = 25)		
	No additional stressors (*N* = 7)	At least one additional stressor (*N* = 18)	Financial stress (*N* = 7)
Both parents below the IFS group mean (*N* = 7)	2	5	0
One parent above the IFS group mean (*N* = 7)	1	6	1
Both parents above the IFS group mean (*N* = 11)	4	7	6
	Time 2 (*N* = 24[a])		
	No additional stressors (*N* = 4)	At least one additional stressor (*N* = 20)	Financial stress (*N* = 7)
Both parents below the IFS group mean (*N* = 9)	1	8	0
One parent above the IFS group mean (*N* = 6)	1	5	2
Both parents above the IFS group mean (*N* = 9)	2	7	5

[a] *N* = 24: One mother did not complete the IFS at Time 2

5.5.3 Family Adjustment and Infant Emotional and Behavioural Outcomes

As noted in the quantitative analysis chapter, parent IFS scores were predictive of infant emotional and behavioural outcomes on the CBCL. The integrated data analysis confirmed the result, demonstrating a link between parent reports of difficulty adjusting to the illness (suggesting a greater impact of the illness on the family) and poor infant emotional and behavioural outcomes. Although difficulties with planning and time-consuming illness routines were common amongst families, some families

talked about having been able to adjust to the disruption. These families also scored the infant below the mean on the CBCL. However, some families were less able to adjust to the disruption caused by the infant's illness. These families talked about the illness routines being time-consuming and exhausting, of the illness having made major changes for the family, and feeling they were not able to plan. The families also discussed worrying about not being able to spend time together and worrying about the effects of the illness on the infant's siblings. The families who struggled to adjust to the infant's illness also scored the infant above the mean on the CBCL.

5.6 Fathers' Engagement

Parents were not asked about fathers' engagement during the interviews. However, 10 of the families spoke about fathers' support at Time 1 and seven discussed fathers' support at Time 2. The discussion in each case was in relation to fathers being supportive, in one case being better than expected and in another case father became more supportive as a result of the infant's illness. No families talked about the father not providing support, though some fathers expressed the view that they wished they could provide more support.

Parents who reported more father engagement (higher scores on the DADS) were compared with parents who reported less father engagement (lower scores on the DADS). There were no differences between the groups in how parents discussed emotions, family relationships, or family responses to the illness. The result supports the quantitative data analysis findings that parent DADS scores were not predictive of other study measures.

5.7 Conclusion

The integrated mixed methods analysis provides additional insights into the experiences of families of infants with serious liver disease than was revealed by the quantitative and qualitative analyses alone.

Lack of extended family support was revealed as a risk factor for mothers' distress, for more problems in family functioning, and for higher perceived impact of the illness on the family. In addition, parent concerns about financial problems were related to greater impact of the illness on the family.

The integrated analysis supported the earlier quantitative analysis result that father engagement was not predictive of parent distress or family functioning.

Finally, the integrated analysis demonstrated that infant emotional and behavioural outcomes were associated with difficulties in family adjustment to disruptions caused by the illness.

The quantitative analysis demonstrated the predictive value of parent reports of greater family impact of the illness for poorer infant emotional outcomes. The role of family adjustment was identified as an important theme in the thematic analysis of the parent interviews. The integrated analysis suggests that difficulties for parents in adjusting to the disruptions caused by the infants' illness, in the context of lack of extended family support and financial stress, may be a mechanism by which parent perceptions of greater family impact of the illness results in poorer infant emotional and behavioural outcomes.

6 Discussion and Conclusions

6.1 Synopsis

This study demonstrates that the perceived impact on the family of serious liver disease during infancy, in combination with illness characteristics, is predictive of infant emotional outcomes one year later. The study also demonstrates that parent perception of the impact of the illness on the family is mediated by lack of extended family support, poor family adjustment to the infant's illness, and financial stress.

This is the first study to prospectively examine the interactional effects of parent and family factors on the infants' emotional and behavioural outcomes. It is unique in standardising the timing of research to the period following the infants' initial diagnosis, and in including both mothers' and fathers' reports. The study extends previous research findings by identifying early predictors of infant outcomes, therefore suggesting possibilities for early identification of families at risk whose infants may benefit from early intervention.

This chapter will consider each of the research hypotheses in turn. The main research findings are discussed in light of previous research. Strengths and limitations of the study are then discussed, followed by recommendations for future research.

6.2 Hypothesis 1

Parents of infants with serious liver disease will have high levels of distress, demonstrated by the presence of psychological symptoms and alterations in family functioning

6.2.1 Parent Psychological Symptoms

The study results do not support the hypothesis that parents of infants with serious liver disease have high levels of psychological symptoms.

The quantitative data analysis found that fathers' rates of psychological symptoms did not differ from the general population at either baseline or follow up. Although mothers' rates of psychological symptoms were significantly greater in comparison with general population data, the finding is likely to reflect a Type I error as outlined below.

As noted in Chapter 3, Quantitative Data Analysis Results, parent DASS scores were not normally distributed. Crawford and Henry also found positively skewed DASS scores in a large non-clinical population study, noting that the scoring scheme of the DASS results in a large proportion of the general population with low scores.[223] In the current study, the raw DASS scores were therefore converted to percentile scores according to Crawford and Henry's conversion tables.[223] While the conversion resulted in normal distribution, the only comparison data that were available were for median rather than mean scores.[223] Non-parametric tests were therefore conducted.

Although mothers' median scores did not change between Time 1 and Time 2, the Wilcoxon signed rank test showed mothers' scores to be significantly higher at Time 2 compared with the population median. The mean percentile scores for mothers increased slightly from Time 1 ($M = 57.29$) to Time 2 ($M = 59.76$). The combination suggests that some individual scores were higher at Time 2 in

comparison with Time 1, resulting in a higher mean score but an unchanged median. Tied scores would result in an alteration in the rankings of scores used in the Wilcoxon signed rank test, leading to a significant finding at the second time point despite both groups having the same median percentile. Although the normative data are based on the general population, rather than on separate data for women and men, research has demonstrated very small non-significant differences in DASS scores due to gender in an Australian population.[244] The normative data median score was therefore suitable for statistical comparison for mothers' and fathers' scores. Finally, the finding was of marginal statistical significance ($P = .048$). Overall, the apparent elevation of mothers' psychological symptom scores at Time 2 is likely to be a Type I error. Future research with a larger sample could help to clarify the differential effects on mothers and fathers.

The finding that parent psychological symptoms are no greater in the study group of parents compared with normative data is in line with previous limited research findings in parents of children with liver disease who have not had a transplant.[171] In paediatric liver transplant research, however, four cross-sectional studies of children and adolescents who have had a liver transplant up to 11 years previously demonstrated significantly greater emotional distress in parents compared with normative data.[142,148,169,170] The self-report measure used in each study is likely to have an influence on the findings. Each study reported high rates of emotional distress in parents using an HRQOL measure. However, one of the studies[148] also included measures of depression and general psychological distress and found no differences in the parents compared with test norms on the psychological measures despite finding elevated levels of emotional distress on the HRQOL measure. The finding suggests higher emotional impact on parents of chronic liver disease and liver transplantation in comparison with normative data, but no greater rates of

psychological symptoms. In retrospect, inclusion of an HRQOL measure in the current study would have allowed comparison with other HRQOL research. The study showing different results from HRQOL measures in comparison with psychological symptom measures was not available at the time the current study was designed. However, the current study extends previous findings regarding psychological symptoms to the early stages of illness following initial diagnosis, and to illness occurring during the developmental period of infancy.

The finding is supported by the results of the qualitative data analysis in the current study. The thematic analysis of the parent interviews revealed specific details about parent distress following a diagnosis of serious liver disease in infancy. Parents described the uncertainties of the illness in relation to diagnosis and treatment, and their awareness of the infants' vulnerability in the face of serious liver disease. Other qualitative research in liver and solid organ transplantation has also reported parent uncertainty, related to parents' feelings of vulnerability about the transplant.[211,219] Further, parent uncertainty has been related to adjustment in parents of children who have had a liver transplant.[210] The current study also identified that feelings of isolation were of concern to parents, in line with results from qualitative research following paediatric organ transplantation.[213] Parents in the current study described the importance of additional life stressors, such as keeping the family going while caring for the sick infant. Other published qualitative research in solid organ transplantation has also reported that dealing with other life stresses such as returning to family routines following discharge from hospital as a concern for parents.[219] The thematic analysis demonstrated the emotional impact of the infants' illness on parents but did not show evidence for an increase in psychological symptoms in the parents, supporting the quantitative data analysis.

The integrated data analysis revealed that parent distress is complex in families of infants with serious liver disease. The analysis indicated that although all parents had been distressed at some point during the infants' illness, many parents did not have psychological symptoms at the time of study participation. As previously noted in Chapter 3, Quantitative Data Analysis Results, there was a wide range of times between infant diagnosis and study participation due to difficulties contacting some parents or arranging a time for the parent interview. It is possible that some parents delayed study participation until they were less distressed. However, the integrated analysis results were not explained by differences in the length of time from the infant diagnosis to completion of the study measures: in the group of parents who scored above the group mean on the DASS, the length of time between diagnosis and interview at Time 1 ranged from 5 to 12 months, while in the parents who scored below the group mean score the range was from 3 to 11 months.

In addition, the integrated analysis did not find a relationship between additional stressors and parent stress. The finding may reflect an altered perspective on life as indicated by some parents, or perhaps that parents focus their emotional energy on the infant rendering other stressors less important. It is also possible that parents under-report their own psychological symptoms in the context of dealing with serious illness in infants.

Finally, the integrated analysis revealed that lack of extended family support is a risk for increased psychological symptoms for mothers, but not for fathers. Previous research has shown an effect of social support, but the specific effect of extended family support has not been previously identified. Simons and colleagues found social support to be associated with less distress in mothers of solid organ and bone marrow transplant candidates, but not in fathers.[173] The novel finding from the current study warrants further investigation.

It would be of interest to compare the study results with research in the post-partum period because the majority of the parents in the current study are parents of newborn children. However, research on psychological symptoms during the post-partum period has largely focussed on mothers rather than fathers. One study of 325 Australian primiparous women used the DASS[245] but only raw sub-scale scores are reported, and the total score, percentile scores and median scores are not available. Due to the non-normal distribution of raw DASS scores in the current study, it is not possible to compare the scores between the studies.

It would also be of interest to compare the current study findings with other research in parents of children with chronic illness, but the DASS has not been reported in parents of children who have a chronic illness.

In summary, the current study results do not support the hypothesis that parents will have elevated levels of psychological symptoms. The inclusion of parent interviews supported the finding and further demonstrated that although all parents have been distressed at some point during the infants' illness, most parents are resilient. Lack of extended family support is a risk factor for the development of psychological symptoms for mothers, but not for fathers.

6.2.2 Alterations in Family Functioning

The study results partially support the hypothesis that parents of infants with serious liver disease would have alterations in family functioning. Mothers' and fathers' ratings of general family functioning were in the healthy range at both time points. However, mothers' ratings of the impact of the infants' illness on the family were similar to other chronic illness groups at both time points, while fathers' ratings of the impact of the infants' illness on the family were significantly lower than reported in other chronic illness groups. However, the qualitative data revealed greater complexity in the families' responses to the infants' illness. Subtle changes in family

relationships were not reflected in the results of the questionnaires. Lack of extended family support and the addition of financial stress were identified as risk factors for poorer family functioning in those parents who scored above the group mean on the study measures.

6.2.2.1 General family functioning

The quantitative data analysis demonstrated that at both time points, mothers' and fathers' ratings of general family functioning were within the healthy range in comparison with healthy populations and compared with families who have a medically ill member ($M = 1.89$).[226,227] The finding is in line with previous studies that have used the FAD General Functioning sub-scale in families of children who have liver disease. One longitudinal study used the FAD General Functioning scale in mothers of children both pre- and post-transplantation and found family functioning to be in the healthy range at both time points.[171] Two other research studies used the FAD General Functioning scale in families of children who have had a liver transplant. Although the studies had participation from some fathers as well as from mothers, fathers' scores were not reported separately. Both studies reported family functioning within the healthy range.[148,180] Although a fourth study used the FAD General Functioning scale in mothers of children being evaluated for a liver transplant,[187] the study only reported correlational data between the FAD and other study measures and the mean scores were not reported.

The current study finding of normal family functioning is in also line with the Ontario Child Health Study, which found healthy family functioning measured by the FAD General Functioning sub-scale in families of children with chronic illness. The researchers found no significant differences in family functioning between the families who had a child with chronic illness in comparison with families with healthy children.[229]

The current study finding is also in line with previous Australian research examining parents of children with cancer. Sawyer and colleagues studied mothers of 22 children with cancer and 21 healthy controls at two time points: 7 weeks after the child's diagnosis and again 1 year later.[246] Family functioning, measured by the FAD General Functioning sub-scale, was within the healthy range on both occasions. However, the mothers' scores were significantly higher at follow up compared with the healthy control group, despite remaining in the healthy range.

The qualitative data analysis in the current study provided additional information about family functioning in this group of parents. The thematic analysis demonstrated changes in relationships in the family and in friendships; parent reports of relationships within the family becoming closer were common. Similar changes in family and social relationships have been noted in families of children who have had a liver transplant[210] and solid organ transplant.[213] Adjustment to the illness was additional to the normative adjustment to the birth of the infant and parents reported disruption to family routines. Families continued to adjust over the course of the first year of the illness, a process that was dependent on the infants' clinical progress. The integrated data analysis revealed that the reported changes in family functioning in response to the illness were not reflected in parent FAD General Functioning scores. The results suggest that while family functioning in general is within the healthy range in the families of infants who have serious liver disease, the instrument is not sensitive to changes in family functioning in response to a significant emotional stressor. The integrated analysis also revealed that lack of extended family support is a risk for higher scores on the FAD, while changes in other social relationships are not. The analysis therefore suggests a specific effect of poor extended family support on family functioning. The finding appears to reflect an interactional effect whereby parents who score higher on the FAD have wider problematic family relationships.

In summary, the current study found family functioning to be in the healthy range three months after the infants' diagnosis of serious liver disease and at follow up. Although the research findings are in line with other research in paediatric chronic liver disease, liver transplant and other chronic childhood disease, the study extends previous research by including parent interviews and an integrated analysis of the study results. The integrated analysis revealed changes in family functioning as well as a specific effect of lack of extended family support on family functioning. The study also extends previous research to families of infants, and to the period following the infants' diagnosis.

6.2.2.2 Impact of the illness on the family

The quantitative data analysis revealed that mothers' reports of the impact of the illness on the family did not significantly differ from the published normative data from families with children with other chronic illness at either time point. However, fathers' reports of the impact of the illness on the family were significantly lower than the comparison data at both time points. The current study is the first to examine both mothers' and fathers' reports of the impact of paediatric liver disease on the family.

The results are in line with other research that has been conducted with mothers of children who have had a liver transplant. DeBolt and colleagues studied the families of 41 children who had had a liver transplant. The informants were all mothers except for two fathers. The researchers found no significant difference between IFS scores in the liver transplant group compared with other families of children with a chronic illness.[247] Similarly, Kaller and colleagues used the IFS in their study of 181 children who had had a liver transplant at least one year previously. Although 16.5% of the informants were fathers, the researchers did not separately analyse mothers' and fathers' data. There was no significant difference in the impact on the family

compared with normative chronic illness data.[189] Given that the vast majority of informants were mothers the result can be assumed to reflect mothers' data.

Rodrigue and colleagues separately studied fathers and mothers of children being evaluated for solid organ or bone marrow transplant. It is difficult to compare their data with the current study for several reasons outlined below, however the research is interesting because mothers and fathers were evaluated separately and comparison with data from other chronic illness groups is included. The research is difficult to directly compare with the current study because the researchers did not report separate analyses for type of transplant, or direct comparison between mothers and fathers. In addition, the researchers report analysis of the IFS sub-scales rather than the Total score. In one of the studies, the fathers of 18 children (five with liver disease) reported a greater impact on the family due to financial issues, disrupted planning and overall family burden compared with other families of children with chronic illness.[181] In another study, the mothers of 36 children (nine with liver disease) also reported significantly more disrupted planning and family burden compared with other families of children with chronic illness.[182] At follow up six months after the child's transplant the mothers reported significantly greater impact due to financial issues, disrupted planning, and caregiver burden compared with the pre-transplant period.[183] While not directly comparable with the current study, the research suggests that both fathers and mothers report greater impact on the family of solid organ or bone marrow transplantation compared with families of children with other chronic illnesses. Differential impact of transplant type cannot be excluded, so direct comparison with liver disease or liver transplantation is not possible.

In contrast to Rodrigue and colleagues' work and the current study results, other researchers have found no significant difference in IFS scores between fathers and mothers of children with cancer[248] or infants with congenital heart disease.[249] Of

interest, the fathers of children with cancer identified themselves as the child's primary caregiver.[248] It is possible that primary caregiver status affects parent report of the impact of the illness and accounts for the different findings in comparison with the current study. The study of infants with congenital heart disease assessed parent reports of the impact of the illness on the family 12 months after the infants had had cardiac surgery.[249] In the current study, only a quarter of the infants had had a transplant at the first time point and only half had had a transplant at follow up. It is possible that major surgery may affect fathers' reports of the impact of the illness on the family.

The integrated data analysis in the current study revealed that financial stress specifically adds to parents' perceived impact of the illness on the family. All families who reported financial problems also had at least one parent who scored above the group mean on the IFS. The integrated analysis finding is in line with Rodrigue and colleagues' findings reported above that reported on the financial impact sub-scale of the IFS.[181,183] The finding is interesting because Rodrigue and colleagues' research was conducted in the United States where the costs of liver transplantation are a considerable burden on families who do not have health insurance. The public health care system in Australia provides transplantation at no cost to families. Nevertheless, the current study results suggest that financial burden identified by parents at interview, such as work disruption and other indirect costs such as car parking and food, remain a significant source of stress for families. The integrated analysis in the current study found that other stressors were common and there was no clear pattern in relation to IFS scores.

The integrated analysis also indicated that lack of extended family support is a risk factor for parent reports of a higher impact of the illness on the family, as was also found for general family functioning and for mothers' psychological symptoms. Social

support has been associated with family adjustment following discharge from hospital after paediatric liver transplant.[250] Werner and colleagues found social support to be a significant predictor of the impact of congenital heart disease on the family for both mothers and fathers.[249] The current study extends previous research in suggesting a specific effect of lack of extended family support, rather than lack of support in general, on parent and family adjustment.

In summary, in the current study mothers' reports of the impact on the family of serious liver disease in infancy are comparable with results from other chronic disease research. Fathers' reports of the impact of the illness on the family are significantly lower than reported in other chronic disease research. Differences in illness types, primary caregiver status or whether the infant has undergone major surgery may account for the observed differences. The current study also found that financial stress and lack of extended family support are risk factors for parent perceptions of an increased impact of the illness on the family. The current study adds to existing research by identifying a differential effect on mothers and fathers of serious liver disease in infancy soon after diagnosis, in contrast with other chronic illness research findings, and by identifying the specific effects of lack of extended family support.

6.3 Hypothesis 2

Fathers' perceived engagement in the infants' care will have an impact on parent distress and family functioning

This is the first study to examine father engagement in infants with chronic liver disease. It is also the first study to examine father engagement in children with a chronic illness using a longitudinal design.

The study results do not support the hypothesis that father engagement would have an impact on parent distress and family functioning.

The quantitative data analysis demonstrated that mothers' reports of father engagement, measured by the DADS questionnaire, did not predict mothers' reports of their own psychological symptoms, family functioning or impact of the illness on the family.

Fathers' reports of their engagement did not predict their reports of their own psychological symptoms or impact of the illness on the family. However, fathers who reported that they spent less time helping in the medical care of the infants also scored more highly (indicating greater problems) on the family functioning measure, the FAD General Functioning scale. Although a statistically significant finding, the results are not likely to be clinically significant. As previously noted, fathers' mean scores on the FAD General Functioning scale are significantly lower than published cut-off scores. Although lower scores on the DADS Amount scale were predictive of higher scores on the FAD, higher scores on the FAD in this situation do not indicate problematic family functioning. It is possible that lower father engagement is a marker of less healthy family functioning, however the analysis of mothers' data did not support the finding.

The quantitative analysis results are confirmed by the integrated data analysis. The integrated analysis found no difference between parents with low or high scores on the DADS in how they discussed emotions, family relationships or the family's response to the illness. It is possible that the lack of findings of an effect of fathers' engagement during the interviews is due to the parents being interviewed together. For example, mothers may be reluctant to report low levels of father engagement or dissatisfaction with the relationship while being interviewed together. However, each parent completed the questionnaires independently and the results support the lack

of findings at interview. It is also possible that early in the course of the infants' illness parents are working together as they adjust to the adversity, resulting in both parents being satisfied with the level of father engagement. This view is supported by the qualitative data that revealed that many parents reported that their immediate family relationships had become closer as a result of the infants' illness.

The finding is in contrast to findings in paediatric diabetes research indicating a correlation between father engagement, lower levels of depression in mothers, and greater anxiety in fathers.[251] Other researchers have also found an association between mothers' ratings of helpfulness of father engagement and maternal psychiatric adjustment, lower impact of the child's chronic illness on the family, and healthier family functioning.[205] However, these studies were cross-sectional in design and therefore only able to assess associations between measures or differences in mean scores between families with high versus low father engagement.

As previously noted in Chapter 1, Introduction, in children with chronic illnesses younger child age is associated with greater father engagement.[203] It is possible that the fathers in the current study are particularly well engaged due to the age of the infants, leading to the lack of statistically significant differences.

The current study has a longitudinal prospective design, therefore the results are more robust than the cross-sectional studies. In addition, the results are supported by the additional qualitative data. It is also possible that differences in findings compared with other chronic illness research are related to the developmental stage of the families.

In summary, the current study has not demonstrated an effect of father engagement on parent distress or family functioning in families who have an infant with serious liver disease. The quantitative findings are confirmed by the qualitative data. This study is the first to examine father engagement of chronic illness occurring during

infancy, and is the first longitudinal study of father engagement in chronic illness. The study extends previous research into father engagement in chronic illness with novel findings of a lack of effect of father engagement on parent and family functioning.

6.4 Hypothesis 3

Parent distress, family functioning and fathers' engagement will have predictive value for the emotional and behavioural outcomes of the infants

The study results partially support the hypothesis. The qualitative data analysis results demonstrate that family demographics, infant illness characteristics and parent perceptions of the impact of the illness on the family are predictive of infant emotional and behavioural outcomes. The combined quantitative and qualitative data analysis demonstrated that parent reports of greater disruption to the family are a risk for worse infant emotional and behavioural outcomes. The study results do not indicate an effect of parent psychological symptoms or father engagement on infant emotional and behavioural outcomes.

6.4.1 Demographic and Illness Characteristics as Predictors of Infant Emotional and Behavioural Outcomes

6.4.1.1 Demographic characteristics

The quantitative analysis of fathers' data identified lower SES as a significant predictor of worse infant emotional outcomes. The hierarchical regression model for mothers did not identify any demographic characteristics as significant predictors of infant emotional or behavioural outcomes.

The differences between mothers' and fathers' results are intriguing. If SES affects infant emotional outcomes it could be expected to do so regardless of the informant, given that it is a shared characteristic for all family members. However, it is possible that SES has a differential influence on fathers' and mothers' perceptions of their

infant's emotional and behavioural functioning. In the current study, it appears that in the lower SES families, fathers perceive greater emotional and behavioural problems in their infants than do the mothers. Such an effect could be accounted for by gender differences. For example, mothers may make greater allowance for the illness in comparison with fathers' assessments when parents consider their infant's emotional functioning. It is also possible that the difference is spurious. In the initial stage of building the multiple regression models, SES had similar beta levels for fathers and mothers (fathers' β = -.398; mothers' β = -.304). It is important not to overestimate the gender difference. A larger sample size could provide sufficient power to clarify whether or not an effect exists.

As previously noted, there is conflicting evidence of the effect of SES on child outcomes following liver or solid organ transplantation.[142,163,191,193,194,197] While SES has been associated with emotional and behavioural problems in children in the general population,[97,199] varied study findings in organ transplantation groups may reflect differences in the measures used. Some researchers have used SES measures[193,194], while others have used family income[191,197] or parent education level[142,163] as proxies for SES. Likewise, different researchers have used different outcome measures, including measures of psychological symptoms in children[163,193,194,197] or HRQOL measures.[142,191]

In summary, SES was a significant predictor of child emotional outcomes in the quantitative analysis of fathers' data but not of mothers' data. The finding may be spurious or may indicate differences in parent perceptions of infant emotional and behavioural functioning according to parent gender. Existing research has reported conflicting findings. There is a need for greater consistency in the choice of measure of SES and of outcome measures in future research to clarify the effects of

demographic variables on the emotional outcomes of infants who have serious liver disease.

6.4.1.2 Illness characteristics

The quantitative data analysis demonstrated that an infant diagnosis of serious liver disease other than Biliary Atresia was predictive of worse infant emotional and behavioural outcomes. The result is supported by the findings of one previous study. Bucuvalas and colleagues reported better emotional functioning in children who had had a liver transplant for treatment of Biliary Atresia compared with children who had a transplant for another liver diagnosis.[142] However, that study was conducted with older children who had had a transplant almost six years previously on average (M = 5.8 years, SD = 3.4 years) and the children with Biliary Atresia had had a transplant earlier than the other children, leading to a greater length of time between transplantation and study participation in the Biliary Atresia group. The favourable outcome in the children with Biliary Atresia may therefore have been due to factors other than the illness itself. The current study demonstrates a differential effect of Biliary Atresia on infant emotional and behavioural outcomes compared with other liver diseases while controlling for length of time since diagnosis. It is possible that there are early differences in emotional outcomes according to diagnosis that persist over time. There are several possible reasons for differences in emotional outcomes between diagnoses. Biliary Atresia has a corrective surgical procedure that may postpone or even prevent the need for a liver transplant in the future. Having a surgical procedure available may offer hope to parents, or reduce feelings of powerlessness or uncertainty, leading to a lower emotional impact of the illness on parents and better emotional outcomes for the infants. Secondly, there are differences in morbidity between different diagnoses as previously noted, possibly leading to varied emotional and behavioural outcomes in the infants.

However, the current study results are in conflict with other research that found no differences in HRQOL following liver transplantation for Biliary Atresia compared with other serious liver disease.[170,180,185] The differing results between studies may be due to the different measures used. Given the conflicting findings in the published studies to date, further research will be required to assess the reasons for this finding.

The current study also demonstrated that greater illness severity was a significant predictor of worse infant emotional and behavioural outcomes. The illness severity measure predicting infant outcomes differed between parents. For mothers, the predictive severity measure was higher rates of outpatient clinic visits, while for fathers the predictive measure was whether the infant had had a liver transplant. The result is supported by prior research in infants with liver disease,[17] and following transplantation in older children.[139]

However, other research has not found an association between severity of illness and child adjustment in children with liver disease.[143,195] Likewise, severity of illness at the time of liver transplantation has not been demonstrated to affect later child HRQOL,[180] or child psychological outcomes following heart transplantation.[193,194]

Different studies have used varied measures of severity and outcomes. The pathways between illness severity and child emotional outcomes remain unclear, but could be related to infant and child distress due to a range of psychosocial stressors such as lengthy periods of hospitalisation, repeated traumatic medical and surgical procedures, altered family relationships, and lack of opportunities for normative social activities and relationships.

6.4.2 Impact of the Illness on the Family

The hierarchical multiple regression analysis demonstrated that parent perceptions of the impact of the illness on the family at Time 1 significantly predicted infant

emotional and behavioural outcomes at Time 2. Mothers' IFS scores contributed 11% to the variation in infant outcomes, while fathers' IFS scores contributed 15% to the variation. There were no problems with collinearity on hierarchical multiple regression analysis, indicating that the parent ratings of the impact of the illness on the family were independent of other variables that might be expected to lead to higher parent ratings of family impact, such as illness severity.

As noted previously, fathers' IFS scores were significantly lower than those found in previous research in families of children with chronic illness. Despite this, fathers' IFS scores were significant predictors of infant emotional and behavioural outcomes. The finding suggests that fathers may underestimate or under-report the impact of the illness on the family. Consideration should therefore be given to using a lower cut-off score for fathers' IFS scores than for mothers' scores when determining which infants may be at risk for poor emotional and behavioural outcomes.

The integrated data analysis supported the quantitative analysis, demonstrating that parent reports of greater difficulty adjusting to the disruption of the illness on the family are a risk for poorer emotional and behavioural outcomes for the infants.

The study results extend previous research findings. Only one previously published study has examined associations between parent IFS scores and child emotional and behavioural functioning in children with liver disease. In a cross-sectional study, Kaller and colleagues found a significant correlation between greater family strain, measured by the IFS, and worse psychosocial functioning in children who had had a liver transplant.[189] The current study provides further evidence for the link, but extends it to the younger age group and to children with either liver disease or who have had a transplant. A larger sample size would be required to assess whether liver disease and liver transplantation have differential effects on parent reports of the impact of the illness on the family.

Other researchers have used different measures of family functioning that may also contain elements of family impact of illness. Paediatric liver transplantation research has demonstrated an association between disruption to family routines and poorer HRQOL in children who have had a transplant,[188] though the research used a measure of family adaptation that had not been validated. Other organ transplantation research has demonstrated a significant correlation between poor family functioning (including family problem solving, organisation, and emotional climate) and poor psychological functioning in children and teenagers who have had a heart transplant.[193,194] Family conflict is also associated with worse emotional outcomes in adolescents who have had a liver transplant[190] or solid organ transplant.[191,192] It is possible that family conflict is a marker for poor family adjustment to transplantation. As previously noted, the current study did not demonstrate problems in general family functioning or predictive effects of family functioning on infant outcomes. However, different measures were used in the current study compared with other studies. Future research should examine the impact of the illness on the family rather than focussing on family functioning per se. Consistency in research approaches would allow greater comparability between studies.

6.4.3 Parent Psychological Symptoms, Family Functioning and Father Engagement as Predictors of Infant Emotional and Behavioural Outcomes

The quantitative analysis did not demonstrate significant predictor effects of parent psychological symptoms, general family functioning or father engagement for infant emotional and behavioural outcomes.

As noted above, the lack of demonstration of psychological symptoms in parents or alterations in general family functioning may reflect the choice of study instruments.

Measures of impact of the illness on parent emotions may have demonstrated an effect on infant outcomes, as has been demonstrated in solid organ transplantation.[140] It is of interest that the family impact measure used in the current study, the IFS, significantly predicted infant outcomes while the measure of family functioning did not, providing further evidence that measuring the impact of illness is more important than measuring psychological symptoms in parents or overall family functioning.

The lack of demonstration of an effect of father engagement on infant outcomes is contrary to the study hypothesis. Although the qualitative data regarding fathers was limited, the data provided further support for the quantitative results. As previously noted, there is a paucity of research examining father engagement in paediatric chronic illness, though there is evidence of a link between greater father engagement and improved HRQOL in adolescents with chronic illness.[204] It is possible that father engagement differs between families who have younger children compared with families who have adolescent children. Fewer differences in father engagement between families with younger children could therefore potentially mask an effect that would become evident during a longer period of follow up. It is also possible that the current study was not sufficiently powered to be able to demonstrate an effect of father engagement.

The mean CBCL Total Problems scores for the infants in the current study were in the normative range (Appendix E). The finding is at odds with research in older children and adolescents who have had a liver transplant, which demonstrated high Internalizing Problems and Total Problems scores in these children even after the somatic symptom items were removed.[89] The CBCL was used in the current study when the infants were at least 18 months old, which is the lower age limit of validity

of the measure. Longer-term follow up may demonstrate different findings over time as problems may further develop with increasing length of illness.

6.5 Strengths of the Current Study

The current study has several strengths that make a valuable contribution to the existing research base. The longitudinal design allows examination of predictive factors for family and infant outcomes. The inclusion of both parents and the mixed methods design provide a more complete picture of parent experiences of dealing with the diagnosis of serious liver disease in infancy. Standardising the research to families of infants soon after diagnosis controls for the developmental stage of the infants and the period of the illness cycle. Similarly, restricting the research to intact families allows examination of the role of father engagement while controlling for the presence of fathers in the family. This study is the first to examine the interplay between parent distress, family adjustment, and father engagement in relation to the emotional and behavioural outcomes of infants with a serious chronic illness. The research therefore considerably adds to the current knowledge base.

6.6 Limitations of the Current Study

A limitation of the current study is that the sample size was only sufficient to detect outcomes with a moderate effect size. It is therefore possible that clinically significant effects were not detected. However, serious liver disease in infancy is rare, requiring prolonged periods of data collection to recruit sufficient numbers of participants, particularly in a country with a relatively small population. Recruitment was compromised by unexpectedly low rates of new diagnoses at the initial recruitment sites, necessitating expansion of the study to other hospitals in Australia. Delays in expanding the study to other treatment centres resulted in a smaller sample size than could have been generated. In retrospect, a multi-centre design from the beginning of

the study would have maximised recruitment and strengthened the results. However, the sample size is comparable with other international research examining serious liver disease in children, and the prospective study design that includes both quantitative and qualitative measures mitigates the effects of the limitation.

While the participation rate of 65% is lower than desirable, it is not unusual in psychosocial research and is understandable given the participant burden for families who are under considerable stress. It is possible that parents who were more distressed or who had more problems adjusting to their infant's diagnosis declined to participate in the research. Unfortunately, limited information was available about non-participants, so it was not possible to determine whether non-participants differed from participants. However, the study had a high follow up rate of 88%, which may have resulted from the study design. The author personally contacted all eligible participants by telephone and personally interviewed all participants at the place and time of their choice. Providing maximal convenience for participants is likely to have assisted with the retention of participants at follow up. Having made an interpersonal connection with participants through the parent interviews is also likely to have improved retention. Some parents commented that they appreciated the opportunity to discuss their experiences.

The range of time periods between infant diagnosis and study participation was high, despite the attempts to standardise study timing. Some families were difficult to contact and some of the participating hospitals did not promptly inform the author of newly diagnosed infants. It is possible that some parents delayed participating until they had better adjusted, potentially leading to bias in the sample. Future research would benefit from a more robust system for identifying possible cases at the time of diagnosis and from attempts to engage parents in the study at an earlier time point leading up to the preferred time of participation.

The generalizability of the study results is limited due to the exclusion of separated families. However, as previously stated, this design allowed standardisation of the presence of fathers in the family and an examination of differences between families with varying father engagement.

Generalizability to migrant groups is also limited due to the requirement for parents to have adequate English language skills to be able to complete the study questionnaires. Although the study sample included migrant families, 12 families were not eligible to participate due to inadequate English. Parents with poor English language skills and their children may be at greater risk of poor psychosocial outcomes due to social isolation compared with other groups. Future research should examine differences between migrant and non-migrant groups.

The study would have been improved by including specific questions in the parent interviews about father engagement and infant emotional outcomes. It is possible that exploration of these issues at interview would have provided a more nuanced understanding of the interaction between fathers' engagement and the other study measures.

The study design included parent self-report measures of the impact of the illness on the family (measured by the IFS) which were predictive of the parents' reports of their infants' emotional outcomes (measured by the CBCL). As noted in Chapter 1, Introduction, use of parental reports in this age group is unavoidable. However, it is possible that parents who experienced the infants' illness as having a higher impact on the family were also more likely to rate their infants' behaviour as more problematic.

As noted in Chapter 1, Introduction, the context of data collection may affect the way in which emotional issues are reported. Conducting the research interviews in a variety of settings, including the family home and the hospital, may have led to

differences in parent reports of the emotional and family impact of the infants' illness depending on the location of the interview. However, as stated above, providing parents with the opportunity to participate in the location of their choice is likely to have improved the study retention rate. A larger sample size would be required to determine the importance of interview location in this context.

It was not possible to include representation in the qualitative analysis across the full range of scores on the study measures. For example, the qualitative analysis included more parents whose infants were below the group mean on the severity measures than parents whose infants were above the mean on these measures. However, in line with usual practice, coding of interview transcripts continued until saturation was reached and the remaining transcripts were reviewed to ensure that no additional themes had been missed. It is unlikely that important information was omitted from the analysis.

Finally, the author is a clinician at one of the research hospitals and was involved in the clinical care of half of the study participants at that site. Participants may have presented different information at this site as a result. It is possible that infant outcomes were different between sites due to differences in assessment and treatment approaches between sites. However, some of the study sites had low numbers of participants and it was not possible to assess differences between sites as a result.

6.7 Clinical Implications and Future Directions

This study has identified several risk factors for poor emotional and behavioural outcomes in infants with serious liver disease. The results suggest that screening families three months following their infant's diagnosis will identify families whose infants could benefit from early intervention.

Screening should consist of asking parents about the adequacy of their extended family support, ascertaining the presence of additional stressors (especially financial stress), and asking parents to complete the 15-item IFS scale. It is recommended that parent mean IFS scores from the current study should be used to identify families at risk, equating to scores above 34 for fathers and scores above 37 for mothers. Such screening could be undertaken by the clinic nurse at a routine clinic visit and would be time-efficient and simple to implement. Families who report lack of extended family support or additional stressors, or families with at least one parent scoring above the cut-off score on the IFS, should be referred to the Social Worker for assessment. Referral for more specialised mental health care could be made following Social Worker assessment if required. Although it would be difficult to measure the efficacy of such an approach, the acceptability of the approach to families and staff could be easily assessed.

The qualitative data revealed the importance to parents of the uncertainties inherent in their infants' illness, heightened by the recognition of the infants' vulnerability. In addition, parents discussed the importance of their trust in the treating team. The findings suggest the importance of maintaining continuity of providers to ensure consistency in service delivery. Such an approach provides opportunities for facilitating communication and parent education. Fostering trusting relationships between the treating team and parents is likely to be reassuring for parents. Contact with other families who have an infant or child with serious liver disease should be encouraged as a means of support for families whose infants have been recently diagnosed. However, treating teams should be aware of such relationships so that additional support can be provided to families if the other children suffer adverse events.

Future research into the impact of serious liver disease in infancy should take a longitudinal approach and focus on longer-term child emotional outcomes. Incorporation of a social support measure, a standardised measure of stressors and a measure of the emotional impact of the illness on parents (rather than a psychological symptom measure) will be important in further assessing the importance of these factors on infant emotional outcomes. Direct observational measures of infant emotional outcomes would provide a clearer picture of infant emotional functioning, but would be more difficult and costly to undertake. Given the rarity of serious liver disease in infants, multi-centre studies are required to generate sufficient data to demonstrate smaller, but clinically important effects.

6.8 Conclusions

This study identifies early risk factors for poor emotional and behavioural outcomes in infants who have been diagnosed with serious liver disease. The study demonstrates that infant outcomes are predicted by parent perceptions of the impact of the illness on the family, mediated by lack of extended family support and financial stress. The results also indicate differential emotional effects on mothers and fathers soon after the infants' diagnosis, which is sustained over the following 12 months. In addition, characteristics of the illness, including severity and primary liver diagnosis, are predictive of infant emotional and behavioural outcomes.

The study provides a detailed understanding of parent experiences following the diagnosis of serious liver disease in infancy. The experience is characterised by uncertainty, awareness of the infant's vulnerability, and feelings of isolation. Dealing with other life pressures can add to parent distress, but can also provide a welcome distraction. The shared experience of the illness typically strengthens family relationships, and meeting other families who have a child with liver disease is also a helpful source of support and understanding for parents. However, poor illness outcomes in other children are highly distressing for parents. Many parents describe feeling that the illness experience results in an altered perspective on life.

This is the first study to prospectively examine the interactional effects of parent and family factors on infant emotional and behavioural outcomes following the infant's diagnosis of serious liver disease. Most previous research has focussed on the post-transplant period and has been cross-sectional in design, limiting the capacity to identify predictive factors for poor emotional and behavioural outcomes for these children. The current study is unique in its inclusion of both parents and the combined use of self-report measures and parent interviews.

Previous research examining families of children who have received a transplant demonstrated high rates of emotional and behavioural problems in children, persisting parent emotional distress, and disruption for families. The current study extends the results to the period following initial diagnosis and to the infant age group.

Future research should include measures of emotional impact, stressors and social supports, and should focus on children's longer-term outcomes.

Taken together, the study results have implications for clinical practice. The results demonstrate that families who report financial stress, who lack extended family support, or who report a greater impact of the infant's illness on the family should be referred for psychosocial assessment. Screening families soon after the infant's diagnosis rather than waiting until the infant is being evaluated for liver transplantation will provide the opportunity for early intervention for vulnerable families.

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Appendix A
Ethics Approval Letters

The University of Sydney

Human Research Ethics Committee
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24 June 2009

Professor Philip Hazell
Psychological Medicine
Concord Clinical School
Thomas Walker Hospital – G03
The University of Sydney
Email: phazell@med.usyd.edu.au

Dear Professor Hazell

Title: **Family Functioning, Parent Stress and Child Outcomes in serious paediatric Liver Disease: A Prospective Longitudinal Study (Ref. No.12000)**
PhD Student: Dr Michael Bowden

Your application was reviewed by the Executive Committee of the Human Research Ethics Committee (HREC), and in doing so has ratified your study to include the PhD student – Dr Michael Bowden.

The Executive Committee acknowledges your right to proceed under the authority of *SSWAHS Human Research Ethics Committee - CRGH.*

Please note, this ratification has been given only in respect of the ethical content of the study.

Any modifications to the study must be approved by *SSWAHS Human Research Ethics Committee - CRGH* before submission to the University of Sydney Human Research Ethics Committee.

Yours sincerely

Marietta Coutinho
Deputy Manager
Human Research Ethics Administration

cc Dr Michael Bowden, Department of Psychological Medicine, Children's Hospital at Westmead – C29 [Email: michael.bowden@nswiop.nsw.edu.au]

Associate Professor Andrew Day, Department of Gastroenterology, Sydney Children's Hospital, High St, Randwick NSW 2021 [Andrew.day@unsw.edu.au]

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Our Ref: *(08/CRGH/153)*

CONCORD
REPATRIATION GENERAL
HOSPITAL

24 November, 2008

Dr Michael Bowden
Department of Psychological Medicine
Children's Hospital at Westmead
Locked Bag 4001
WESTMEAD NSW 2145

Dear Dr Bowden,

Re: 08/CRGH/153 CH62/6/2008-124
Family Functioning, Parent Stress and Child Outcomes in serious paediatric Liver Disease: A Prospective Longitudinal Study

Thank you for submitting the above multi-centre project for single ethical and scientific review. This project was first considered by the Sydney South West Area Health Service Human Research Ethics Committee – CRGH Zone at its meeting held on 25 September 2008. This Human Research Ethics Committee (HREC) has been accredited by the NSW Department of Health as a lead HREC under the model for single ethical and scientific review.

This lead HREC is constituted and operates in accordance with the National Health and Medical Research Council's *National Statement on Ethical Conduct in Human Research* and the *CPMP/ICH Note for Guidance on Good Clinical Practice.*

I am pleased to advise that the Committee has granted ethical approval of this research project. The documents reviewed and approved include:

Participant Information Sheet (The Children's Hospital Westmead) – Version 2 dated 3 November 2008.
Participant Consent Form – (The Children's Hospital Westmead) – Version 2 dated 3 November 2008.
Participant Information Sheet (Sydney Children's Hospital) – Version 2 dated 3 November 2008.
Participant Consent Form – (Sydney Children's Hospital) – Version 2 dated 3 November 2008.
Qualitative Interview Questions – Version 1 dated 19 August 2008.

Please note the following conditions of approval:

1. You will immediately report anything which might warrant review of ethical approval of the project in the specified format, including:
   - unforeseen events that might affect continued ethical acceptability of the project, (including Serious Adverse Events).

2. Proposed changes to the research protocol, conduct of the research, or length of HREC approval will be provided to the HREC for review in the specified format.

3. The HREC will be notified, giving reasons, if the project is discontinued at a site before the expected date of completion.

4. You will provide an annual report to the HREC, and at completion of the study in the specified format.

5. You will adhere to the study protocol at all times.

HREC approval is valid for four (4) years subject to the supply of an annual progress report. The first report should be sent to the Concord Hospital Research Office by **30/11/2009.**

Should you have any queries about the HREC's consideration of your project please contact the Executive Officer - Ms Virginia Turner on (02) 9767-5622. The HREC Terms of Reference, Standard Operating Procedures, membership and standard forms are available from the website: www.sswahs.nsw.gov.au/concord/ethics .

You are reminded that this letter constitutes ethical approval only. You must not commence this research project at a site until separate authorisation from the Chief Executive or delegate of that site has been obtained.

Please forward a copy of this letter to all site investigators for submission to the relevant Research Governance Officer.

We wish you every success in your research.

Please quote the above file number in all correspondence.

Yours sincerely,

Dr Garry Pearce
Chairman
SSWAHS Human Research Ethics Committee – CRGH

☐ ***Please complete and return a copy of this page to the Concord Hospital Research Office as acknowledgment of your acceptance of the Conditions of Ethical Approval.***

M. BOWDEN — **Printed Name**
Chief Investigator

Signature

1/12/08 — **Date**

the children's hospital at Westmead

Research and Development
Contact for this correspondence:
Name: Carolyn Casey
Email: CarolyB3@chw.edu.au
Phone: (02) 9845 3017
Facsimile: (02) 9845 1317

Corner Hawkesbury Road
and Hainsworth Street
Locked Bag 4001
Westmead NSW 2145
Sydney Australia
DX 8213 Parramatta
Tel +61 2 9845 0000
Fax +61 2 9845 3489
www.chw.edu.au
ABN 53 188 579 090

JMG:\DATA\Research\GOVERNANCE-SSA Applications\Approval Letters & Memos\09.CHW.11 Ltr to Dr M Bowden.rtf

30 January 2009

Dr Michael Bowden
Psychological Medicine

Dear Dr Bowden,

HREC reference number: 08/CRGH/153
SSA reference number: SSA/09/CHW/11
Project title: Family Functioning, Parent Stress and Child Outcomes in serious paediatric Liver Disease: A Prospective Longitudinal Study

Thank you for submitting an application for authorisation of this project. I am pleased to inform you that authorisation has been granted for this study to take place here at The Children's Hospital at Westmead.

The following conditions apply to this research project. These are additional to those conditions imposed by the Human Research Ethics Committee that granted ethical approval:

1. Proposed amendments to the research protocol or conduct of the research which may affect the ethical acceptability of the project, and which are submitted to the lead HREC for review, are copied to the research governance officer;

2. Proposed amendments to the research protocol or conduct of the research which may affect the ongoing site acceptability of the project are to be submitted to the research governance officer.

Yours Sincerely,
per
Ms Carolyn Casey
Research Governance Officer

Date: 20/08/2008 Reference: 08/CRGH/153 Online Form

(f) Authorisation by Chief Executive (or delegate)

HREC Reference number:	08/CRGH/153
Project Title (in full):	Family Functioning, Parent Stress and Child Outcomes in serious paediatric Liver Disease: A Prospective Longitudinal Study
Principal Investigator:	Dr Michael Bowden

This research is: ☑ authorised ○ not authorised

Specify, conditions applying to authorisation or reasons for not authorising.

My signature indicates that I authorise/ do not authorise this research project to commence at this site.

Name of Chief Executive (or delegate): Chris Cowell
Name of Organisation: CHW
Date: 29/1/09
Signature: .....

Version 1.0 59 C/5143/7940/1

the children's hospital at Westmead

Corner Hawkesbury Road
and Hainsworth Street
Locked Bag 4001
Westmead NSW 2145
Sydney Australia
DX 8213 Parramatta
Tel +61 2 9845 0000
Fax +61 2 9845 3489
www.chw.edu.au
ABN 53 188 579 090

INFORMATION SHEET

Family Risk and Resilience in Paediatric Liver Disease

Principal Investigators:
Dr M Bowden, Dept of Psychological Medicine, CHW, ph: 9845 2005
A/Prof A Day, Dept of Gastroenterology, SCH, ph: 9382 1752

Associate Investigators:
Dr M Stormon, Dept of Gastroenterology, CHW, ph: 9845 3999
Dr T O'Loughlin, Dept of Gastroenterology, CHW, ph: 9845 3999
Dr D Lemberg, Dept of Gastroenterology, SCH, ph: 9382 1752
Dr R Jackson, Dept of Gastroenterology, SCH, ph: 9382 1752
Ms V Jermyn, Dept of Gastroenterology, CHW, ph: 9845 3999
Ms G Tomsic, Dept of Gastroenterology, CHW, ph: 9845 3999
Ms D Carmody, Dept of Social Work, CHW, ph: 9845 2623
Mr N Teofilovic, Dept of Psychological Medicine, CHW, ph: 9845 2005

We would like you to consider participating in a research study that will be conducted in the Department of Psychological Medicine at The Children's Hospital at Westmead (CHW), in conjunction with the Department of Gastroenterology at Sydney Children's Hospital (SCH) at Randwick.

What is the study about?
This study is trying to find out how your child's liver disease affects you as parents and the relationships within the family and how these are related to the physical and psychological health outcomes for your child. The information we get from this study will help us to develop our services to best meet the needs of families who have children with liver disease.

Who can participate in the study?
We are inviting all families who have an infant diagnosed with liver disease at the Children's Hospital at Westmead or the Sydney Children's Hospital at Randwick to participate in this study.

What will the study involve?
If you decide to participate in the study, this is what we will ask you to do:

1. Complete and sign the consent form and return it to your doctor.
2. Dr Michael Bowden will contact you to arrange a convenient time for a family interview to take place.
3. We will send you four (4) questionnaires for each parent to complete before the interview. These will take approximately 45 minutes to complete. There are no right or wrong answers; the questionnaires ask about your feelings, your experiences of having a child with a physical illness and about your family.
4. If you would like to participate in the study but have trouble completing the questionnaires, please call Dr Michael Bowden for help (02 9845 2005). If you prefer, you can bring the questionnaires to your hospital appointment and we will help you to fill them in or arrange an interpreter if needed.
5. All family members are invited to the interview. Dr Michael Bowden will talk to you about the relationships in the family, about your experiences of having a child with liver disease and

any other concerns you may have. This interview takes about 45 minutes to one hour and will be taped so that it can be accurately recorded and analysed.

6. About 12 months after your child's diagnosis, Dr Michael Bowden will contact you again to arrange a follow up interview. You will be sent the same questionnaires as previously, with one additional questionnaire that asks about your child's behaviour and feelings, which takes about 20 minutes to complete. The interview will be the same as previously.
7. We will be happy to give you feedback of your results at the end of the study if you wish.
8. You and your family will receive the usual clinical assessment, care and treatment from your treating health care team.
9. This is all that you will be asked to do, though we may ask to follow up again at a later date. If so, we will ask you if you wish to participate then and will give you full details. There will be no requirement for you to participate further if you do not wish to.

Are there any benefits for my child participating in the study?
There are no known benefits for your child in participating in this study. We hope that the results from this study will help us to learn more about the support needs of the families of children we care for.

Are there any side-effects and risk associated with this study?
There are no known side-effects or risks associated with participation in this study. We expect that the time needed to complete the questionnaires and the interviews is the only cost to you in taking part in this study. Talking about difficult health issues in your child may be distressing. If so, we will provide de-briefing or counselling and will discuss with you whether you would like a referral for further assistance.

Other information
Your confidentiality will be maintained throughout the study period by not using your or your child's name on any study documents. We need to keep track of your information and will identify your data only with your child's medical record number and date of birth. Only group data will be reported so that individuals will not be able to be identified in any publication that may arise from the study.

The audiotape of your interview will be transcribed and then the tape will be destroyed. The study data, including the transcribed data, will be stored in a locked cabinet that is only accessible to the study group. It will be stored for a period of seven (7) years after completion of the study to comply with Hospital guidelines. After this it will be destroyed through a secure document destruction company employed by the Children's Hospital at Westmead.

Participation in this project is entirely voluntary. If you decide not to take part or decide to withdraw at any time, please be assured that this will not affect your child's care at the Hospital or your relationship with Hospital staff.

If you have any questions about the conduct of this study, please do not hesitate to discuss them with the principal investigators, Dr Michael Bowden (02 9845 2005) or A/Prof Andrew Day (02 9382 1752).

This project has been approved by the Ethics Committee of Sydney South West Area Health Service – Concord Repatriation General Hospital. If you have any concerns or complaints about the conduct of the study, you may contact the Secretary of the Ethics Committee on 02 9767 5622. Alternatively, if you would like to speak to someone at the Children's Hospital at Westmead, you may contact the Research Governance Officer, Carolyn Casey on 02 9845 1316.

This Information Sheet is for you to keep. We will also give you a copy of the signed consent form.

the children's hospital at Westmead

Corner Hawkesbury Road
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Fax +61 2 9845 3489
www.chw.edu.au
ABN 53 188 579 090

PARTICIPANT CONSENT FORM

Family Risk and Resilience in Paediatric Liver Disease

I _____(name) of

_____(address)

have read and understand the Information Sheet for the above named research study, and have

discussed the study with _____

- I have been made aware of the procedures involved in the study, including any known or expected inconvenience, risk, discomfort or potential side effect and of their implications as far as they are currently known by the researchers.
- I understand that during the course of this study, my medical records may be accessed by the researchers, by regulatory authorities or by the Sydney South West Area Health Service – Concord Repatriation General Hospital in order to verify results and determine that the study is being carried out correctly.
- I freely choose to participate in this study and understand that I can withdraw at any time and this decision will not otherwise affect my or my child's involvement with the Children's Hospital at Westmead or with the Sydney Children's Hospital.
- I also understand that the research study is strictly confidential.
- I hereby agree to participate in this research study.

Name of Child (Please Print): _____

Name of Parent/s (Please Print): _____

Signature of Parent/s: _____Date: _____

Name of Person who Conducted Informed Consent Discussion (Please Print):

_____

Signature: _____ Date: _____
of Person who Conducted Informed Consent Discussion

SOUTH EASTERN SYDNEY
ILLAWARRA
NSW HEALTH

RESEARCH SUPPORT OFFICE - NORTHERN HOSPITAL NETWORK

Room G71, East Wing
Edmund Blacket Building
The Prince of Wales Hospital
Cnr High & Avoca Sts
RANDWICK NSW 2031
Tel: (02) 9382 3587
Fax: (02) 9382 2813

22 April 2009

Dr Andrew Day
Department of Gastroenterology
Sydney Children's Hospital
High Street
RANDWICK NSW 2031

Dear Dr Day

RE: SSA Reference: 08/G/222
HREC / AU RED Reference: 08/CRGH/153 CH62/6/2008
Project Title: Family functioning, Parent Stress and Child Outcomes in serious paediatric liver disease: A Prospective Longitudinal Study.

I refer to your Site Specific Assessment application for the above titled project. I am pleased to advise that on 22 April 2009 the Acting Executive Director Sydney Children's Hospital granted authorisation for the above project to commence at Sydney Children's Hospital.

The following conditions apply to this research project. These are additional to any conditions imposed by the Human Research Ethics Committee that granted ethical approval:

1. Proposed amendments to the research protocol or conduct of the research which may affect the ethical acceptability of the project, and are submitted to the lead HREC for review, are copied to the Research Governance Officer.

2. Proposed amendments to the research protocol or conduct of the research which may affect the ongoing site acceptability of the project are to be submitted to the Research Governance Officer.

If you have any queries relating to the above please contact the Research Support Office on 9382 3587.

Yours sincerely

Ms Marie Malica
Manager, Research Support Office
Research Governance Officer

SSA 2008-222 - Dr Andrew Day - Approval ltr 22-4-2009.doc

South Eastern Sydney and Illawarra Area Health Service
Locked Mail Bag 8808 South Coast Mail Centre NSW 2521
Level 4 Lawson House Wollongong Hospital
Tel (02) 4253 4888 Fax (02) 4253 4878
ABN 78 390 886 131

INFORMATION SHEET

Family Risk and Resilience in Paediatric Liver Disease

Principal Investigators:
Dr M Bowden, Dept of Psychological Medicine, CHW, ph: 9845 2005
A/Prof A Day, Dept of Gastroenterology, SCH, ph: 9382 1752

Associate Investigators:
Dr M Stormon, Dept of Gastroenterology, CHW, ph: 9845 3999
Dr T O'Loughlin, Dept of Gastroenterology, CHW, ph: 9845 3999
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Dr R Jackson, Dept of Gastroenterology, SCH, ph: 9382 1752
Ms V Jermyn, Dept of Gastroenterology, CHW, ph: 9845 3999
Ms G Tomsic, Dept of Gastroenterology, CHW, ph: 9845 3999
Ms D Carmody, Dept of Social Work, CHW, ph: 9845 2623
Mr N Teofilovic, Dept of Psychological Medicine, CHW, ph: 9845 2005

We would like you to consider participating in a research study that will be conducted in the Department of Psychological Medicine at The Children's Hospital at Westmead (CHW), in conjunction with the Department of Gastroenterology at Sydney Children's Hospital (SCH) at Randwick.

What is the study about?
This study is trying to find out how your child's liver disease affects you as parents and the relationships within the family and how these are related to the physical and psychological health outcomes for your child. The information we get from this study will help us to develop our services to best meet the needs of families who have children with liver disease.

Who can participate in the study?
We are inviting all families who have an infant diagnosed with liver disease at the Children's Hospital at Westmead or the Sydney Children's Hospital at Randwick to participate in this study.

What will the study involve?
If you decide to participate in the study, this is what we will ask you to do:

1. Complete and sign the consent form and return it to your doctor.
2. Dr Michael Bowden will contact you to arrange a convenient time for a family interview to take place.
3. We will send you four (4) questionnaires for each parent to complete before the interview. These will take approximately 45 minutes to complete. There are no right or wrong answers; the questionnaires ask about your feelings, your experiences of having a child with a physical illness and about your family.
4. If you would like to participate in the study but have trouble completing the questionnaires, please call Dr Michael Bowden for help (02 9845 2005). If you prefer, you can bring the questionnaires to your hospital appointment and we will help you to fill them in or arrange an interpreter if needed.
5. All family members are invited to the interview. Dr Michael Bowden will talk to you about the relationships in the family, about your experiences of having a child with liver disease and any other concerns you may have. This interview takes about 45 minutes to one hour and will be taped so that it can be accurately recorded and analysed.

6. About 12 months after your child's diagnosis, Dr Michael Bowden will contact you again to arrange a follow up interview. You will be sent the same questionnaires as previously, with one additional questionnaire that asks about your child's behaviour and feelings, which takes about 20 minutes to complete. The interview will be the same as previously.
7. We will be happy to give you feedback of your results at the end of the study if you wish.
8. You and your family will receive the usual clinical assessment, care and treatment from your treating health care team.
9. This is all that you will be asked to do, though we may ask to follow up again at a later date. If so, we will ask you if you wish to participate then and will give you full details. There will be no requirement for you to participate further if you do not wish to.

Are there any benefits for my child participating in the study?
There are no known benefits for your child in participating in this study. We hope that the results from this study will help us to learn more about the support needs of the families of children we care for.

Are there any side-effects and risk associated with this study?
There are no known side-effects or risks associated with participation in this study. We expect that the time needed to complete the questionnaires and the interviews is the only cost to you in taking part in this study. Talking about difficult health issues in your child may be distressing. If so, we will provide de-briefing or counselling and will discuss with you whether you would like a referral for further assistance.

Other information
Your confidentiality will be maintained throughout the study period by not using your or your child's name on any study documents. We need to keep track of your information and will identify your data only with your child's medical record number and date of birth. Only group data will be reported so that individuals will not be able to be identified in any publication that may arise from the study.

The audiotape of your interview will be transcribed and then the tape will be destroyed. The study data, including the transcribed data, will be stored in a locked cabinet that is only accessible to the study group. It will be stored for a period of seven (7) years after completion of the study to comply with Hospital guidelines. After this it will be destroyed through a secure document destruction company employed by the Children's Hospital at Westmead.

Participation in this project is entirely voluntary. If you decide not to take part or decide to withdraw at any time, please be assured that this will not affect your child's care at the Hospital or your relationship with Hospital staff.

If you have any questions about the conduct of this study, please do not hesitate to discuss them with the principal investigators, Dr Michael Bowden (02 9845 2005) or A/Prof Andrew Day (02 9382 1752).

This project has been approved by the Ethics Committee of Sydney South West Area Health Service – Concord Repatriation General Hospital. If you have any concerns or complaints about the conduct of the study, you may contact the Secretary of the Ethics Committee on 02 9767 5622. Alternatively if you would like to speak to someone at Sydney Children's Hospital Randwick, you may contact the Research Governance Officer, Marie Malica on (02) 9382 3583.

This Information Sheet is for you to keep. We will also give you a copy of the signed consent form.

PARTICIPANT CONSENT FORM

Family Risk and Resilience in Paediatric Liver Disease

I _____(name) of

_____(address)

have read and understand the Information Sheet for the above named research study, and have

discussed the study with _____

- I have been made aware of the procedures involved in the study, including any known or expected inconvenience, risk, discomfort or potential side effect and of their implications as far as they are currently known by the researchers.
- I understand that during the course of this study, my medical records may be accessed by the researchers, by regulatory authorities or by the Sydney South West Area Health Service – Concord Repatriation General Hospital in order to verify results and determine that the study is being carried out correctly.
- I freely choose to participate in this study and understand that I can withdraw at any time and this decision will not otherwise affect my or my child's involvement with the Children's Hospital at Westmead or with the Sydney Children's Hospital.
- I also understand that the research study is strictly confidential.
- I hereby agree to participate in this research study.

Name of Child (Please Print): _____

Name of Parent/s (Please Print): _____

Signature of Parent/s: _____Date: _____

Name of Person who Conducted Informed Consent Discussion (Please Print):

_____

Signature: _____ Date: _____
of Person who Conducted Informed Consent Discussion

Family Risk and Resilience in Paediatric Liver Disease
Version 2, 03.11.2008

Page 1 of 1

Queensland Government

Queensland **Health**

CHILDREN'S HEALTH SERVICES

DISTRICT MANAGEMENT

Enquiries to: Linda Hardy
Telephone: 3636 7591
Facsimile: 3636 7215
Our Ref:

8 December 2010

Dr Michael Bowden
The Children's Hospital
Locked Bag 4001
WESTMEAD NSW 2145

Dear Dr Bowden

Re: Family functioning, Parent Stress and Child Outcomes in serious paediatric liver disease

I am pleased to advise the above project was approved by the Royal Children's Hospital Executive on 6 December 2010.

Sincerely

Linda Hardy
Chief Operating Officer
Royal Children's Hospital
Children's Health Services
Cc Dr Looi Ee

Office
Chief Operations Manager
Level 5 Woolworths Medical Bldg

Postal
Herston Rd
Herston Qld 4029

Phone
3636 8262

Fax
3636 7215

QLD CHILDREN'S HEALTH SERVICES (RCH)
HUMAN RESEARCH ETHICS COMMITTEE

Professor John Pearn (Chair) 3365 5323
Mrs Amanda Smith (Co-ordinator) 3636 9167

Queensland Government

Queensland **Health**

Level 3, RCH Foundation Building
Royal Children's Hospital
Herston QLD 4029 Australia
Telephone (07) 3636 9167
Facsimile (07) 3365 5455

19th October 2010

Dr Michael Bowden
The Children's Hospital
Locked Bag 4001
Westmead NSW 2145

Dear Dr Bowden,

HREC Reference number: HREC/10/QRCH/94
Project title: Family Functioning, Parent Stress & Child Outcomes in Serious Paediatric Liver Disease: A Prospective Longitudinal Study.

Thank you for submitting the above project for ethical review.

This HREC is constituted and operates in accordance with the National Health and Medical Research Council's (NHMRC) *National Statement on Ethical Conduct in Human Research (2007), NHMRC and Universities Australia Australian Code for the Responsible Conduct of Research (2007)* and the *CPMP/ICH Note for Guidance on Good Clinical Practice.*

I am pleased to advise the proposal meets the requirements of the National Statement on Ethical Conduct in Human Research and the Committee is happy to give approval. The documents reviewed and approved include:

Document	*Version*	*Date*
Application		
Covering Letter		05 October 2010
Correspondence - Concord Repatriation General Hospital		15 September 2010
Correspondence - South Eastern Sydney Illawarra		22 April 2009
Correspondence - The Children's Hospital at Westmead		30 January 2009
Correspondence - Concord Repatriation General Hospital		24 November 2008
NEAF Application		
Questionnaire: Qualitative Interview Questions		
Patient Information Sheet/Consent Form: QCHS Info & Consent	6	04 October 2010
Patient Information Sheet/Consent Form: The Children's Hospital at Westmead	1	19 August 2008
Patient Information Sheet/Consent Form: Sydney Children's Hospital Randwick	1	19 August 2008

Please note the following conditions of approval:

1. We require an annual progress report (or sooner if the project is completed) concerning the study. This must include progress to date or outcome in the case of completed research. (In accordance with National Statement 5.5.3)

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2. In accordance with the National Statement (3.3.12), before beginning the clinical phase of the research, researchers should register clinical trials in a publicly accessible domain.

3. If the project does not proceed, the Committee must be informed as soon as possible. (In accordance with National Statement 5.5.6)

4. The Committee must be informed of any potential or realised problem with bioethical implications, if such occurs during the conduct of the research project.

5. Any serious adverse event (SAE) that arises in the context of this research, or involving a researcher conducting this research, must be reported to the Ethics Committee within 72 hours and reported to the sponsor (if applicable) within the stipulated time frame.

   Serious Adverse Event Reports that are generated off-site may be (a) Serious Unexpected Adverse Reactions or (b) Serious Events which the Research Team believes cannot be related to the research intervention. The Research team must report incidents of (a) during multi-centre trials. Such are required to be submitted to the Chair of the QLD Children's Health Services District Ethics Committee (RCH) on receipt by the researcher. A summary of the SAE reports is to accompany the submission. Information required includes; patient details (age & sex), adverse event, outcome and the likelihood of the event being related to the study drug/device/procedure.

   With respect to all SAEs, the researcher must provide his or her opinion as to whether the SAE is directly related to the research intervention. A copy of the SAE Summary must be provided. (This can be obtained from the Ethics Officer)

6. Amendments to the research project which may affect the ongoing ethical acceptability of a project must be submitted to the HREC for review. Major amendments should be reflected in a revised online NEAF (accompanied by all relevant updated documentation and a cover letter from the principal investigator, providing a brief description of the changes, the rationale for the changes, and their implications for the ongoing conduct of the study). Hard copies of the revised NEAF, the cover letter and all relevant updated documents with tracked changes must also be submitted to the HREC coordinator as per standard HREC SOP. Further advice on submitting amendments is available from: http://www.health.qld.gov.au/cpic/documents/ethics/researcher_userguide.pdf

7. The Ethics Committee will conduct a randomly identified audit of a proportion of research projects approved by the Committee. That audit process will look at such issues as;
   a. Security of Documents
   b. Consent Form Register
   c. Serious Adverse Events Register
   d. Withdrawal of Participants – who and why
   e. The de-identification of data

8. We require researchers to give a declaration of intention to publish their findings in a refereed journal or similar peer-reviewed forum. Your work must be in accordance with the following:

   - National Statement on Ethical Conduct in Human Research: http://www.nhmrc.gov.au/publications/synopses/_files/e72.pdf
   - Queensland Health Management Research Policy: http://www.health.qld.gov.au/cpic/documents/ethics/research_policy.pdf
   - Joint NHMRC / AVCC Statement and Guidelines on Research Practice (1997): http://www.nhmrc.gov.au/funding/policy/researchprac.htm
   - Declaration of Helsinki: http://www.health.qld.gov.au/ethics/Documents/24938_policy.pdf
   - Guidelines under Section 95 of the Privacy Act1995 and Guidelines approved under Section 95A of the Privacy Act 1995. http://www.comlaw.gov.au/ComLaw/Legislation/ActCompilation1.nsf/0/B471AB909A18D172CA25725C0083858A/$file/Privacy1988_WD02HYP.pdf
   - Queensland Health Privacy Guidelines IS42 & IS42A: http://qheps.health.qld.gov.au/privacy/resources.htm

9. Researchers should note, if not QLD Health employees, a Blue Card may be required for contact with children.

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10. The Researcher must send the **'Notification of Commencement of Research Protocol'** as soon as research begins. Status of the project will remain as 'Not Started' until this form is received.

Should you have any queries about the HREC's consideration of your project please contact Amanda Smith (Co-ordinator) or Professor John Pearn (Chairperson). The HREC terms of Reference, Standard Operating Procedures, membership and standard forms are available from: http://www.health.qld.gov.au/cpic/ethics/reagu_homepage.asp

You are reminded that this letter constitutes ethical approval only. You must not commence this research project at a site until separate authorisation from the District CEO or Delegate of that site has been obtained.

A copy of this approval must be submitted to the District Executive with a completed Institutional Approval Form for authorisation from the CEO or Delegate to conduct this research within the Children's Health Service District.

The HREC wishes you every success in your research.

With kind regards,

for Professor Alan Isles
Deputy Chair
Queensland Children's Health Services (RCH) Human Research Ethics Committee

Cc: Ethics Committee Files
Dr Looi Ee, Paediatric Gastroenterology, RCH

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Queensland Government
Queensland Health

QLD Children's Health Services (RCH)
Parent/Guardian Information Sheet

Project Title	***Family Risk and Resilience in Paediatric Liver Disease***
Investigators	*Dr Michael Bowden (CHW), Dr Usha Krishnan (SCH) and Dr Looi Ee (RCH)*
Contact Person	*Dr Michael Bowden*
Address	*Department of Psychological Medicine* *Children's Hospital Westmead* *Locked Bag 4001* *Westmead NSW 2145*
Phone Number	*02 9845 2005 (Dr Bowden)* *07 3636 7887 (Dr Ee)*

We would like you to consider participating in a research study that will be conducted in the Department of Psychological Medicine at The Children's Hospital Westmead, NSW (CHW), in conjunction with the Department of Gastroenterology at Sydney Children's Hospital Randwick, NSW (SCH) and the Paediatric Gastroenterology, Hepatology & Nutrition Service, Royal Children's Hospital, Brisbane, QLD (RCH).

What is the study about?
This study is trying to find out how your child's liver disease affects you as parents and the relationships within the family and how these are related to the physical and psychological health outcomes for your child. The information we get from this study will help us to develop our services to best meet the needs of families who have children with liver disease. The study forms part of a PhD project by Dr M Bowden.

Who can participate in the study?
We are inviting all families who have an infant diagnosed with liver disease at the Children's Hospital at Westmead, the Sydney Children's Hospital at Randwick and the Royal Children's Hospital at Brisbane to participate in this study.

What will the study involve?
If you decide to participate in the study, this is what we will ask you to do:

1. Complete and sign the consent form and return it to your doctor.
2. Dr Michael Bowden will contact you to arrange a convenient time for a family interview to take place.
3. We will send you four (4) questionnaires for each parent to complete before the interview. These will take approximately 45 minutes to complete. There are no right or wrong answers; the questionnaires ask about your feelings, your experiences of having a child with a physical illness and about your family.
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7. We will be happy to give you feedback of your results at the end of the study if you wish.
8. You and your family will receive the usual clinical assessment, care and treatment from your treating health care team.
9. This is all that you will be asked to do, though we may ask to follow up again at a later date. If so, we will ask you if you wish to participate then and will give you full details. There will be no requirement for you to participate further if you do not wish to.

Are there any benefits for my child participating in the study?
There are no known benefits for your child in participating in this study. We hope that the results from this study will help us to learn more about the support needs of the families of children we care for.

Are there any side-effects and risk associated with this study?
There are no known side-effects or risks associated with participation in this study. We expect that the time needed to complete the questionnaires and the interviews is the only cost to you in taking part in this study. Talking about difficult health issues in your child may be distressing. If so, we will provide de-briefing or counselling and will discuss with you whether you would like a referral for further assistance.

Other information
Your confidentiality will be maintained throughout the study period by not using your or your child's name on any study documents. We need to keep track of your information and will identify your data only with your child's medical record number and date of birth. Only group data will be reported so that individuals will not be able to be identified in any publication that may arise from the study, including in the PhD thesis that will result from this study. Research data may be accessed by auditors, ethics committee or regulatory authorities.

The audiotape of your interview will be transcribed and then the tape will be destroyed. The study data, including the transcribed data, will be stored in a locked cabinet that is only accessible to the study group. It will be stored for a period of seven (7) years after completion of the study to comply with Hospital guidelines. After this it will be destroyed through a secure document destruction company employed by the Children's Hospital at Westmead.

Participation in this project is entirely voluntary. If you decide not to take part or decide to withdraw at any time, please be assured that this will not affect your child's care at the Hospital or your relationship with Hospital staff.

If you have any questions about the conduct of this study, please do not hesitate to discuss them with the principal investigators, Dr Michael Bowden (02 9845 2005), Dr Usha Krishnan (02 9382 1752) or Dr Looi Ee (07 36367887).

This project has been approved by the Ethics Committee of Sydney South West Area Health Service – Concord Repatriation General Hospital. Should you wish to discuss the study with someone not directly involved, in particular, any matters concerning policies, information about the conduct of the study or your rights as a participant, or you wish to make a confidential complaint, at any time, you may contact the Secretary of the Ethics Committee on 02 9767 5622. Alternatively, if you would like to speak to someone at the Royal Children's Hospital Brisbane, you may contact the Co-ordinator of the Ethics Committee on 07 3636 9167. If this phone is unattended, please leave a message and your call will be answered as soon as possible.

This Information Sheet is for you to keep. We will also give you a copy of the signed consent form.

Queensland Government
Queensland Health

QLD Children's Health Services (RCH)
Parent/Guardian and Patient Consent Form

Family Risk and Resilience in Paediatric Liver Disease

Parent/Guardian

I have read the above information. I have asked all of my questions and I have gotten answers. I agree to enrol my child in this study.

_____ _____
Signature of Parent/Guardian Date

CHIEF INVESTIGATOR

I have fully explained to the parent/guardian ..... the nature and purpose of the program and the procedures to be employed as described above and such risks as are involved in their performance, and I have provided the parent/guardian with a copy of the Patient Information Sheet.

_____ _____
Signature of Investigator Date

_____ _____
Print Name Position

INDEPENDENT WITNESS

I have witnessed the receipt of a Patient Information Sheet by the parent/guardian and exchanging of information between the investigator and the parent/guardian about the study.

An auditor witness would optimally discuss the study with the subject and witness the subject signature

_____ _____
Signature of Witness Date

_____ _____
Print Name Position

PARTICIPANT

I have read the above information. I have asked all of my questions and I have gotten answers. I agree to take part in this study.

_____ _____
Signature of Child/Adolescent Date

The Royal Children's Hospital Melbourne

Research & Ethics

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13 July 2011

Prof Winita Hardikar
Department of Gastroenterology and Nutrition
RCH

Dear Prof Hardikar,

Re: HREC 31051 A
Family Functioning, Parent Stress and Child Outcomes in Serious Paediatric Liver Disease

Please find attached the RCH HREC Approval Certificate for the above project.

Please note that this application was recommended for Chairman's approval (expedited review). All Chair approvals are ratified at the subsequent Human Research Ethics Committee (HREC) meeting. In the interim the HREC require the approved materials to be used, as listed on the attached Approval Certificate.

Also, please note the conditions of ethics approval which have been listed on the certificate.

The Committee wishes you well with your research study.

Yours sincerely,

Ethics and Research Department, on behalf of the
RCH Human Research Ethics Committee

The Royal Children's Hospital Human Research Ethics Committee (RCH HREC) is constituted according to the National Health and Medical Research Council's 'National Statement on Ethical Conduct in Humans Research (2007). The committee operates in accordance with these guidelines and is registered with the NHMRC.

The **Royal Children's Hospital** Melbourne

RCH HUMAN RESEARCH ETHICS COMMITTEE APPROVAL

HREC REF. No:	**31051 A**
PROJECT TITLE:	Family Functioning, Parent Stress and Child Outcomes in Serious Paediatric Liver Disease
DOCUMENTS APPROVED:	PGIL v8 dated 24 April 2011 Qualitative Interview Questions v1 dated 19 August 2008 Information Sheet v1 dated 19 August 2008 DAD-F Validated DAD-M Validated Family Assessment Device Validated Impact on Family Scale - Validated DASS - Validated Cover Letter 1 v2 dated 7 May 2011 Cover Letter 2 v2 dated 7 May 2011 Cover Letter 3 v2 dated 7 May 2011 Cover Letter 4 v2 dated 7 May 2011 Cover Letter 5 v2 dated 7 May 2011 Cover Letter 6 v2 dated 7 May 2011
APPROVED PROTOCOL:	Study Protocol v2 dated 24 April 2011
PRINCIPAL INVESTIGATOR:	Winita Hardikar
DATE OF ORIGINAL APPROVAL:	**13 July 2011**
DURATION:	36 months
DATE OF APPROVAL EXPIRY:	**13 July 2014**
SIGNED:	..... 13/7/11

COMMITTEE REPRESENTATIVE

APPROVED SUBJECT TO THE FOLLOWING CONDITIONS:

ALL PROJECTS

1. Must comply with the Investigator's Responsibilities in Research available at http://www.rch.org.au/emplibrary/ethics/Investigators_Responsibilities_in_Research.pdf
2. Any proposed change in the protocol or approved documents or the addition of documents (including flyers, brochures, advertising material etc) must be submitted to the Human Research Ethics Committee (HREC) for approval prior to implementation.
3. The Principal Investigator must notify Ethics & Research of:
   - Any serious adverse effects of the study on participants and steps taken to deal with them.
   - Any unforeseen events (e.g. protocol violations or complaints).
   - Investigators withdrawing from or joining the project.
4. A progress report must be submitted annually and at the conclusion of the project.
5. RCH HREC approval must remain current for the entire duration of the project. If the project is not completed in the allocated time a renewal request must be submitted to the Ethics & Research Department. Investigators undertaking projects without current HREC approval risk their indemnity, funding and publication rights.

CLINICAL TRIALS

6. Must comply with Good Clinical Practice (GCP) available at http://www.tga.gov.au/docs/pdf/euguide/ich/ich13595.pdf
7. Must report all internal (occurring in RCH participants) Serious Adverse Events (SAE) to the sponsor and the RCH HREC within 72 hours of occurrence.
8. Must report all Suspected Unexpected Serious Adverse Reactions (SUSARS) to the Therapeutic Goods

The Royal Children's Hospital Melbourne

PARENT/GUARDIAN INFORMATION LETTER AND CONSENT FORM

HREC Project Number: **31051**

Research Project Title: **Family Risk and Resilience in Paediatric Liver Disease**

Dear Parent

We would like you to consider participating in a research study that will be conducted in the Department of Psychological Medicine at The Children's Hospital Westmead, NSW (CHW), in conjunction with the Department of Gastroenterology at Sydney Children's Hospital Randwick, NSW (SCH), the Paediatric Gastroenterology, Hepatology & Nutrition Service, Royal Children's Hospital, Brisbane, QLD (RCHQ) and the Department of Gastroenterology and Nutrition, The Royal Children's Hospital, Melbourne VIC (RCHV).

What is the study about?
This study is trying to find out how your child's liver disease affects you as parents and the relationships within the family and how these are related to the physical and psychological health outcomes for your child. The information we get from this study will help us to develop our services to best meet the needs of families who have children with liver disease. We hope that around 30 parents in total will participate in this study. The study forms part of a PhD project by Dr M Bowden.

Who can participate in the study?
We are inviting all families who have an infant diagnosed with liver disease at the Children's Hospital at Westmead, the Sydney Children's Hospital at Randwick, the Royal Children's Hospital at Brisbane and The Royal Children's Hospital at Melbourne to participate in this study.

What will the study involve?
If you decide to participate in the study, this is what we will ask you to do:

1. Complete and sign the consent form and return it to your doctor.
2. Dr Michael Bowden will contact you to arrange a convenient time for a family interview to take place.
3. We will send you four (4) questionnaires for each parent to complete before the interview. These will take approximately 30 minutes to complete. There are no right or wrong answers; the questionnaires ask about your feelings, your experiences of having a child with a physical illness and about your family.
4. If you would like to participate in the study but have trouble completing the questionnaires, please call Dr Michael Bowden for help (02 9845 2005). If you prefer, you can bring the questionnaires to your hospital appointment and we will help you to fill them in.
5. All family members are invited to the interview. Dr Michael Bowden will talk to you about the relationships in the family, about your experiences of having a child with liver disease and any other concerns you may have. This interview takes about 45 minutes to one hour and will be tape recorded so that it can be accurately recorded and analysed.
6. About 12 months after your child's diagnosis, Dr Michael Bowden will contact you again to arrange a follow up interview. You will be sent the same questionnaires as previously, with one additional questionnaire that asks about your child's behaviour and feelings, which takes about 20 minutes to complete. The interview will be the same as previously.
7. At the end of the project, we will send you a summary of the results. This will be of the whole group of participants, not your individual results.
8. You and your family will receive the usual clinical assessment, care and treatment from your treating health care team.
9. This is all that you will be asked to do, though we may ask to follow up again at a later date. If so, we will ask you if you wish to participate then and will give you full details. There will be no requirement for you to participate further if you do not wish to.

Are there any benefits for my child participating in the study?
There are no known benefits for you or your child in participating in this study. However, some participants may benefit from having a comprehensive psychological assessment that they would otherwise not receive. We hope that the results from this study will help us to learn more about the support needs of the families of children we care for.

Are there any side-effects and risk associated with this study?
There are no known side-effects or risks associated with participation in this study. We expect that the time needed to complete the questionnaires and the interviews is the only inconvenience to you in taking part in this study. Talking about difficult health issues in your child may be distressing. If so, we will refer you to a counsellor for further assistance.

Other information
Your confidentiality will be maintained throughout the study period by not using your or your child's name on any study documents. We need to keep track of your information and will identify your data only with your child's medical record number and date of birth. Only group data will be reported so that individuals will not be able to be identified in any publication that may arise from the study, including in the PhD thesis that will result from this study. Research data may be accessed by auditors, ethics committee or regulatory authorities.

The audiotape of your interview will be transcribed and then the tape will be destroyed. The study data, including the transcribed data, will be stored in the Department of Psychological Medicine at the Children's Hospital at Westmead, in a locked cabinet that is only accessible to the study group. It will be stored for a period of seven (7) years after completion of the study to comply with Hospital guidelines. After this it will be destroyed through a secure document destruction company employed by the Children's Hospital at Westmead.

Participation in this project is entirely voluntary. If you decide not to take part or decide to withdraw at any time, please be assured that this will not affect your child's care at the Hospital or your relationship with Hospital staff.

If you have any questions about the conduct of this study, please do not hesitate to discuss them with the principal investigators, Dr Michael Bowden (02 9845 2005), Dr Usha Krishnan (02 9382 1752), Dr Looi Ee (07 36367887) or A/Prof Winita Hardikar (03 9345 7998)

This project has been approved by the Ethics Committee of Sydney South West Area Health Service – Concord Repatriation General Hospital.

Should you wish to discuss the study with someone not directly involved, in particular, any matters concerning policies, information about the conduct of the study or your rights as a participant, or you wish to make a confidential complaint, at any time, you may contact the Secretary of the Ethics Committee on 02 9767 5622. Alternatively, if you would like to speak to someone at The Royal Children's Hospital Melbourne, you may contact the Co-ordinator of the Ethics Committee on (03) 9345 5044. If you have any concerns about the project or the way it is being conducted, and would like to speak to someone independent of the project, please contact: Director, Ethics & Research, The Royal Children's Hospital on telephone: (03) 9345 5044.

This Information Sheet is for you to keep. We will also give you a copy of the signed consent form.

Yours sincerely

Dr Michael Bowden
Department of Psychological Medicine
Children's Hospital Westmead

A/Prof Winita Hardikar
Department of Gastroenterology and Clinical Nutrition
The Royal Children's Hospital Melbourne

The Royal Children's Hospital Melbourne

CONSENT FORM

HREC Project Number: **31051**

Research Project Title: **Family Risk and Resilience in Paediatric Liver Disease**

Version Number: 8 **Version Date:** 24/04/2011

- I voluntarily consent for my child to take part in this research project
- I believe I understand the purpose, extent and possible effects of my child's involvement in this project.
- I have had an opportunity to ask questions and I am satisfied with the answers I have received.
- I understand that this project has been approved by The Royal Children's Hospital Human Research Ethics Committee and will be carried out in line with the National Statement on Ethical Conduct in Human Research (2007).
- I understand I will receive a copy of Parent/Guardian Information Letter and Consent Form.

_____
Child's Name

_____	_____	_____
Parent/Guardian Name	Parent/Guardian Signature	Date

I have supplied an Information Letter and Consent Form to the parent/guardian who has signed above, and believe that they understand the purpose, extent and possible effects of their child's involvement in this project.

_____	_____	_____
Research Team Member Name	Research Team Member Signature	Date

Note: All parties signing the Consent Form must date their own signature.

Appendix B
Publications Arising from the Thesis

ORIGINAL ARTICLE: HEPATOLOGY

Family Adjustment and Parenting Stress When an Infant Has Serious Liver Disease: The Australian Experience

*Michael R. Bowden, †Michael Stormon, ‡Winita Hardikar, §Looi C. Ee, ||Usha Krishnan, ¶Diana Carmody, †Vicki Jermyn, *Mee-Mee Lee, †Edward V. O'Loughlin, †Janine Sawyer, ‡Kathe Beyerle, ||Daniel A. Lemberg, #Andrew S. Day, **Campbell Paul, and ††Philip Hazell

ABSTRACT

Objectives: Parenting stress, problems in family functioning, and lack of fathers' engagement in treatment are associated with poor quality of life in children with chronic illnesses. The aim of the present study was to examine these characteristics in families of infants with serious liver disease in Australia, to inform the provision of mental health care for these families.

Methods: From September 2009 to May 2013, 42 parents of infants recently diagnosed as having serious liver disease (defined as liver disease that may require transplantation in the future) completed questionnaires about family function, impact of the infant's illness on the family, parent stress symptoms, and fathers' engagement in the care of the child. Participants were recruited from 4 metropolitan children's hospitals in Australia.

Results: Parents reported psychological symptoms at similar rates to normative populations. Their reports of family functioning were significantly below mean scores in previously published populations with a medically ill family member (population mean 1.89; mothers mean 1.59; fathers mean 1.61, $P < 0.001$). Disruption to family roles was significantly correlated with psychological symptoms for mothers ($r = 0.48$, $P < 0.01$) and fathers ($r = 0.31$, $P < 0.05$). Greater helpfulness of fathers was correlated with lower depression in mothers ($r = -0.35$, $P < 0.05$), and fathers' anxiety was correlated with their increased engagement ($r = 0.40$, $P < 0.01$).

Conclusions: When parents report the presence of psychological symptoms, symptoms are likely to be present in both parents and are associated with difficulties adjusting to disrupted family roles. Father engagement may be protective of mothers' mental health.

Key Words: family relationships, fathers, infants, liver disease, psychological stress

(*JPGN* 2015;60: 717 722)

What Is Known

- Pediatric liver disease requiring transplantation, although uncommon, is associated with significant stress for the children and their parents.
- Previous studies of children who have had a liver transplant report that family functioning is unaffected.

What Is New

- Contrary to previous research, we found limited impact on parent stress but a significant detrimental impact on family functioning.
- When families are distressed, both parents report psychological symptoms and altered family functioning.
- Anxious fathers are more engaged in infant care.
- Greater father engagement is protective of maternal mental health.

Received November 20, 2014; accepted January 23, 2015.

From the *Departments of Psychological Medicine, the †Gastroenterology, the ‡Department of Gastroenterology and Clinical Nutrition, the §Queensland Paediatric Gastroenterology, Hepatology, and Nutrition Service, Royal Children's Hospital, Melbourne, the ¶Social Work, Children's Hospital, Westmead, Sydney, the ||Department of Gastroenterology, Sydney Children's Hospital, Randwick, Australia, the #Department of Paediatrics, University of Otago, Christchurch, New Zealand, the **Department of Psychiatry, University of Melbourne, Melbourne, and the ††Discipline of Psychiatry, Sydney Medical School, Sydney, Australia.

Address correspondence and reprint requests to Michael R. Bowden, BMed, PhD, Department of Psychological Medicine, Children's Hospital Westmead, Locked Bag 4001, Westmead, NSW 2145, Australia (e-mail: michael.bowden@health.nsw.gov.au).

The present study forms part of the research for a PhD degree (M.R.B.).

The authors report no conflicts of interest.

DOI: 10.1097/MPG.0000000000000742

Pediatric liver disease is rare, but is associated with serious morbidity such as repeated hospitalization, multiple interventional procedures, and the potential need for transplantation (1,2). Parents of children with chronic illnesses experience high rates of stress (3 5). Stress in parents of children who have had a liver transplant is associated with poor adherence to medical treatment (6,7) and consequently poor health outcomes (6), suggesting the potential clinical importance of identifying and managing parental stress in this situation.

Studies of family functioning after pediatric liver transplantation (6,8) show generally favorable outcomes posttransplant, although clinical experience suggests disruption to family routines and relationships, which can also be associated with poorer quality of life in these children (9).

Fathers' involvement in treatment has received little attention in the literature to date. Greater father engagement in the treatment of children and adolescents with chronic illness has been linked with improved maternal mental health and family functioning (10), as well as improved treatment adherence and quality of life

in the children (11). A family systems approach to studying children with chronic illness may therefore assist with identifying potential targets for intervention.

Although a noncategorical approach to studying children with chronic illness has been advocated (12), a meta-analysis showed significant differences between the behavioral outcomes for children between types of illnesses (13), suggesting that studying individual illnesses is important for service planning. Because liver disease in children is rare, many previous studies include small numbers of children across a wide range of ages and developmental stage. This may bias study outcomes because of the inherent differences in children across developmental stages, as well as the differences between families with children of different ages. In addition, previous pediatric liver transplant studies have shown variable findings because of sampling heterogeneity (13). Hence, studying children of similar developmental stage at a similar point in their illness may be important in identifying specific illness effects.

We aimed to identify characteristics of families of infants who present with serious liver disease to inform clinical practice in the provision of mental health care for these families, particularly given that the birth of an infant is a period of adjustment for all of the new parents. We hypothesized that having an infant with serious liver disease will result in alterations in family functioning and psychological symptoms for some parents. We further hypothesized that fathers' engagement in the care of the infant will be associated with family and parent symptoms.

METHODS

Participants

In Australia, there are 3 pediatric liver transplant units located in Sydney, Brisbane, and Melbourne. The present study includes consecutive patients from all of the 3 centers and from an additional children's hospital in Sydney that does not offer transplants. The parents of infants (ages <2 years) newly diagnosed as having serious liver disease were eligible to participate. Serious liver disease was defined as liver disease that may require transplantation in the future. Exclusion criteria were inadequate parental English to allow self-completion of the questionnaires and families with separated parents. Two-parent families were sought to examine father engagement. Parents were approached to participate approximately 3 months after the initial diagnosis, to allow early normative stress reactions to the diagnosis to resolve (14). Participants were enrolled between September 2009 and May 2013.

Ethics approval was obtained from each hospital's human research ethics committee. Written informed consent was obtained from all of the participants.

Study Instruments

Severity of Illness

There is no available standardized measure of severity in this patient population. Pediatric end-stage liver disease scores are valid for children who are in the end stage of liver disease (15), but not for children with less severe disease. Therefore, we calculated the ratio of days in hospital since onset of illness (the number of days in hospital divided by the number of days between onset of illness and the time of data collection) as a proxy for illness severity.

Family Functioning

Each parent separately completed the Family Assessment Device (FAD) (16), a validated 60-item self-report questionnaire assessing family functioning. It has normative data for healthy, clinical (psychiatric), and medically ill patients (17). It provides a measure of family functioning across 7 subscales. The Problem Solving subscale reflects a family's capacity to solve practical and emotional problems. The Communication subscale measures directness and clarity of communication in the family. The Roles subscale evaluates role allocation and accountability. Affective Responsiveness reflects family members' capacity for and appropriateness of emotional response. Affective Involvement refers to the interest that family members show for each other. Behavior Control is a measure of how well the family manages members' behavior. Finally, General Functioning covers aspects of each of the other 6 subscales and measures overall family functioning. Higher scores are associated with greater dysfunction in each subscale.

Impact of the Infant's Illness on the Family

The Impact on Family Scale (IFS) (18) is a 27-item self-report questionnaire validated for use with families who have a child with a chronic illness. It provides a total score, with higher scores representing greater impact on the family of the child's illness. Each parent completed the IFS individually.

Parent Psychological Distress

The Depression Anxiety Stress Scale (DASS) (19) is a 42-item validated self-report scale that asks about symptoms of depression, anxiety, and stress experienced during the preceding 1 week. It provides scores for depression, anxiety, stress, and a total symptom score. Higher scores indicate higher levels of psychological symptoms. Again, each parent completed the DASS independently.

Fathers' Engagement

The Dad's Active Disease Support Scale (20) is a 24-item validated self-report scale that asks about how much the father is engaged in tasks related to the sick child's illness and the helpfulness of his engagement. There is a separate form for mothers and fathers with scores for both the amount and the helpfulness of fathers' involvement. Higher scores reflect more involvement and greater perceived helpfulness.

Statistical Analysis

Comparisons with published population norms were made using 1-sample t tests, whereas comparisons of continuous variables were made using 2-sample t tests and analysis of variance. Univariate regression was used to analyze whether continuous demographic variables significantly predicted the study measures. Demographic variables were analyzed against the General Functioning subscale of the FAD, the total score of the DASS, the IFS total score, and the Dad's Active Disease Support Scale. Pearson correlations were used to examine associations between study measures. $P < 0.05$ was considered significant.

RESULTS

Descriptive Statistics

Participant Characteristics

A total of 42 eligible families agreed to participate in the study (participation rate 65%; Fig. 1). Two eligible families could not be contacted. Of the 21 contactable families who declined to participate, 8 declined citing time constraint, 1 declined because their child was ill, 1 declined because the mother was experiencing

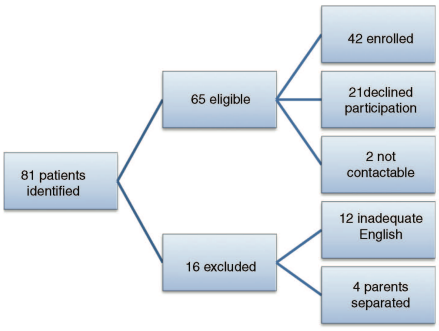

FIGURE 1. Patients screened for study enrolment.

postpartum depression, and the remainder either did not give a reason or stated that they were not interested in the study. There were 15 boy infants (36%), the most common diagnosis was biliary atresia (60%), and one-fourth of the infants had received a liver transplant by the time of study participation (Table 1). There was a wide range of time between the infants' diagnosis and study participation because some families were difficult to contact to arrange completion of study measures (Table 1).

Infant sex, diagnosis, illness severity, age at diagnosis, and treatment hospital were not significant predictors of any study measure, with the exception that greater illness severity predicted fathers' higher ratings of the impact of the illness on the family, explaining 11% of the variation ($R^2 = 0.11$, $F(1, 40) = 5.14$, $\beta = 0.34$, $P = 0.03$).

Socioeconomic status (SES) significantly predicted fathers' reports of problems in family functioning as measured by the FAD General Functioning subscale and mothers' reports of the helpfulness of fathers' engagement in the care of the child, but was not predictive of any other study measures. There was a negative association with family functioning (lower SES predicted more problems in family functioning), accounting for 18% of the variation ($R^2 = 0.18$, $F(1, 40) = 8.46$, $\beta = -0.42$, $P = 0.006$), and a positive association with fathers' engagement (higher SES predicted greater helpfulness of fathers' engagement), predicting 13% of the variation ($R^2 = 0.13$, $F(1, 40) = 5.89$, $\beta = 0.36$, $P = 0.02$).

TABLE 1. Participant characteristics (N = 42)

	N (%)
Child sex male	15 (36)
Diagnosis*	
Biliary atresia	25 (60)
α_1-Antitrypsin deficiency	6 (14)
Alagille syndrome	3 (7)
Autoimmune hepatitis	3 (7)
Cryptogenic hepatitis	3 (7)
Citrullinemia	1 (2)
Hepatoblastoma	1 (2)
Liver transplant at study participation	11 (26)
	Median (range)
Child age at diagnosis, days	61 (2–700)
Mother's age, y	34 (19–46)
Father's age, y	36 (20–53)
Length of time from diagnosis to study participation, days	206 (80–485)
Ratio of days in hospital from diagnosis to study participation	0.18 (0.00–0.98)

* Totals do not add to 100% because of rounding.

Family Functioning

Mean scores on the FAD General Functioning subscale were compared with published norms (mean 1.89) for families with a medically ill family member (16). Both mothers and fathers scored significantly below the norms (Table 2). A total of 79% of mothers and 74% of fathers, however, scored family functioning above the norms on at least 1 subscale of the FAD. There were no significant differences between mothers' and fathers' scores on the FAD.

Impact of the Infant's Illness on the Family

Scores on the IFS were compared with published norms means (mean 48.03) (21). Mothers' mean scores on the IFS did not significantly differ from published norms, whereas fathers' mean scores were significantly lower than published norms (Table 2). Despite this, 52% of mothers and 43% of fathers scored above the published norms. Fathers' scores were significantly lower than mothers' scores ($t(41) = 2.02$, $P = 0.05$).

Parent Stress

Parent total DASS scores were also compared with normative populations (mean 19.9 for women, 16.6 for men) (22). Neither mothers' nor fathers' scores were significantly different from norms. Compared with rates reported during the postpartum period for women (depression mean 5.10; anxiety mean 3.33; stress mean 10.00) (23), mothers' scores were not significantly different (depression mean 6.52, standard deviation [SD], $t(41) = 1.13$, $P = 0.27$; anxiety mean 5.05, SD 7.03, $t(41) = 1.58$, $P = 0.12$; stress mean 11.57, SD 8.25, $t(41) = 1.24$, $P = 0.22$).

Examination of the DASS subscales showed that 21% of mothers and 12% of fathers scored in the mild symptom severity range or higher on at least 1 subscale.

Fathers' total DASS mean scores were lower than mothers', with the difference approaching significance ($t(41) = 1.94$, $P = 0.06$).

Fathers' Engagement

Mothers and fathers agreed on their reports on the extent and helpfulness of fathers' engagement in the care of the infant. Mothers (mean 73.76, SD 19.43) and fathers (mean 75.28, SD 16.85) did not differ significantly on the amount of fathers' engagement, $t(41) = -0.527$, $P = 0.60$. There were also no significant differences in their reports of the helpfulness of fathers' engagement (mothers' mean 76.12, SD 15.63; fathers' mean 72.17, SD 16.13), $t(41) = 1.22$, $P = 0.23$.

Correlations Between Study Measures

Family Functioning, Impact of the Illness on the Family, and Parent Stress Symptoms

There were several significant correlations between mothers' ratings of family functioning and their psychological symptoms (Table 3). Mothers' reports of difficulties in problem solving

TABLE 2. Family characteristics compared with published population means

	Mean (SD)	Mean difference	95% confidence interval of the difference
Family function			
FAD General Functioning[a]			
Mothers	1.59*** (0.37)	−0.30	−0.41 to −0.18
Fathers	1.61*** (0.41)	−0.28	−0.41 to −0.15
Impact of illness			
IFS total score[b]			
Mothers	47.69 (12.83)	−0.34	−4.34 to 3.66
Fathers	44.07* (11.13)	−3.96	−7.42 to −0.49
Parent psychological symptoms			
DASS total score			
Mothers[c]	23.14 (21.86)	3.24	−3.57 to 10.05
Fathers[d]	16.79 (21.7)	0.19	−6.58 to 6.95

DASS = Depression Anxiety Stress Scale; FAD = Family Assessment Device; IFS = Impact on Family Scale; SD = standard deviation.
[a] Population mean (medically ill samples) 1.89.
[b] Population mean 48.03.
[c] Population mean (women) 19.9.
[d] Population mean (men) 16.6.
* $P < 0.05$.
*** $P < 0.001$.

correlated with their stress scores, whereas their ratings of problems in family roles and general functioning were correlated with all of the psychological symptom scores (depression, anxiety, stress, and total scores on the DASS). Mothers' scores on the IFS were correlated with maternal total DASS scores, anxiety, and depression.

There were fewer significant correlations between fathers' scores of family functioning and psychological symptoms (Table 3). Fathers' scores of communication were correlated with their stress scores, and their reports of problems in family roles were correlated with stress and total symptoms.

Both mothers' and fathers' reports of higher impact of the child's illness on the family correlated with problems in family roles (mothers: $r = 0.46$, $P = 0.002$; fathers: $r = 0.48$, $P = 0.001$).

Fathers' Engagement

Fathers who were more anxious rated themselves as more engaged and rated their engagement as more helpful. Fathers' total DASS scores were also correlated with their ratings of the helpfulness of their engagement. Mothers' ratings of the helpfulness of fathers' engagement negatively correlated with mothers' depression (Table 3). There was no significant correlation between the impact of the infant's illness on the family and fathers' engagement.

DISCUSSION

To our knowledge, the present study is the first to examine the interaction among family functioning, parental stress, and fathers' engagement in families of infants with serious liver disease.

TABLE 3. Pearson correlations between parent psychological symptoms (measured on the Depression, Anxiety, and Stress Scale) and ratings of family functioning, impact of the child's illness on the family, and fathers' engagement (N = 42)

	Depression	Anxiety	Stress	Total symptoms
Mothers				
FAD subscale				
Problem Solving	0.28	0.14	0.31*	0.26
Communication	0.16	0.11	0.12	0.14
Roles	0.42**	0.52**	0.40**	0.48**
Affective Responsiveness	−0.17	−0.08	−0.13	−0.17
Affective Involvement	0.20	0.38*	0.26	0.30
Behavior Control	−0.06	0.09	0.00	0.01
General Functioning	0.33*	0.37*	0.37*	0.38*
Impact on Family Scale				
Total score	0.34*	0.49**	0.28	0.39*
DADS amount	−0.11	−0.05	−0.04	−0.07
DADS helpfulness	−0.35*	−0.27	−0.19	−0.29
Fathers				
FAD subscale				
Problem Solving	0.17	0.09	0.21	0.17
Communication	0.29	0.16	0.31*	0.28
Roles	0.29	0.26	0.32*	0.31*
Affective Responsiveness	0.14	0.10	0.16	0.15
Affective Involvement	0.22	0.18	0.24	0.23
Behavior Control	0.15	0.11	0.12	0.13
General Functioning	0.12	0.04	0.19	0.13
Impact on Family Scale				
Total score	0.20	0.11	0.30	0.22
DADS amount	0.17	0.40**	0.17	0.25
DADS helpfulness	0.26	0.41**	0.27	0.33*

DADS = Dad's Active Disease Support Scale; FAD = Family Assessment Device.
* $P < 0.05$.
** $P < 0.01$.

It is the largest psychosocial study of infants with liver disease in Australia to date. We limited age and duration of illness effects by recruiting only infants, instead of children at varying stages of development. We examined the family as a system, taking into account parent psychological symptoms and considering the extent of fathers' engagement in the care of the child. We found significant correlations between parent psychological symptoms and family functioning, particularly in relation to role disruption, which was also associated with higher perceived impact of the infant's illness on the family. This finding is in line with another Australian study, which found that disrupted family routine was associated with poorer quality of life in children who have had a liver transplant (9). In addition, the mothers in our study who reported greater helpfulness of fathers in the care of the infant also reported lower levels of depression.

Previous research in this area has been undertaken with infants and children who have had a liver transplant, instead of with those who have serious liver disease. Consistent with previous research in pediatric liver transplantation, our group of families of infants with serious liver disease reported general family functioning to be comparable with population norms (6,8). Compared with previous research, however, more parents in the present study reported problems in at least 1 domain of family functioning. The difference may be because of our study being restricted to parents of infants soon after the diagnosis of liver disease instead of in a post liver transplant group. A US study of children with chronic illness (24), which included a large infant cohort, found normal family functioning overall but abnormalities across domains of family functioning associated with severity of illness in the infant group. Severity of illness in our study was associated with fathers' reports of the impact of the illness and not with family functioning.

Parents' scores on the DASS were comparable with population means (22,23), suggesting that they are no more stressed than members of the general population without a sick infant. This is surprising given the high level of stress these families are under and suggests that present measures are inadequate or parents may underreport psychological symptoms. This needs further study to elucidate. Nevertheless, we found significant correlations between parent symptoms and family functioning. This was particularly apparent for role disruption for mothers, which was associated with all of the 3 psychological symptom areas of depression, anxiety, and stress. For fathers, family role disruption and communication were the only areas of family functioning associated with stress symptoms. For our participants, role adjustment appears to be the biggest issue. This may offer an opportunity for early detection and intervention, to minimize psychological symptoms in both parents.

Fathers' engagement in the medical care of chronically ill children has not been widely studied. In our study, fathers' engagement was protective: we found less depression in mothers who rated fathers as more helpful. These findings are in line with another study of chronically ill children (10). That study, however, also found that fathers' engagement was associated with less impact of the disease on the family and improved family functioning, which differs from our findings.

We found that fathers who rated themselves as more engaged in the care of the infant also reported greater anxiety. It is not clear from our cross-sectional study whether the fathers' anxiety led them to become more engaged in the medical care of the infant; it is also possible that fathers who had preexisting anxiety were more engaged even before the infant's diagnosis. Although more anxious fathers are more engaged, their engagement does not reduce the impact of the child's illness on the family. Mothers' and fathers' depression and anxiety scores were correlated, suggesting an interactional effect whereby the infant's illness creates anxiety and/or depression in both parents together. Overall, the findings suggest that when an infant's diagnosis of serious liver disease has disruptive effects on parents and families, multiple areas of functioning tend to be affected.

A limitation of the study was the small sample size. Some families were difficult to contact, leading to a large range of time from diagnosis to participation. It is possible that this resulted in bias, for example, some parents may have delayed participation until they had better adjusted to the infant's diagnosis. This also raised the issue that one-quarter of the infants had already had a transplant by the time of study participation. Study numbers were low to test for significant differences between hospitals for infants who had or had not received a transplant or to compare the data from the nontransplant center with that from the other hospitals. This is a potential bias because there may be differences between those families whose infant has or has not had a transplant and warrants further investigation. In addition, the cross-sectional nature of our data limits the inferences that can be drawn regarding causation. Another potential source of bias is the number of eligible families who did not participate, particularly the 10 families who declined participation because of their child being ill (1 family), mental illness in a parent (1 family), and the 8 families who cited lack of time to participate. These families may have been more stressed than those who agreed to participate. Finally, the data are limited to English-speaking families of children with serious liver disease in Australia and may not generalize to other populations. Our study, however, extends observations from international studies in chronic illness to the specific situation of liver disease in infants and to the Australian context in particular.

Our study has a number of practical implications. First, parents may underreport psychological stress symptoms and family difficulties, resulting in parents not being referred for psychological assessment. Enquiring about disruption to family routines may clarify which parents warrant additional psychological assessment. It is therefore important to ask about stress symptoms and about disruption to family roles and routines, with early referral to psychological services. Alternatively, routine psychosocial evaluation several months after the infant's initial diagnosis could identify those families who could benefit from early intervention. Second, psychological symptoms in 1 parent should suggest the presence of similar symptoms in the other parent. Finally, engaging fathers in the care of the infant may be protective for mothers, although it does not appear to improve family function. Mental health assessment and management is an important part of the health care for these families.

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Appendix C
Family Assessment Device

Items Used for the General Functioning Sub-scale Score

1. Planning family activities is difficult because we misunderstand each other
2. In times of crisis, we can turn to each other for support
3. We cannot talk to each other about the sadness we feel
4. Individuals are accepted for what they are
5. We avoid discussing our fears and concerns
6. We can express feelings to each other
7. There are lots of bad feelings in the family
8. We feel accepted for what we are
9. Making decisions is a problem for our family
10. We are able to make decisions about how to solve problems
11. We don't get along well together
12. We confide in each other

Appendix D
Impact on Family Scale

Items Used for the 15-Item Total Score

1. Because of the illness, we are not able to travel out of the city.
2. People in the neighbourhood treat us specially because of my child's illness.
3. We have little desire to go out because of my child's illness.
4. It is hard to find a reliable person to take care of my child.
5. Sometimes we have to change plans about going out at the last minute because of my child's state.
6. We see family and friends less because of the illness.
7. Sometimes I wonder whether my child should be treated "specially" or the same as a normal child
8. I think about not having more children because of the illness.
9. I don't have much time left over for other family members after caring for my child.
10. Our family gives up things because of my child's illness.
11. Fatigue is a problem for me because of my child's illness.
12. I live from day to day and don't plan for the future.
13. Nobody understands the burden I carry.
14. Travelling to the hospital is a strain on me.
15. Sometimes I feel like we live on a rollercoaster; in crisis when my child is acutely ill, OK when things are stable.

Items Used for the 19-Item Total Score

1. The illness is causing financial problems for the family.
2. Time is lost from work because of hospital appointments.
3. I am cutting down the hours I work to care for my child.
4. Additional income is needed in order to cover medical expenses.
5. Because of the illness, we are not able to travel out of the city.
6. People in the neighbourhood treat us specially because of my child's illness.
7. We have little desire to go out because of my child's illness.
8. It is hard to find a reliable person to take care of my child.
9. Sometimes we have to change plans about going out at the last minute because of my child's state.
10. We see family and friends less because of the illness.
11. Sometimes I wonder whether my child should be treated "specially" or the same as a normal child
12. I think about not having more children because of the illness.
13. I don't have much time left over for other family members after caring for my child.
14. Our family gives up things because of my child's illness.
15. Fatigue is a problem for me because of my child's illness.
16. I live from day to day and don't plan for the future.
17. Nobody understands the burden I carry.
18. Travelling to the hospital is a strain on me.
19. Sometimes I feel like we live on a rollercoaster; in crisis when my child is acutely ill, OK when things are stable.

Appendix E
Participant Characteristics SPSS Output Tables

Family Demographics

			Statistic	Std. Error
Socio Economic Index	Mean		60.893	3.7378
	95% Confidence Interval for Mean	Lower Bound	53.344	
		Upper Bound	68.442	
	5% Trimmed Mean		60.788	
	Median		65.100	
	Variance		586.802	
	Std. Deviation		24.2240	
	Minimum		23.3	
	Maximum		100.0	
	Range		76.7	
	Interquartile Range		46.5	
	Skewness		-.117	.365
	Kurtosis		-1.340	.717
Infant Age at Diagnosis (Days)	Mean		117.90	27.805
	95% Confidence Interval for Mean	Lower Bound	61.75	
		Upper Bound	174.06	
	5% Trimmed Mean		92.53	
	Median		60.50	
	Variance		32470.430	
	Std. Deviation		180.196	
	Minimum		2	
	Maximum		700	
	Range		698	
	Interquartile Range		47	
	Skewness		2.734	.365
	Kurtosis		6.102	.717
Mother's Age	Mean		32.95	1.060
	95% Confidence Interval for Mean	Lower Bound	30.81	
		Upper Bound	35.09	
	5% Trimmed Mean		33.06	
	Median		34.00	
	Variance		47.217	
	Std. Deviation		6.871	
	Minimum		19	
	Maximum		46	
	Range		27	
	Interquartile Range		10	
	Skewness		-.224	.365
	Kurtosis		-.630	.717
Father's Age	Mean		35.12	1.108
	95% Confidence Interval for Mean	Lower Bound	32.88	
		Upper Bound	37.36	
	5% Trimmed Mean		35.04	
	Median		36.00	
	Variance		51.522	
	Std. Deviation		7.178	
	Minimum		20	
	Maximum		53	
	Range		33	
	Interquartile Range		9	
	Skewness		-.064	.365
	Kurtosis		.214	.717

Infant Gender

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Male	15	35.7	35.7	35.7
	Female	27	64.3	64.3	100.0
	Total	42	100.0	100.0	

Birth Order

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	1	15	35.7	35.7	35.7
	2	14	33.3	33.3	69.0
	3	6	14.3	14.3	83.3
	4	5	11.9	11.9	95.2
	5	1	2.4	2.4	97.6
	7	1	2.4	2.4	100.0
	Total	42	100.0	100.0	

Birth Order (First born or not first born)

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	First born	15	35.7	35.7	35.7
	Not first born	27	64.3	64.3	100.0
	Total	42	100.0	100.0	

Infant Diagnosis

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Biliary Atresia	25	59.5	59.5	59.5
	Alpha 1 AT	6	14.3	14.3	73.8
	Alagille Syndrome	3	7.1	7.1	81.0
	Autoimmune Hepatitis	3	7.1	7.1	88.1
	Cryptogenic Hepatitis	3	7.1	7.1	95.2
	Citrullinemia	1	2.4	2.4	97.6
	Hepatoblastoma	1	2.4	2.4	100.0
	Total	42	100.0	100.0	

Diagnosis (Biliary Atresia or Other severe liver disease)

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Biliary Atresia	25	59.5	59.5	59.5
	Other severe liver disease	17	40.5	40.5	100.0
	Total	42	100.0	100.0	

Recruitment Hospital

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	CHW[a]	18	42.9	42.9	42.9
	SCH[b]	5	11.9	11.9	54.8
	RCHB[c]	8	19.0	19.0	73.8
	RCHM[d]	11	26.2	26.2	100.0
	Total	42	100.0	100.0	

[a]CHW Children's Hospital Westmead
[b]SCH Sydney Children's Hospital
[c]RCHB Royal Children's Hospital Brisbane
[d]RCHM Royal Children's Hospital Melbourne

Timing of Study Participation

			Statistic	Std. Error
Time from diagnosis to Time 1 (Days)	Mean		230.62	14.830
	95% Confidence Interval for Mean	Lower Bound	200.67	
		Upper Bound	260.57	
	5% Trimmed Mean		226.35	
	Median		206.00	
	Variance		9237.559	
	Std. Deviation		96.112	
	Minimum		80	
	Maximum		485	
	Range		405	
	Interquartile Range		143	
	Skewness		.740	.365
	Kurtosis		-.132	.717
Time between Time 1 and Time 2 (Days)	Mean		428.27	11.377
	95% Confidence Interval for Mean	Lower Bound	405.20	
		Upper Bound	451.34	
	5% Trimmed Mean		422.74	
	Median		405.00	
	Variance		4788.814	
	Std. Deviation		69.201	
	Minimum		343	
	Maximum		608	
	Range		265	
	Interquartile Range		89	
	Skewness		1.271	.388
	Kurtosis		.742	.759

Participation at Time 2

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	No/withdrawn	5	11.9	11.9	11.9
	Yes	37	88.1	88.1	100.0
	Total	42	100.0	100.0	

Illness Severity Measures

			Statistic	Std. Error
Ratio of outpatient visits since diagnosis at Time 1	Mean		.0434	.00479
	95% Confidence Interval for Mean	Lower Bound	.0337	
		Upper Bound	.0531	
	5% Trimmed Mean		.0403	
	Median		.0377	
	Variance		.001	
	Std. Deviation		.03102	
	Minimum		.00	
	Maximum		.18	
	Range		.18	
	Interquartile Range		.04	
	Skewness		2.049	.365
	Kurtosis		6.908	.717
Ratio of outpatient visits since diagnosis at Time 2	Mean		.0380	.00382
	95% Confidence Interval for Mean	Lower Bound	.0302	
		Upper Bound	.0457	
	5% Trimmed Mean		.0365	
	Median		.0321	
	Variance		.001	
	Std. Deviation		.02321	
	Minimum		.01	
	Maximum		.10	
	Range		.09	
	Interquartile Range		.03	
	Skewness		.796	.388
	Kurtosis		.046	.759
Ratio of days admitted since diagnosis at Time 1	Mean		.2430	.03368
	95% Confidence Interval for Mean	Lower Bound	.1750	
		Upper Bound	.3110	
	5% Trimmed Mean		.2269	
	Median		.1747	
	Variance		.048	
	Std. Deviation		.21825	
	Minimum		.00	
	Maximum		.98	
	Range		.98	
	Interquartile Range		.31	
	Skewness		1.237	.365
	Kurtosis		1.619	.717
Ratio of days admitted since diagnosis at Time 2	Mean		.1326	.02074
	95% Confidence Interval for Mean	Lower Bound	.0906	
		Upper Bound	.1747	
	5% Trimmed Mean		.1217	
	Median		.1158	
	Variance		.016	
	Std. Deviation		.12614	
	Minimum		.00	
	Maximum		.49	
	Range		.49	
	Interquartile Range		.16	
	Skewness		1.183	.388
	Kurtosis		1.018	.759

Liver transplant at Time 1 or not

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	No liver transplant at Time 1	31	73.8	73.8	73.8
	Liver transplant at Time 1	11	26.2	26.2	100.0
	Total	42	100.0	100.0	

Liver transplant at Time 2 or not

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	No liver transplant at Time 2	18	42.9	48.6	48.6
	Liver transplant at Time 2	19	45.2	51.4	100.0
	Total	37	88.1	100.0	
Missing	Withdrawn	5	11.9		
Total		42	100.0		

Infant CBCL T-score

			Statistic	Std. Error
Mother CBCL Total Problems T-Score	Mean		46.78	1.901
	95% Confidence Interval for Mean	Lower Bound	42.93	
		Upper Bound	50.64	
	5% Trimmed Mean		46.20	
	Median		45.00	
	Variance		133.730	
	Std. Deviation		11.564	
	Minimum		28	
	Maximum		78	
	Range		50	
	Interquartile Range		14	
	Skewness		.763	.388
	Kurtosis		.762	.759
Father CBCL Total Problems T-Score	Mean		46.41	1.940
	95% Confidence Interval for Mean	Lower Bound	42.47	
		Upper Bound	50.34	
	5% Trimmed Mean		46.18	
	Median		46.00	
	Variance		139.248	
	Std. Deviation		11.800	
	Minimum		28	
	Maximum		72	
	Range		44	
	Interquartile Range		19	
	Skewness		.111	.388
	Kurtosis		-.928	.759

Appendix F
Family Demographics and Illness Variables Normality Assessment

Family Demographics Score Distributions

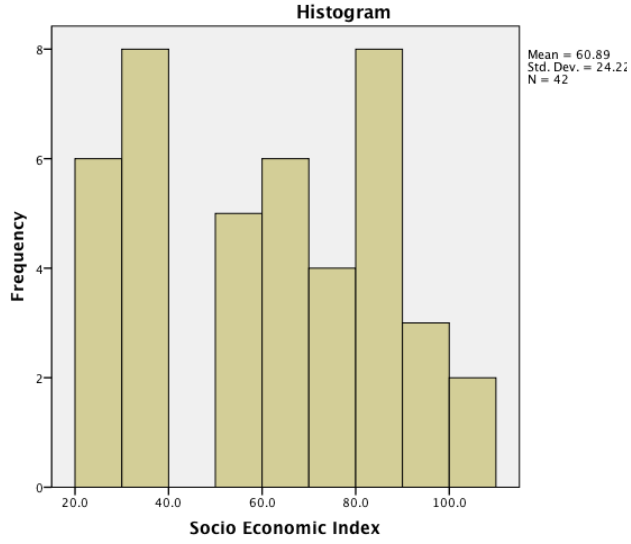
Tests of Normality

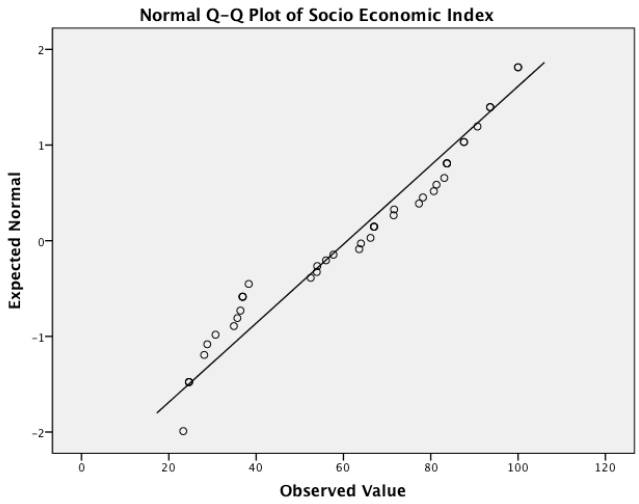
	Kolmogorov-Smirnov[a]		
	Statistic	df	Sig.
Socio Economic Index	.158	42	.010
Mother's Age	.108	42	.200*
Father's Age	.097	42	.200*

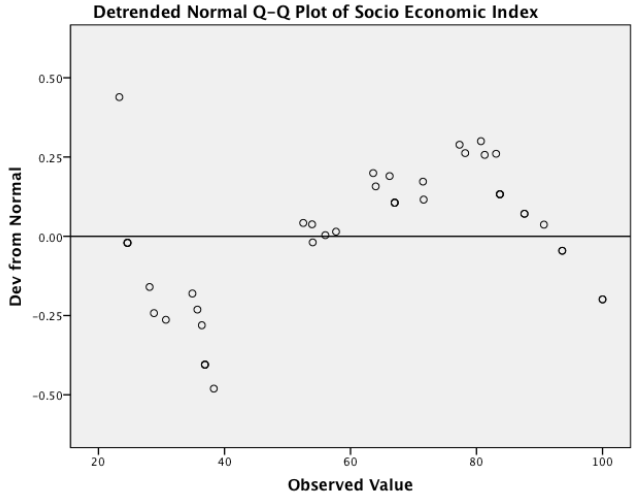
*. This is a lower bound of the true significance.

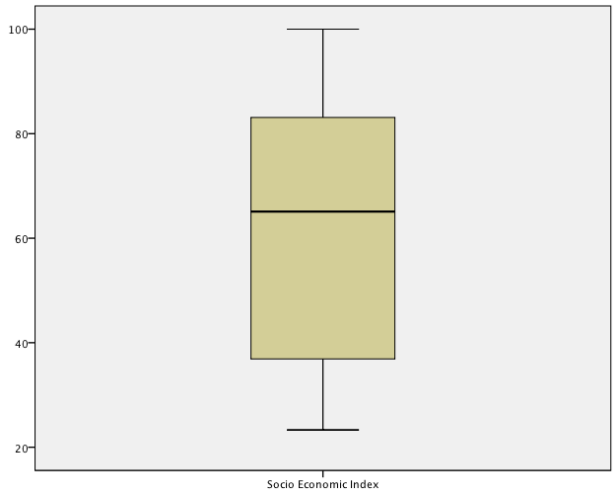
a. Lilliefors Significance Correction

Socio Economic Index

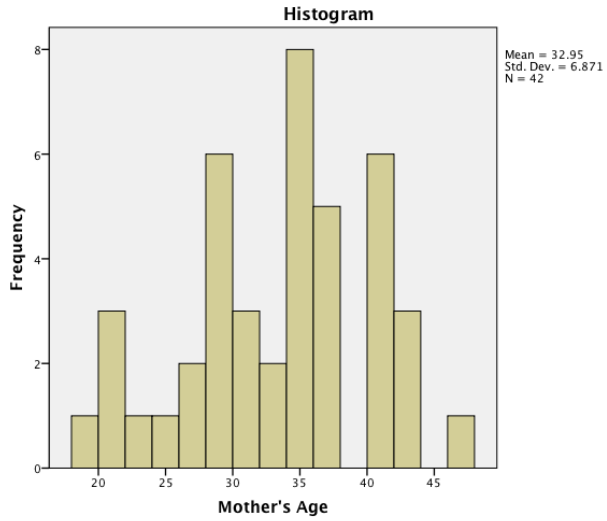


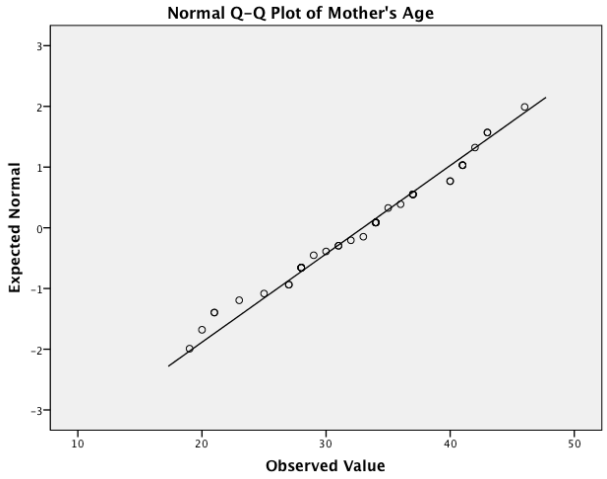


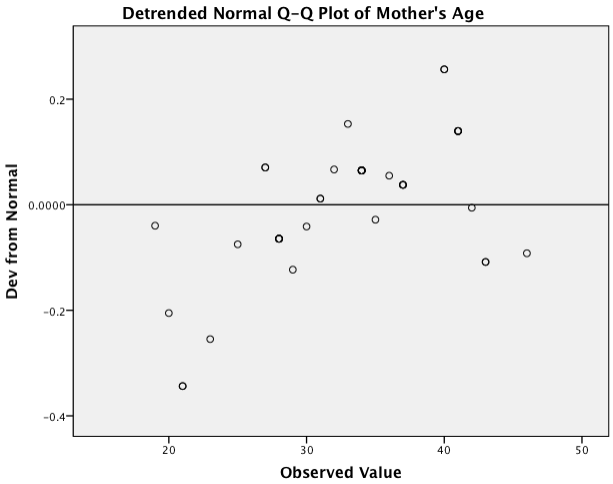


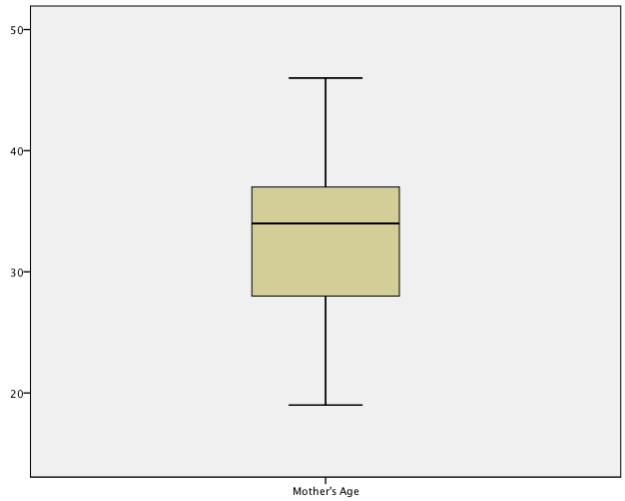


Mother's Age

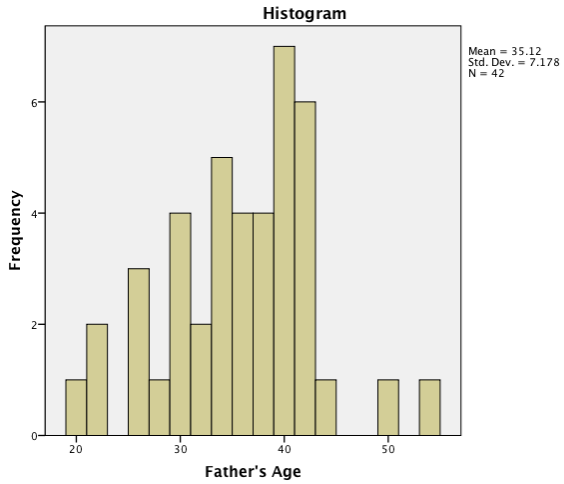

Histogram
Mean = 32.95
Std. Dev. = 6.871
N = 42
Frequency
Mother's Age

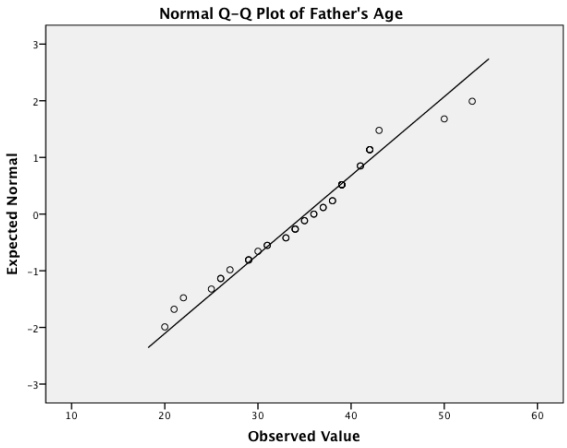

Normal Q-Q Plot of Mother's Age
Expected Normal
Observed Value

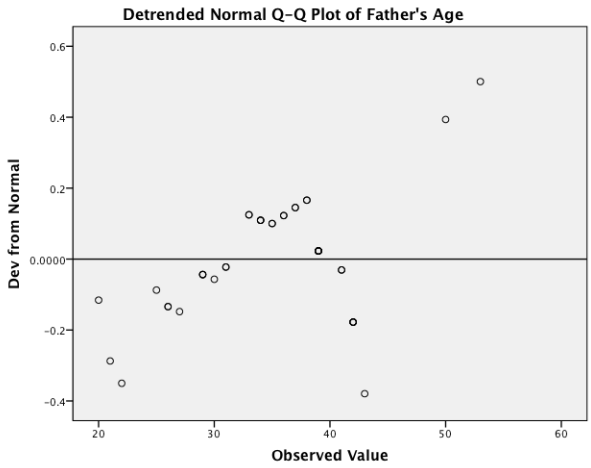

Detrended Normal Q-Q Plot of Mother's Age
Dev from Normal
Observed Value

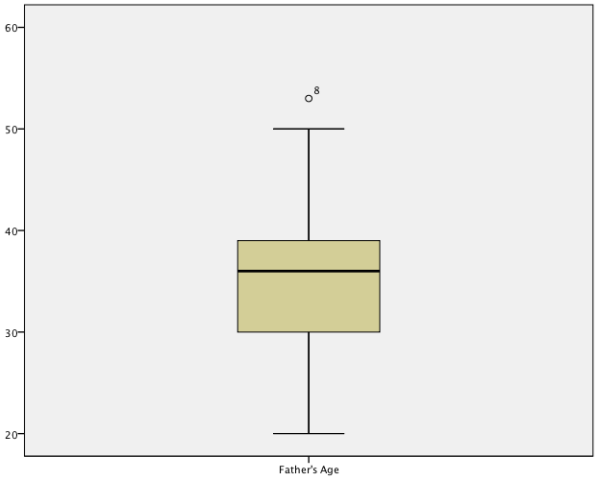

Mother's Age

Father's Age


Histogram
Mean = 35.12
Std. Dev. = 7.178
N = 42
Frequency
Father's Age


Normal Q-Q Plot of Father's Age
Expected Normal
Observed Value


Detrended Normal Q-Q Plot of Father's Age
Dev from Normal
Observed Value


Father's Age

Square Root Transformation Ratios of Outpatient Visits (OPD) and Days Admitted to Hospital (ADM)

Case Processing Summary

	Cases					
	Valid		Missing		Total	
	N	Percent	N	Percent	N	Percent
Sqrt transformation ratio OPD T1	42	100.0%	0	0.0%	42	100.0%
Sqrt transformation ratio OPD T2	37	88.1%	5	11.9%	42	100.0%
Sqrt transformation ratio ADM T1	42	100.0%	0	0.0%	42	100.0%
Sqrt transformation ratio ADM T2	37	88.1%	5	11.9%	42	100.0%

Descriptives

			Statistic	Std. Error
Sqrt transformation ratio OPD T1	Mean		.1960	.01102
	95% Confidence Interval for Mean	Lower Bound	.1738	
		Upper Bound	.2183	
	5% Trimmed Mean		.1945	
	Median		.1943	
	Variance		.005	
	Std. Deviation		.07139	
	Minimum		.00	
	Maximum		.42	
	Range		.42	
	Interquartile Range		.10	
	Skewness		.326	.365
	Kurtosis		1.915	.717
Sqrt transformation ratio OPD T2	Mean		.1859	.00976
	95% Confidence Interval for Mean	Lower Bound	.1661	
		Upper Bound	.2057	
	5% Trimmed Mean		.1840	
	Median		.1791	
	Variance		.004	
	Std. Deviation		.05937	
	Minimum		.10	
	Maximum		.31	
	Range		.21	
	Interquartile Range		.09	
	Skewness		.255	.388
	Kurtosis		-.765	.759
Sqrt transformation ratio ADM T1	Mean		.4352	.03617
	95% Confidence Interval for Mean	Lower Bound	.3621	
		Upper Bound	.5082	
	5% Trimmed Mean		.4349	
	Median		.4179	
	Variance		.055	
	Std. Deviation		.23442	
	Minimum		.00	
	Maximum		.99	
	Range		.99	
	Interquartile Range		.35	
	Skewness		.070	.365
	Kurtosis		-.378	.717
Sqrt transformation ratio ADM T2	Mean		.3150	.03046
	95% Confidence Interval for Mean	Lower Bound	.2532	
		Upper Bound	.3768	
	5% Trimmed Mean		.3122	
	Median		.3402	
	Variance		.034	
	Std. Deviation		.18527	
	Minimum		.00	
	Maximum		.70	
	Range		.70	
	Interquartile Range		.27	
	Skewness		.108	.388
	Kurtosis		-.643	.759

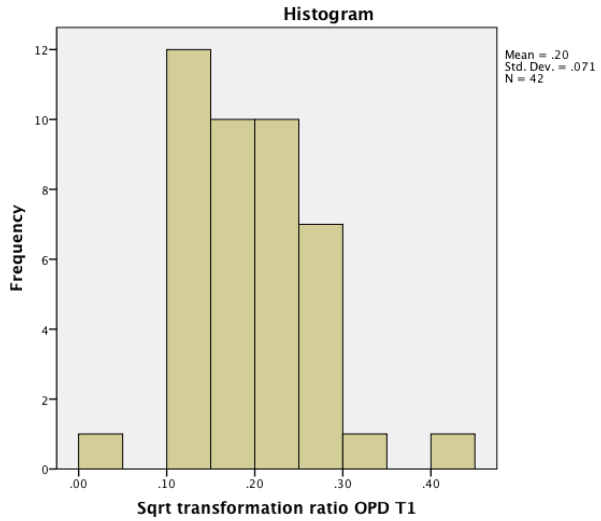
Tests of Normality

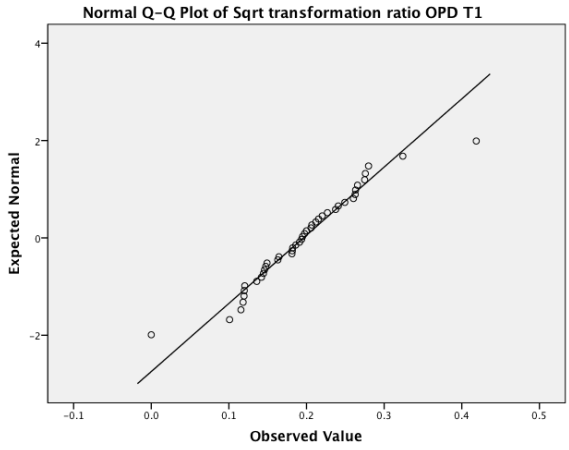
	Kolmogorov-Smirnov[a]		
	Statistic	df	Sig.
Sqrt transformation ratio OPD Time 1	.082	42	.200*
Sqrt transformation ratio OPD Time 2	.085	37	.200*
Sqrt transformation ratio ADM Time 1	.085	42	.200*
Sqrt transformation ratio ADM Time 2	.102	37	.200*

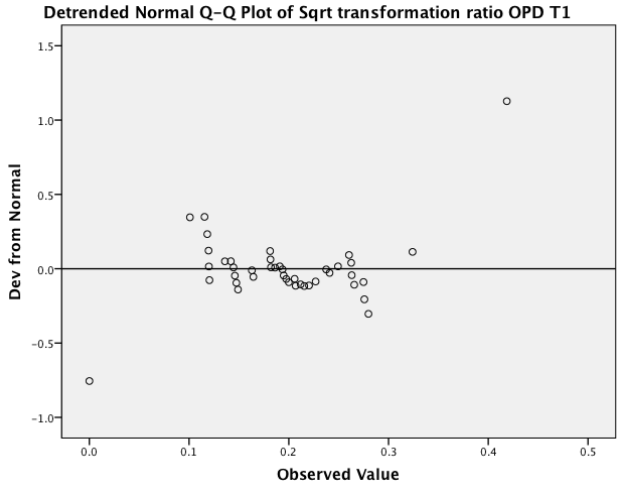
*. This is a lower bound of the true significance.

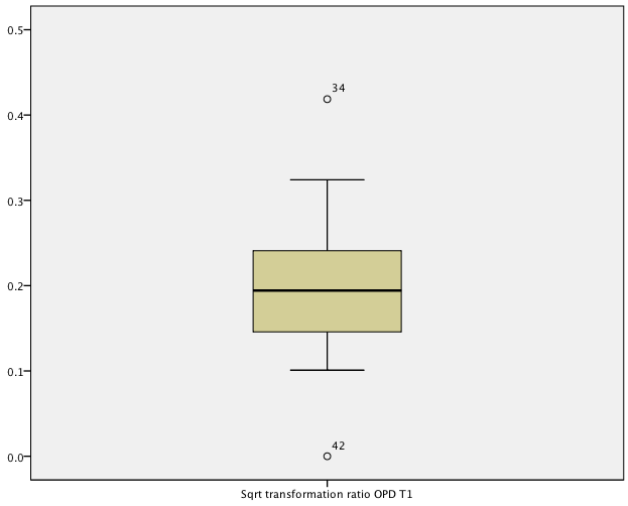
a. Lilliefors Significance Correction

Sqrt transformation ratio OPD Time 1

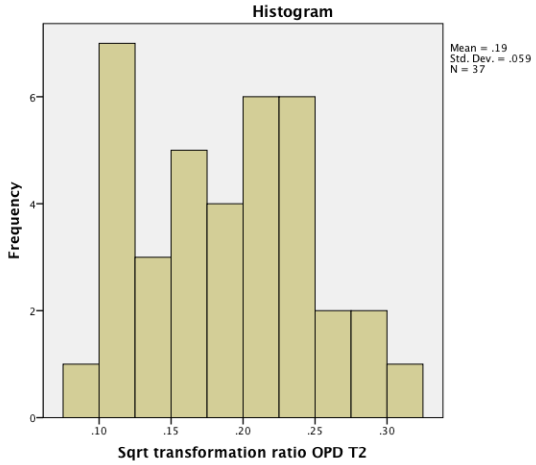


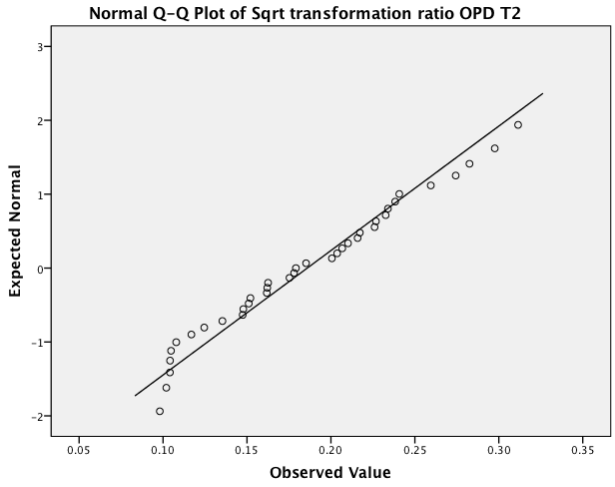


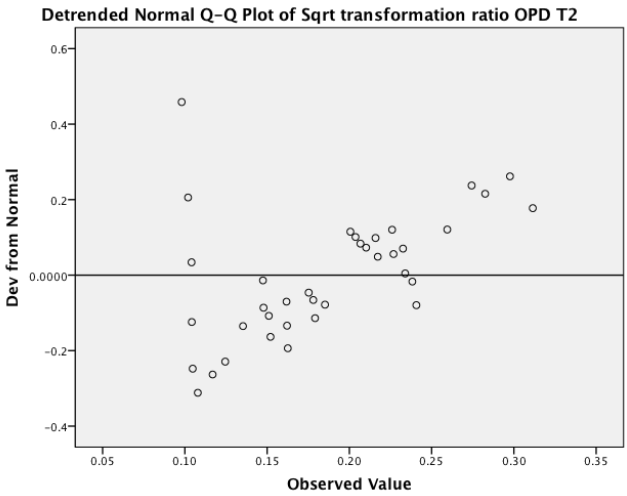


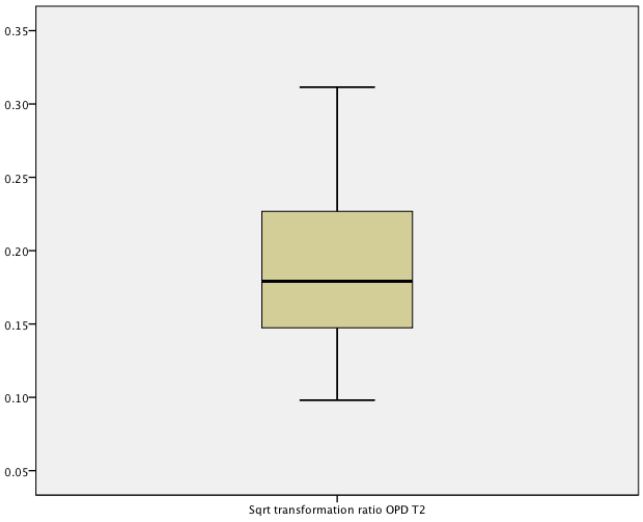


Sqrt transformation ratio OPD Time 2

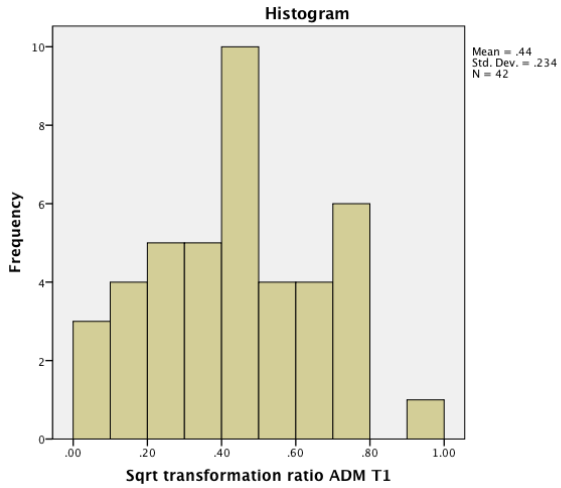


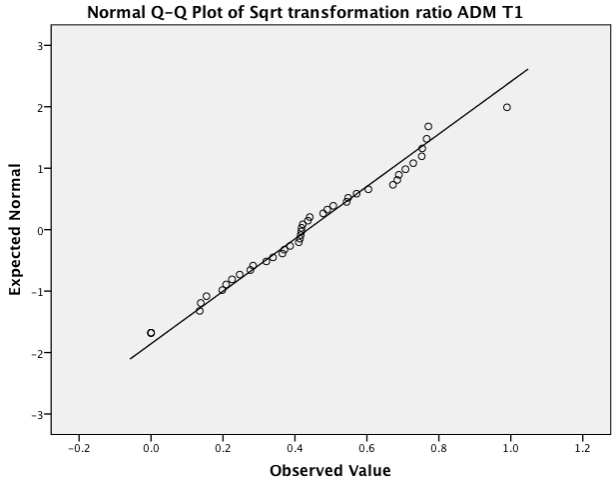


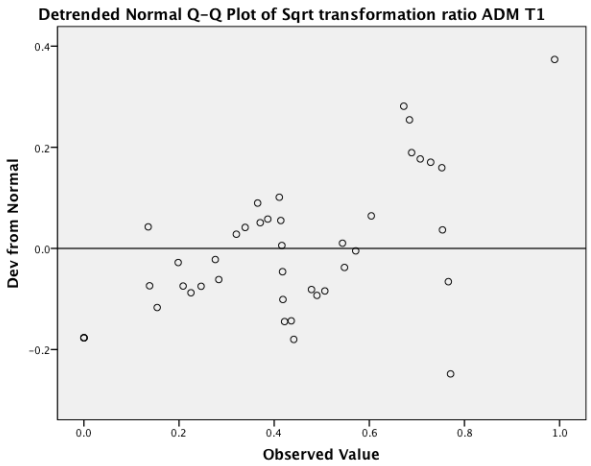


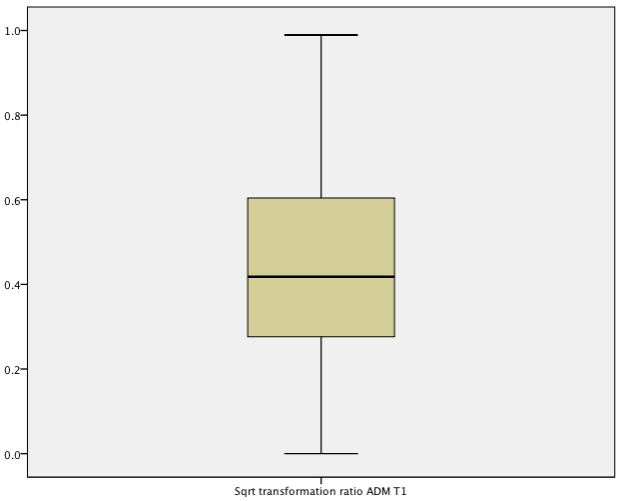


Sqrt transformation ratio ADM Time 1

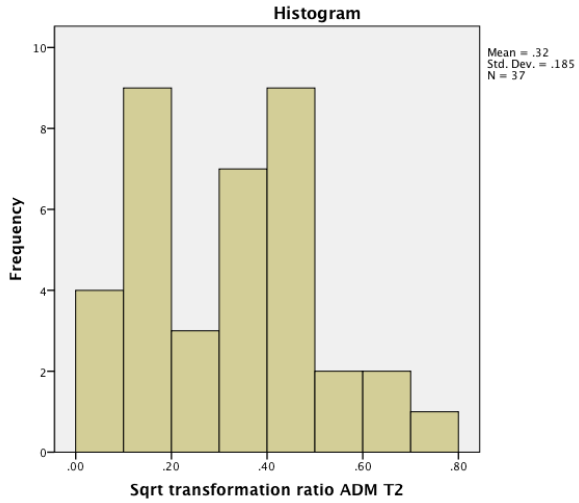


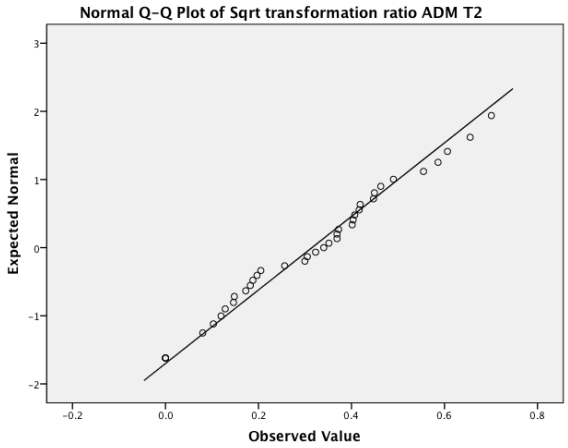


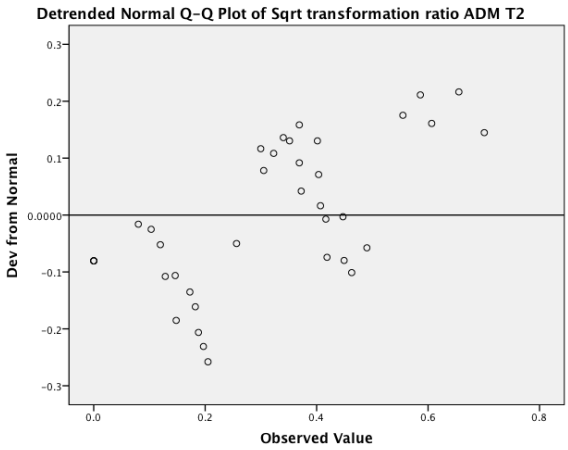


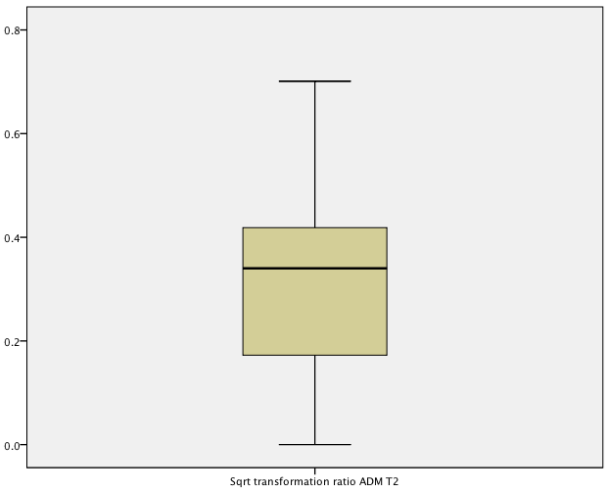


Sqrt transformation ratio ADM Time 2









Appendix G
Study Instruments Normality Assessment

DASS Total Raw Score

Case Processing Summary

	Cases					
	Valid		Missing		Total	
	N	Percent	N	Percent	N	Percent
Mother DASS Total Time 1	42	100.0%	0	0.0%	42	100.0%
Mother DASS Total Time 2	37	88.1%	5	11.9%	42	100.0%
Father DASS Total Time 1	42	100.0%	0	0.0%	42	100.0%
Father DASS Total Time 2	37	88.1%	5	11.9%	42	100.0%

DASS Scores Distribution and Normality

DASS Total Raw Score Descriptives

			Statistic	Std. Error
Mother DASS Total Time 1	Mean		23.14	3.373
	95% Confidence Interval for Mean	Lower Bound	16.33	
		Upper Bound	29.95	
	5% Trimmed Mean		20.97	
	Median		16.00	
	Variance		477.735	
	Std. Deviation		21.857	
	Minimum		0	
	Maximum		90	
	Range		90	
	Interquartile Range		21	
	Skewness		1.565	.365
	Kurtosis		2.085	.717
Mother DASS Total Time 2	Mean		22.46	2.822
	95% Confidence Interval for Mean	Lower Bound	16.74	
		Upper Bound	28.18	
	5% Trimmed Mean		21.58	
	Median		17.00	
	Variance		294.644	
	Std. Deviation		17.165	
	Minimum		0	
	Maximum		63	
	Range		63	
	Interquartile Range		26	
	Skewness		.794	.388
	Kurtosis		-.250	.759
Father DASS Total Time 1	Mean		16.79	3.348
	95% Confidence Interval for Mean	Lower Bound	10.02	
		Upper Bound	23.55	
	5% Trimmed Mean		13.59	
	Median		12.00	
	Variance		470.904	
	Std. Deviation		21.700	
	Minimum		0	
	Maximum		126	
	Range		126	
	Interquartile Range		16	
	Skewness		3.456	.365
	Kurtosis		15.565	.717
Father DASS Total Time 2	Mean		13.24	1.966
	95% Confidence Interval for Mean	Lower Bound	9.26	
		Upper Bound	17.23	
	5% Trimmed Mean		12.39	
	Median		10.00	
	Variance		143.078	
	Std. Deviation		11.962	
	Minimum		0	
	Maximum		46	
	Range		46	
	Interquartile Range		18	
	Skewness		.904	.388
	Kurtosis		.153	.759

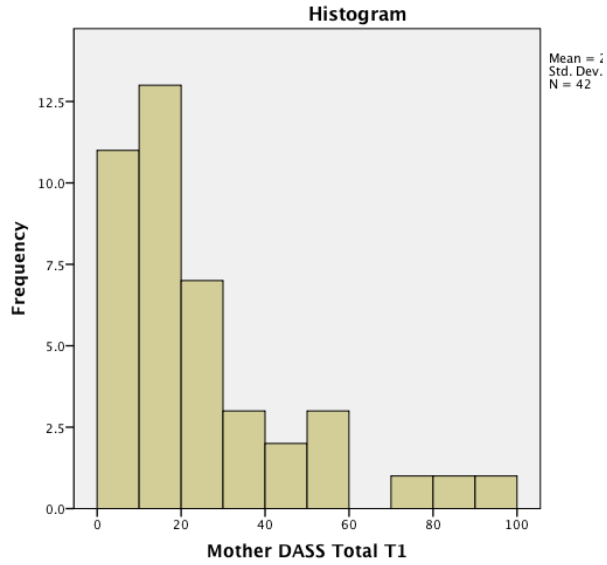
DASS Total Raw Score

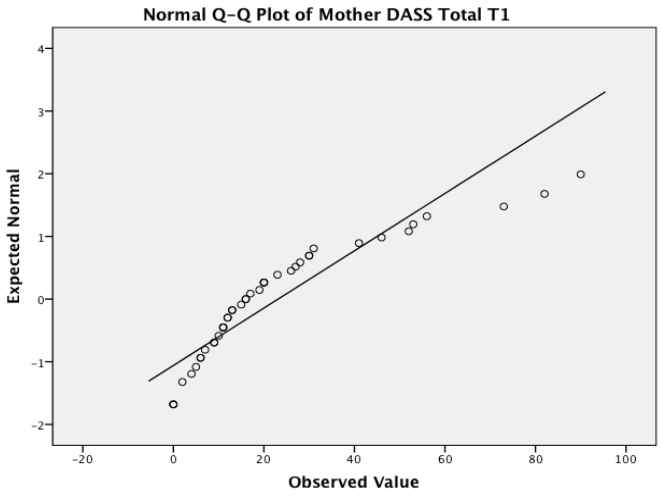
Tests of Normality

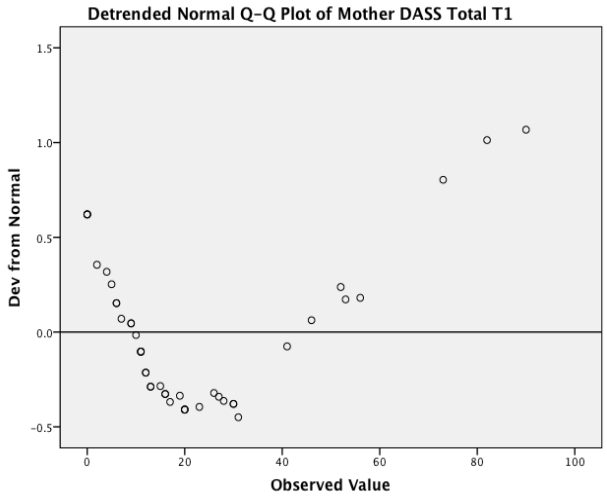
	Kolmogorov-Smirnov[a]		
	Statistic	df	Sig.
Mother DASS Total Time 1	.200	42	.000
Mother DASS Total Time 2	.138	37	.071
Father DASS Total Time 1	.234	42	.000
Father DASS Total Time 2	.214	37	.000

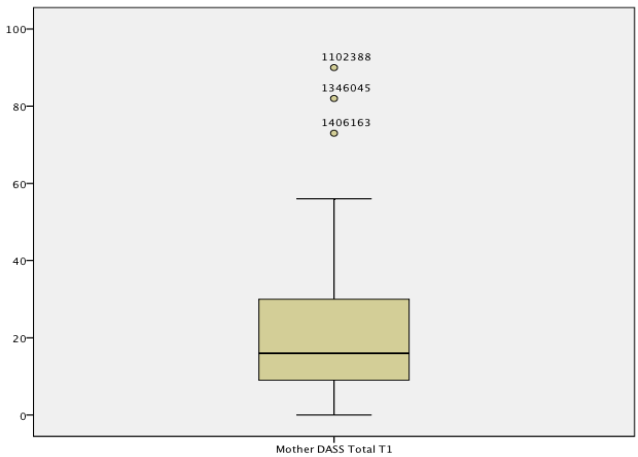
a. Lilliefors Significance Correction

Mother DASS Total Raw Score Time 1

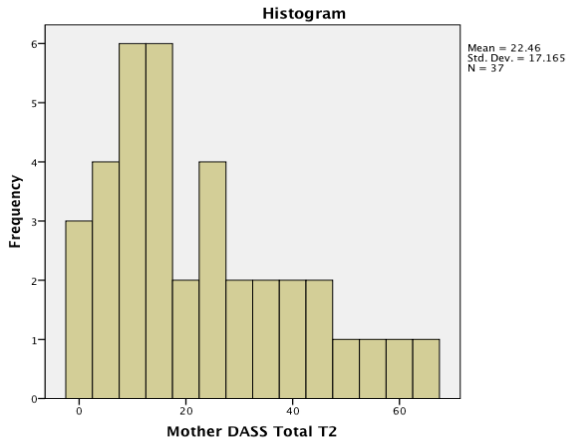




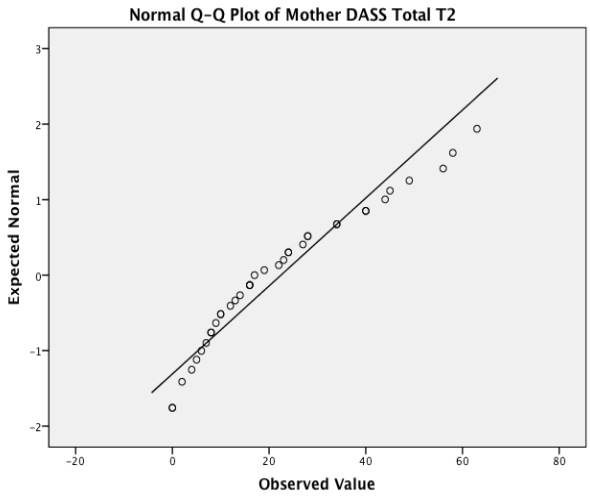


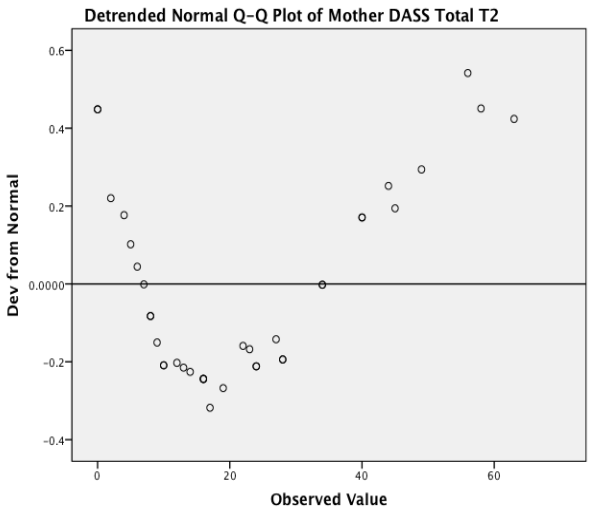


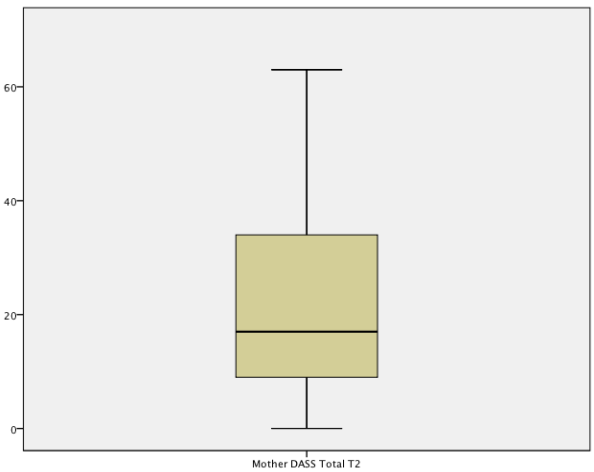
Mother DASS Total Raw Score Time 2



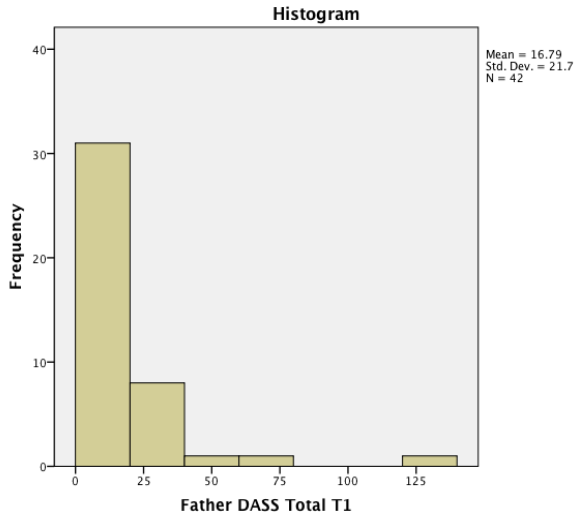
Mother DASS Total Raw Score Time 2 cont.

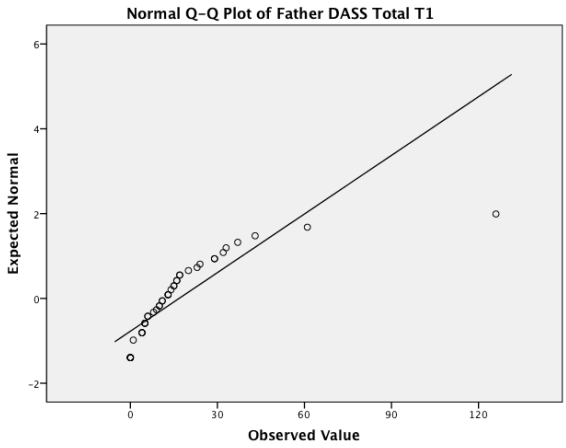


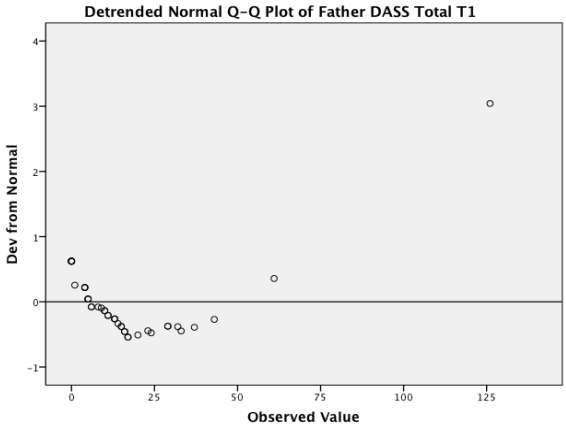




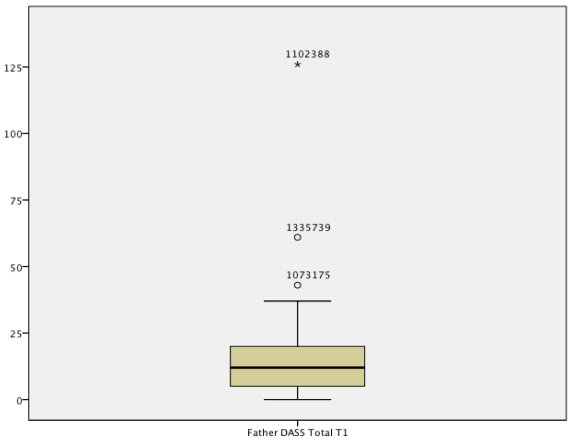
Father DASS Total Raw Score Time 1







Father DASS Total Raw Score Time 1 cont.



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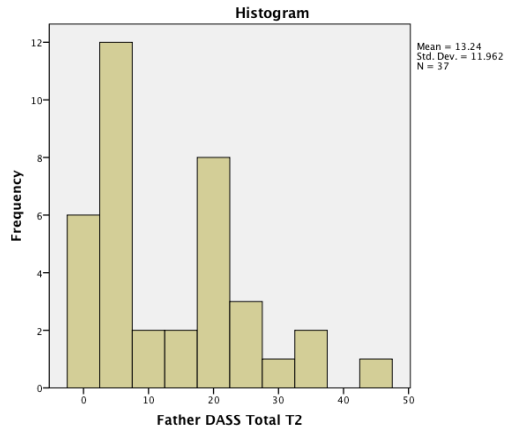
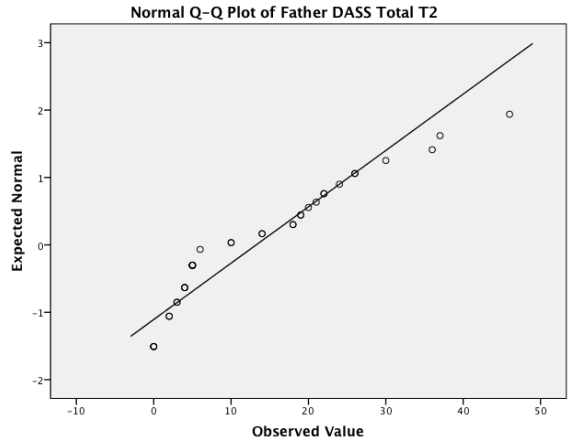
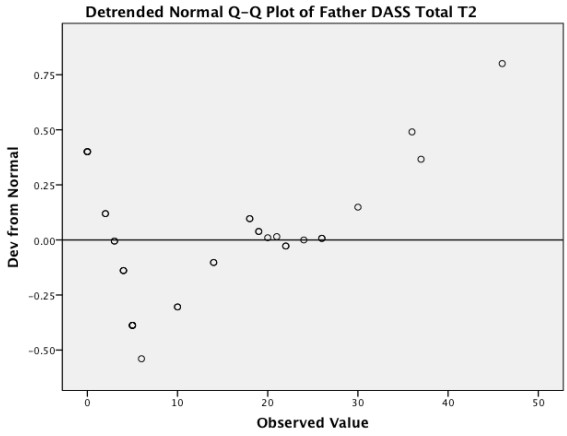
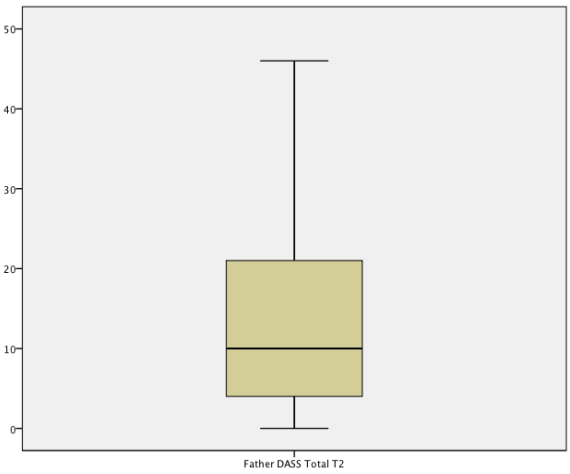








DASS Total Percentile Score

Case Processing Summary

	Cases					
	Valid		Missing		Total	
	N	Percent	N	Percent	N	Percent
Mother DASS Total Percentile Time 1	42	100.0%	0	0.0%	42	100.0%
Mother DASS Total Percentile Time 2	37	88.1%	5	11.9%	42	100.0%
Father DASS Total Percentile Time 1	42	100.0%	0	0.0%	42	100.0%
Father DASS Total Percentile Time 2	37	88.1%	5	11.9%	42	100.0%

DASS Percentile Score Descriptives

			Statistic	Std. Error
Mother DASS Total Percentile Time 1	Mean		57.29	4.391
	95% Confidence Interval for Mean	Lower Bound	48.42	
		Upper Bound	66.15	
	5% Trimmed Mean		57.90	
	Median		60.00	
	Variance		809.672	
	Std. Deviation		28.455	
	Minimum		5	
	Maximum		99	
	Range		94	
	Interquartile Range		43	
	Skewness		-.254	.365
	Kurtosis		-.949	.717
Mother DASS Total Percentile Time 2	Mean		59.76	4.642
	95% Confidence Interval for Mean	Lower Bound	50.34	
		Upper Bound	69.17	
	5% Trimmed Mean		60.81	
	Median		60.00	
	Variance		797.300	
	Std. Deviation		28.237	
	Minimum		5	
	Maximum		96	
	Range		91	
	Interquartile Range		49	
	Skewness		-.441	.388
	Kurtosis		-.997	.759
Father DASS Total Percentile Time 1	Mean		45.93	4.497
	95% Confidence Interval for Mean	Lower Bound	36.85	
		Upper Bound	55.01	
	5% Trimmed Mean		45.38	
	Median		47.50	
	Variance		849.385	
	Std. Deviation		29.144	
	Minimum		5	
	Maximum		99	
	Range		94	
	Interquartile Range		48	
	Skewness		.123	.365
	Kurtosis		-1.124	.717
Father DASS Total Percentile Time 2	Mean		42.76	4.828
	95% Confidence Interval for Mean	Lower Bound	32.97	
		Upper Bound	52.55	
	5% Trimmed Mean		42.22	
	Median		40.00	
	Variance		862.300	
	Std. Deviation		29.365	
	Minimum		5	
	Maximum		92	
	Range		87	
	Interquartile Range		50	
	Skewness		.197	.388
	Kurtosis		-1.585	.759

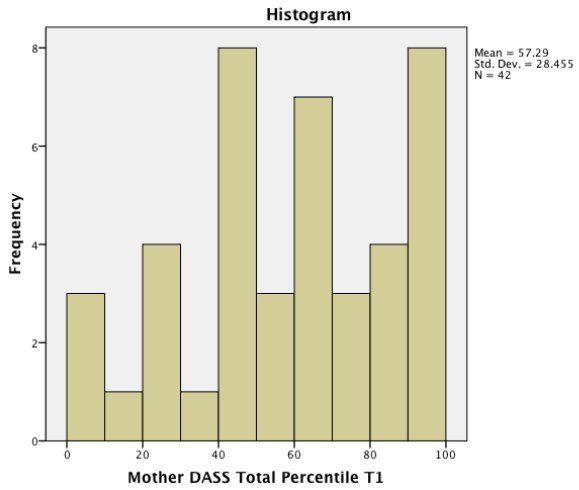
DASS Percentile Score

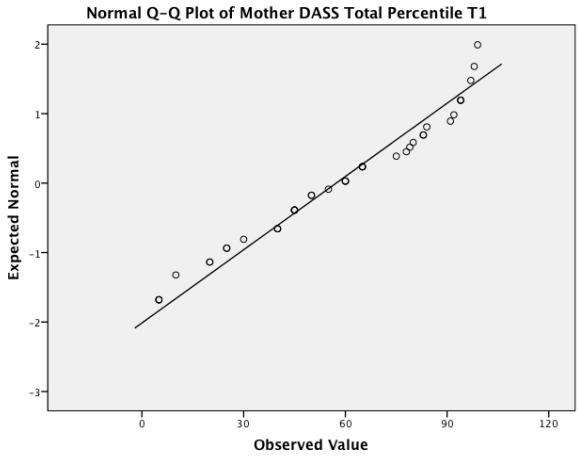
Tests of Normality

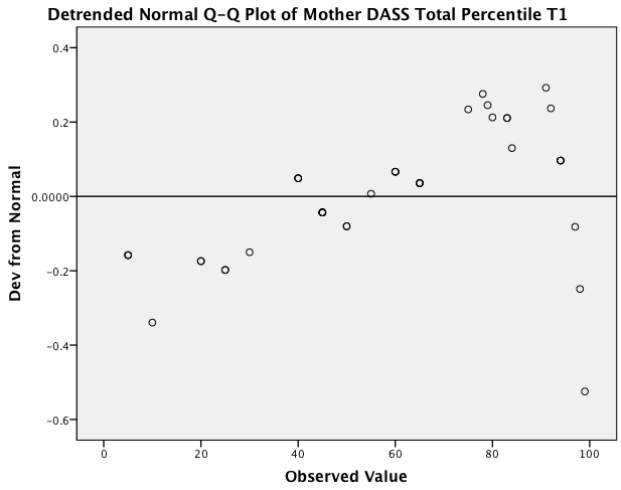
	Kolmogorov-Smirnov[a]		
	Statistic	df	Sig.
Mother DASS Total Percentile Time 1	.100	42	.200*
Mother DASS Total Percentile Time 2	.138	37	.074
Father DASS Total Percentile Time 1	.123	42	.115
Father DASS Total Percentile Time 2	.240	37	.000

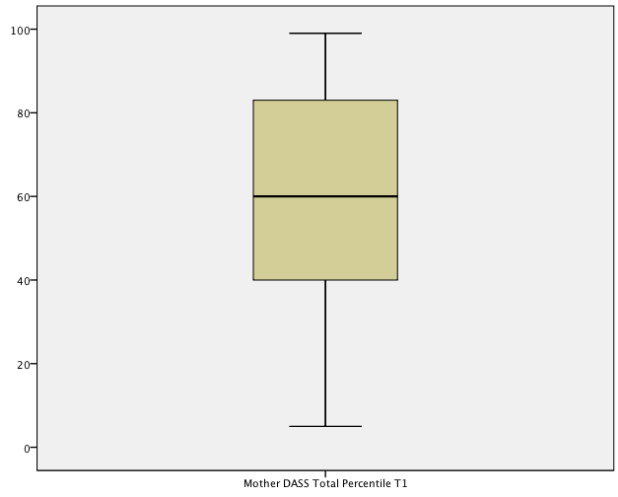
*. This is a lower bound of the true significance.
a. Lilliefors Significance Correction

Mother DASS Total Percentile Time 1

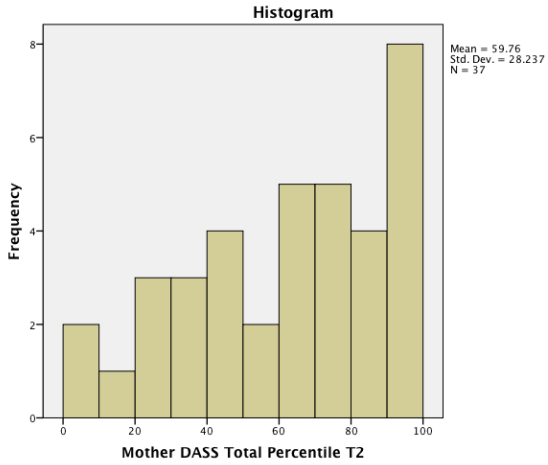








Mother DASS Total Percentile Time 2



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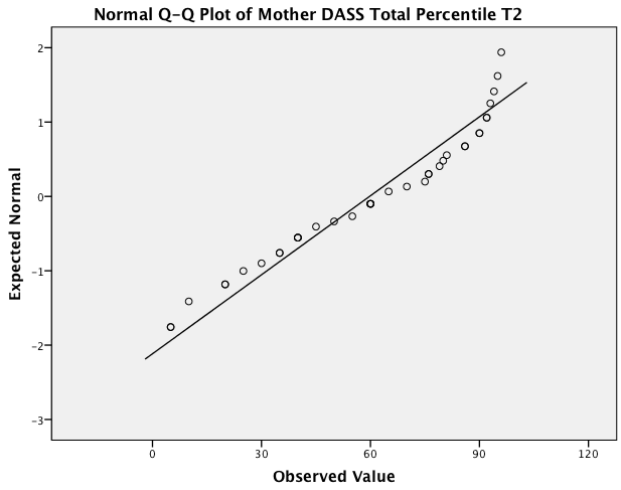
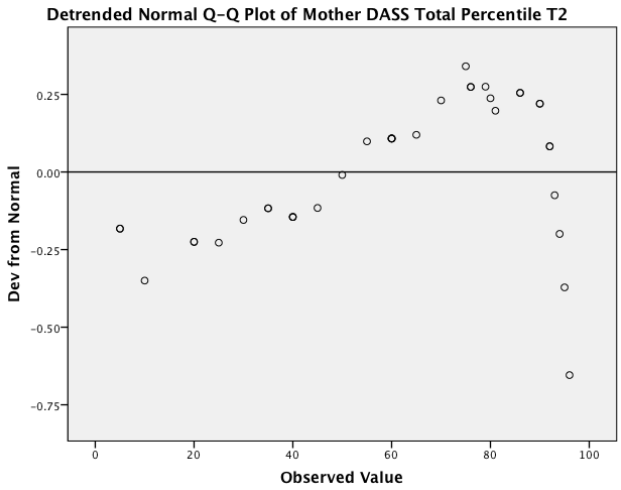
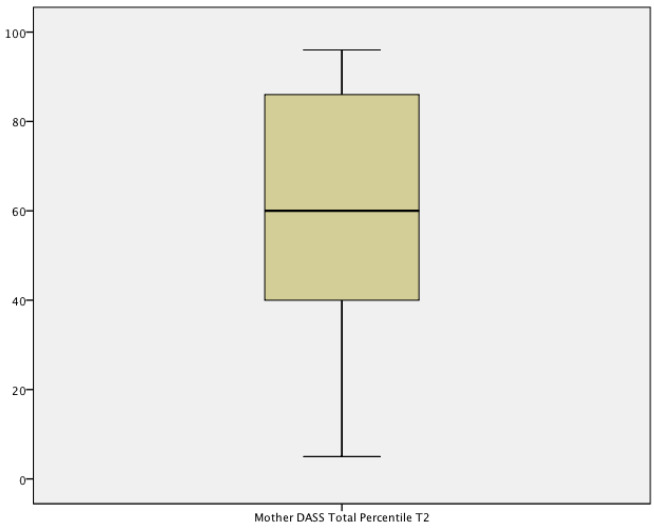
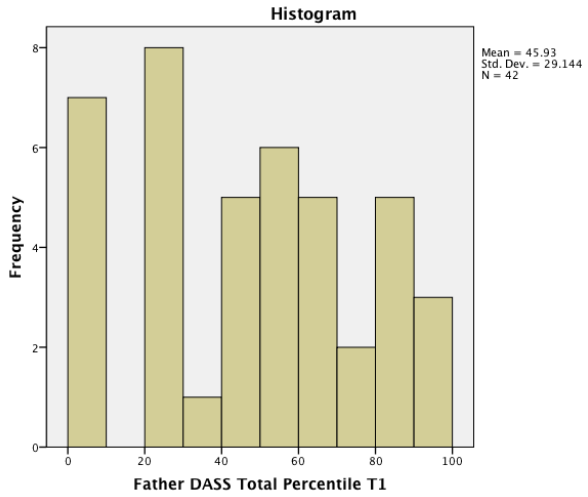
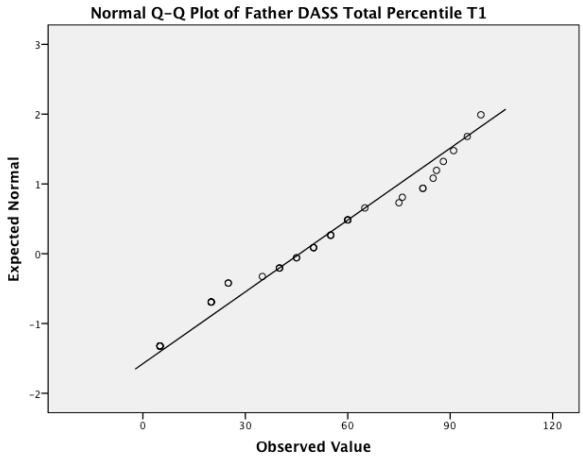


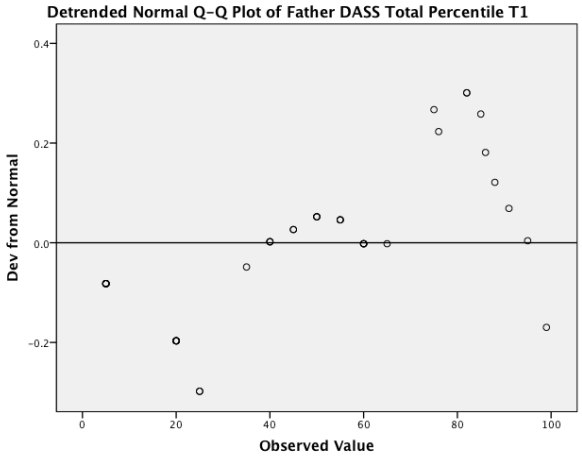


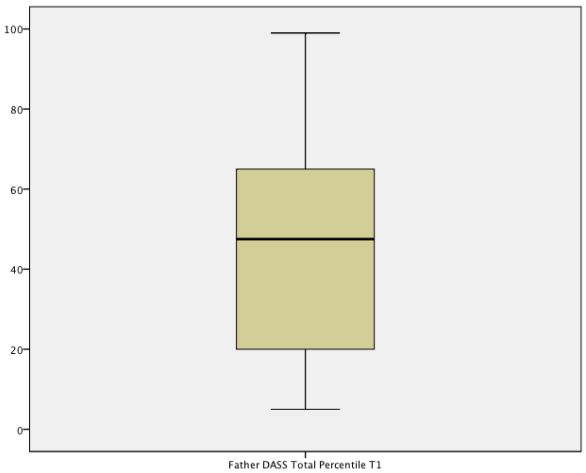


Father DASS Total Percentile Time 1

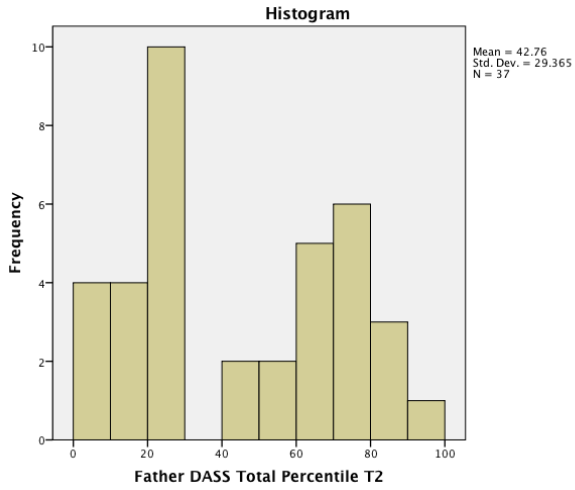


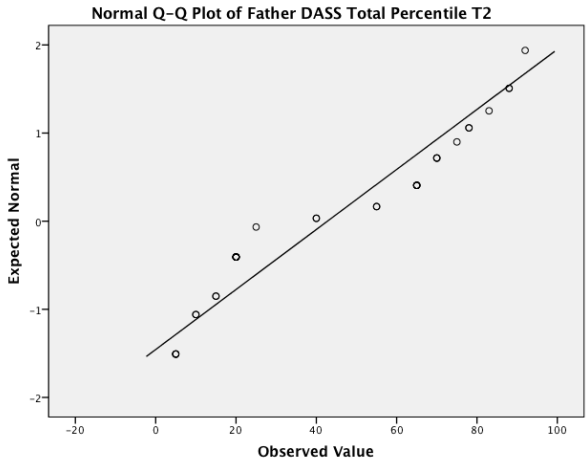


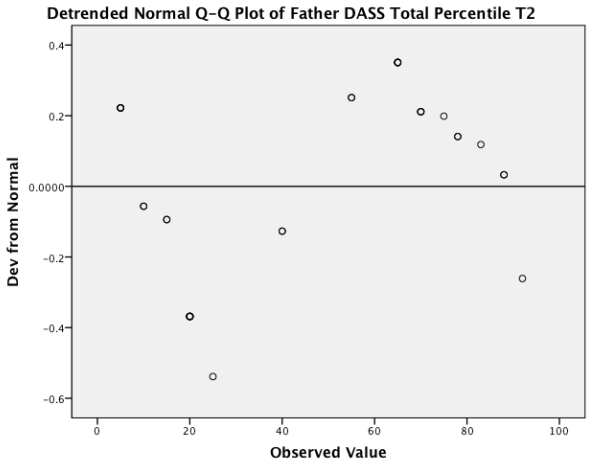


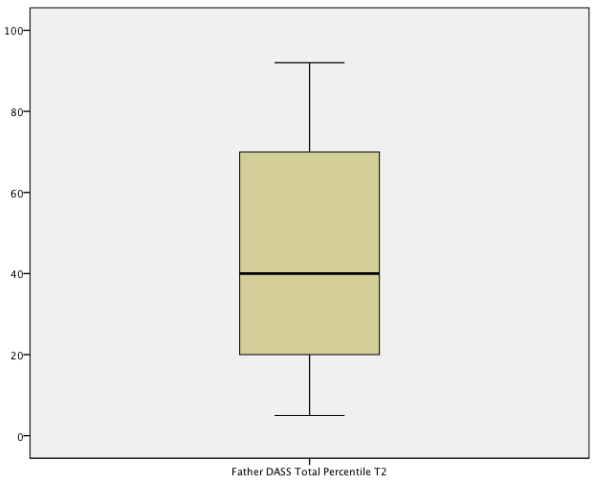


Father DASS Total Percentile Time 2









FAD Scores Distribution and Normality

Case Processing Summary

	Cases					
	Valid		Missing		Total	
	N	Percent	N	Percent	N	Percent
Mother FAD General Functioning Time 1	42	100.0%	0	0.0%	42	100.0%
Mother FAD General Functioning Time 2	37	88.1%	5	11.9%	42	100.0%
Father FAD General Functioning Time 1	42	100.0%	0	0.0%	42	100.0%
Father FAD General Functioning Time 2	37	88.1%	5	11.9%	42	100.0%

Descriptives

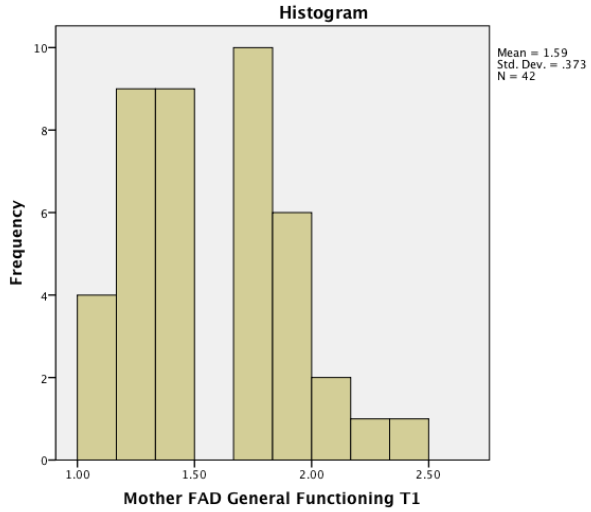
			Statistic	Std. Error
Mother FAD General Functioning Time 1	Mean		1.5934	.05763
	95% Confidence Interval for Mean	Lower Bound	1.4770	
		Upper Bound	1.7098	
	5% Trimmed Mean		1.5837	
	Median		1.5000	
	Variance		.139	
	Std. Deviation		.37348	
	Minimum		1.00	
	Maximum		2.50	
	Range		1.50	
	Interquartile Range		.60	
	Skewness		.221	.365
	Kurtosis		-.703	.717
Mother FAD General Functioning Time 2	Mean		1.6078	.07207
	95% Confidence Interval for Mean	Lower Bound	1.4617	
		Upper Bound	1.7540	
	5% Trimmed Mean		1.5941	
	Median		1.5800	
	Variance		.192	
	Std. Deviation		.43841	
	Minimum		1.00	
	Maximum		2.50	
	Range		1.50	
	Interquartile Range		.67	
	Skewness		.471	.388
	Kurtosis		-.862	.759
Father FAD General Functioning Time 1	Mean		1.6117	.06336
	95% Confidence Interval for Mean	Lower Bound	1.4837	
		Upper Bound	1.7396	
	5% Trimmed Mean		1.5922	
	Median		1.5400	
	Variance		.169	
	Std. Deviation		.41063	
	Minimum		1.00	
	Maximum		2.75	
	Range		1.75	
	Interquartile Range		.59	
	Skewness		.509	.365
	Kurtosis		.025	.717
Father FAD General Functioning Time 2	Mean		1.6541	.06399
	95% Confidence Interval for Mean	Lower Bound	1.5243	
		Upper Bound	1.7838	
	5% Trimmed Mean		1.6443	
	Median		1.7500	
	Variance		.152	
	Std. Deviation		.38926	
	Minimum		1.00	
	Maximum		2.75	
	Range		1.75	
	Interquartile Range		.59	
	Skewness		.211	.388
	Kurtosis		.195	.759

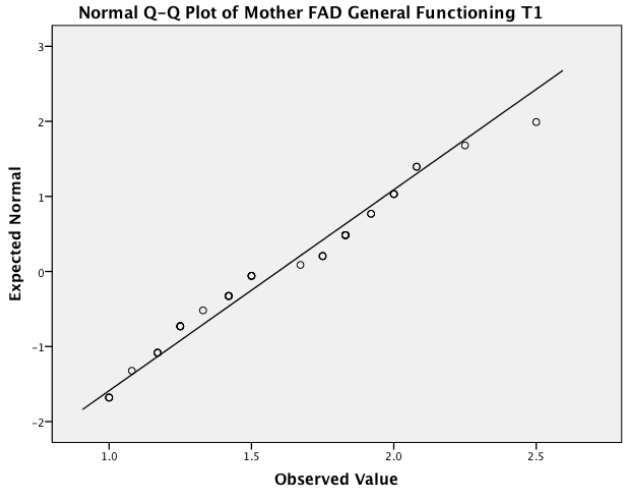
Tests of Normality

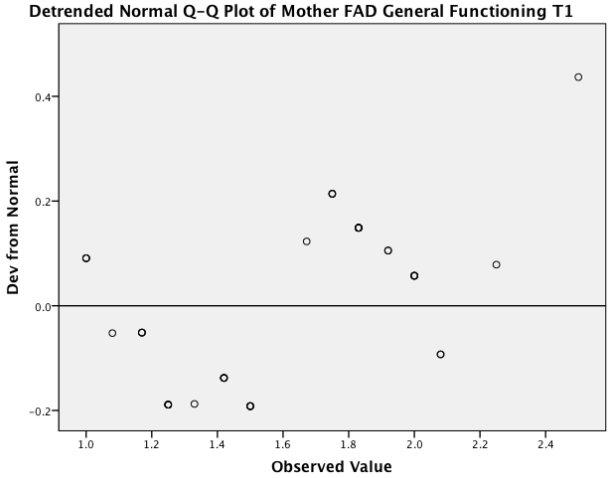
	Kolmogorov-Smirnov[a]		
	Statistic	df	Sig.
Mother FAD General Functioning Time 1	.123	42	.117
Mother FAD General Functioning Time 2	.144	37	.050
Father FAD General Functioning Time 1	.107	42	.200*
Father FAD General Functioning Time 2	.165	37	.012

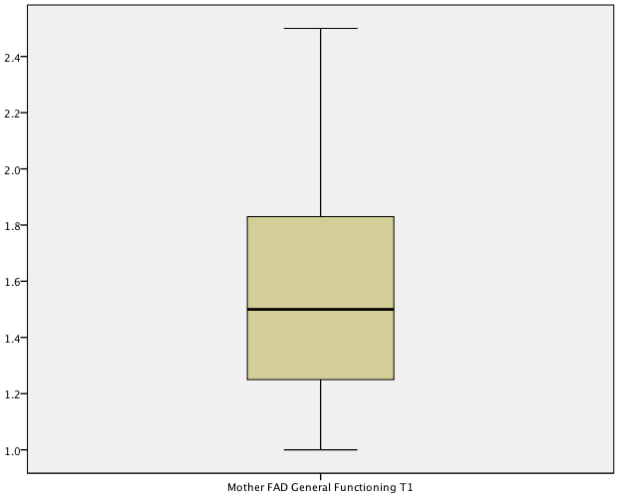
*. This is a lower bound of the true significance.
a. Lilliefors Significance Correction

Mother FAD General Functioning Time 1

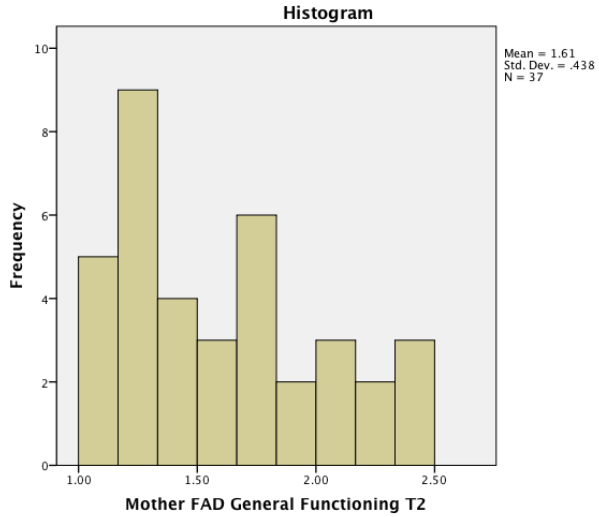


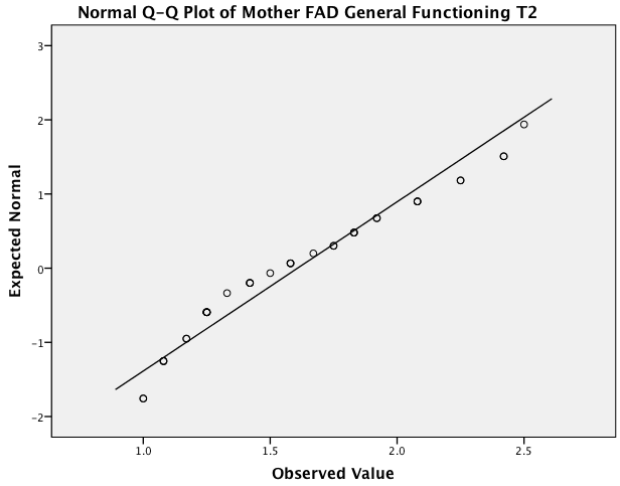


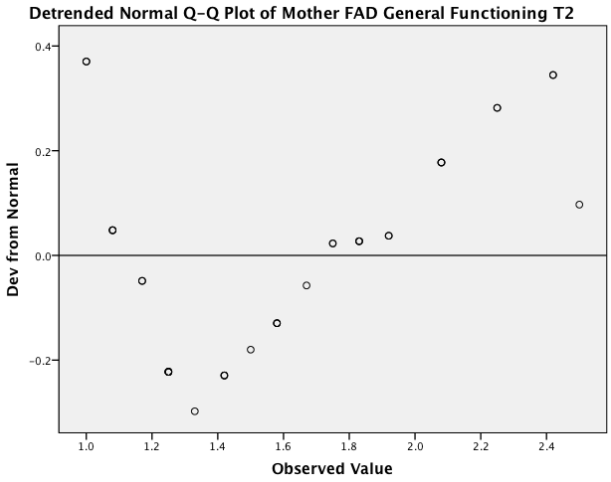


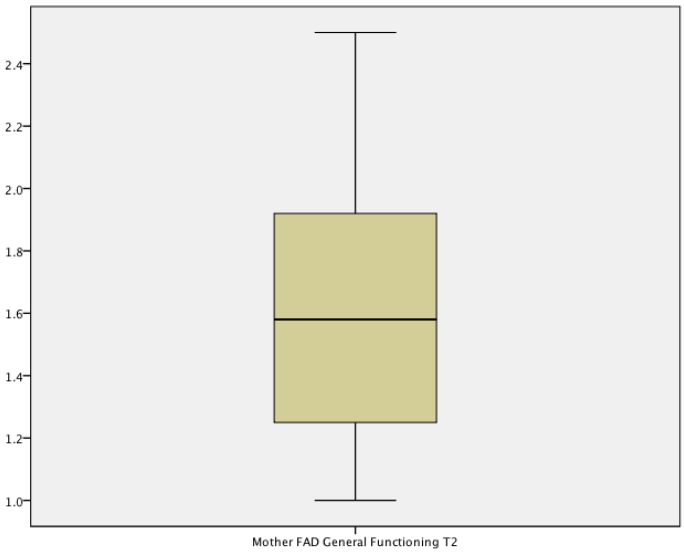


Mother FAD General Functioning Time 2

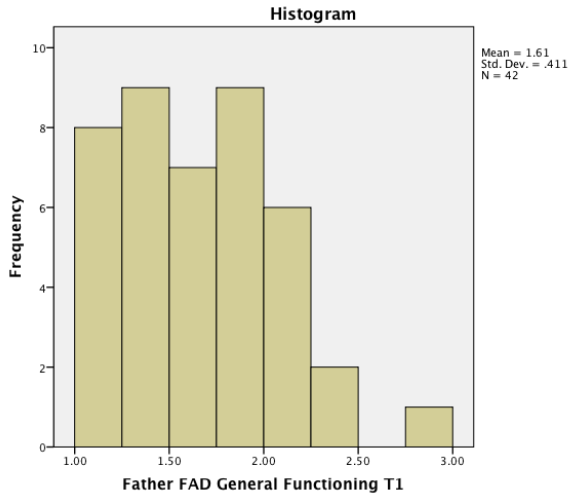


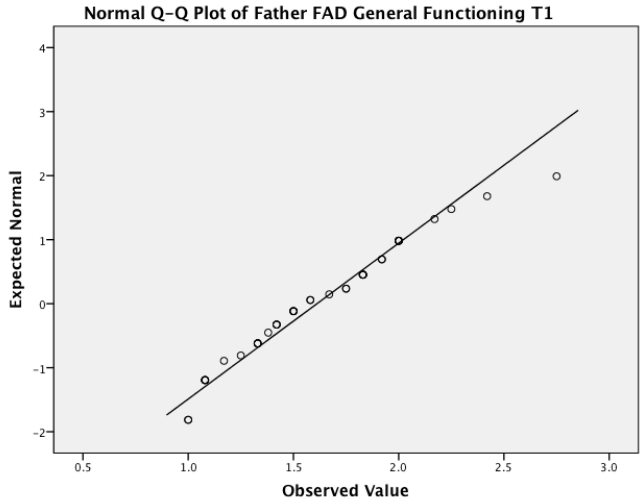


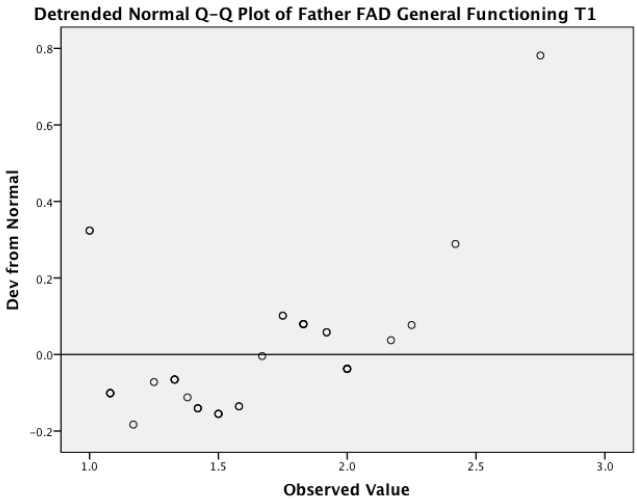


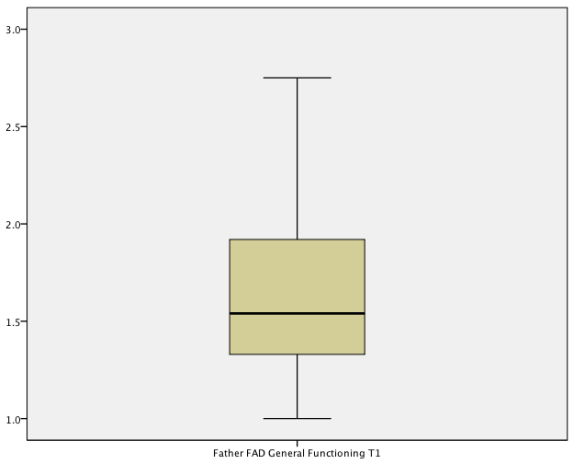


Father FAD General Functioning Time 1

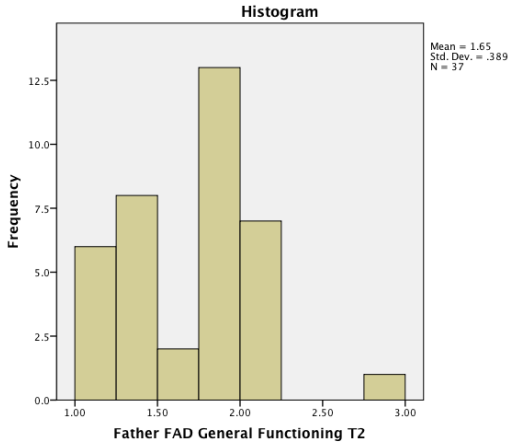


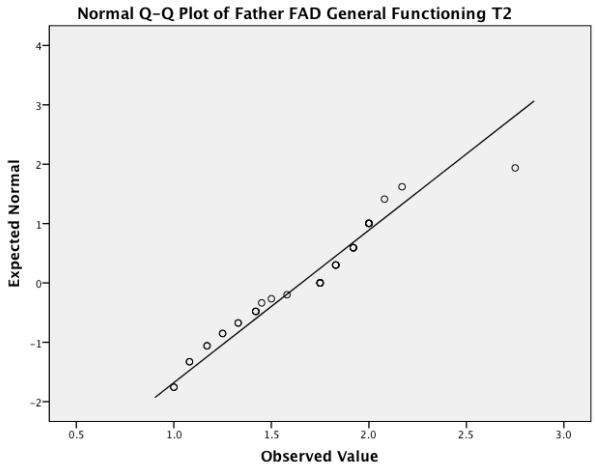


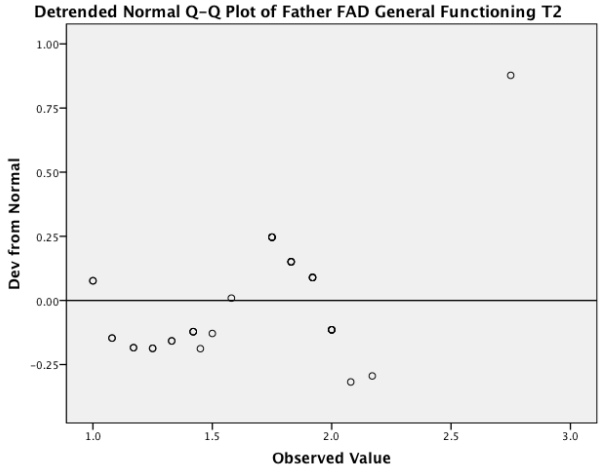




Father FAD General Functioning Time 2







IFS 15-item Total Scores Distribution and Normality

Case Processing Summary

	Cases					
	Valid		Missing		Total	
	N	Percent	N	Percent	N	Percent
Mother IFS 15 item total Time 1	42	100.0%	0	0.0%	42	100.0%
Mother IFS 15 item total Time 2	36	85.7%	6	14.3%	42	100.0%
Father IFS 15 item total Time 1	42	100.0%	0	0.0%	42	100.0%
Father IFS 15 item total Time 2	37	88.1%	5	11.9%	42	100.0%

Descriptives

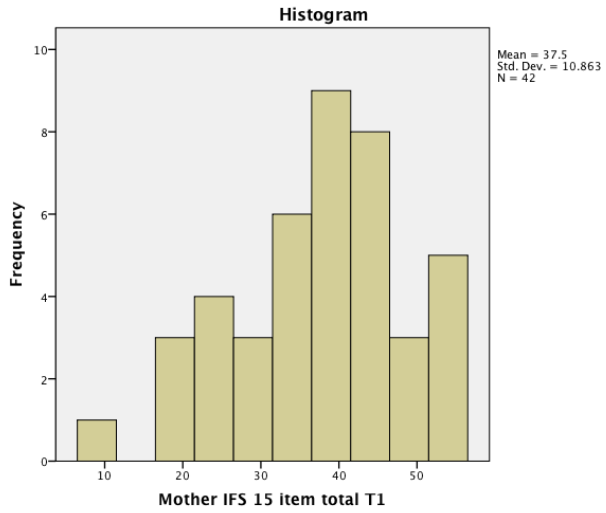
			Statistic	Std. Error
Mother IFS 15 item total Time 1	Mean		37.50	1.676
	95% Confidence Interval for Mean	Lower Bound	34.11	
		Upper Bound	40.89	
	5% Trimmed Mean		37.85	
	Median		38.00	
	Variance		118.012	
	Std. Deviation		10.863	
	Minimum		9	
	Maximum		56	
	Range		47	
	Interquartile Range		13	
	Skewness		-.475	.365
	Kurtosis		.054	.717
Mother IFS 15 item total Time 2	Mean		35.92	2.068
	95% Confidence Interval for Mean	Lower Bound	31.72	
		Upper Bound	40.12	
	5% Trimmed Mean		35.23	
	Median		35.00	
	Variance		153.964	
	Std. Deviation		12.408	
	Minimum		15	
	Maximum		75	
	Range		60	
	Interquartile Range		16	
	Skewness		.831	.393
	Kurtosis		1.398	.768
Father IFS 15 item total Time 1	Mean		34.24	1.362
	95% Confidence Interval for Mean	Lower Bound	31.49	
		Upper Bound	36.99	
	5% Trimmed Mean		34.46	
	Median		35.50	
	Variance		77.942	
	Std. Deviation		8.828	
	Minimum		13	
	Maximum		51	
	Range		38	
	Interquartile Range		13	
	Skewness		-.385	.365
	Kurtosis		-.529	.717
Father IFS 15 item total Time 2	Mean		33.41	1.940
	95% Confidence Interval for Mean	Lower Bound	29.47	
		Upper Bound	37.34	
	5% Trimmed Mean		33.01	
	Median		31.00	
	Variance		139.192	
	Std. Deviation		11.798	
	Minimum		15	
	Maximum		59	
	Range		44	
	Interquartile Range		19	
	Skewness		.296	.388
	Kurtosis		-.490	.759

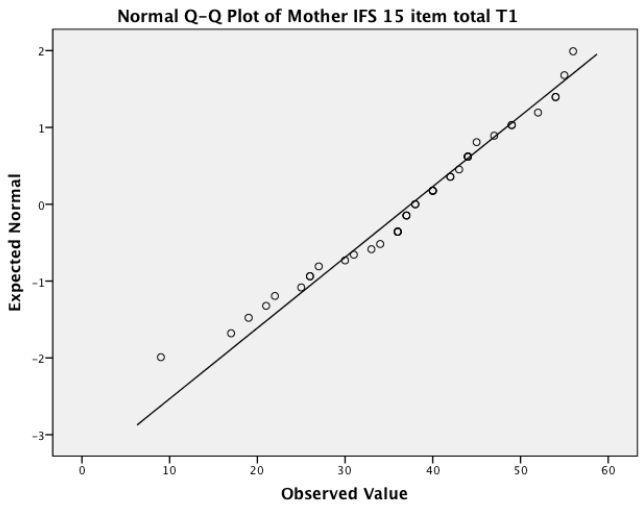
Tests of Normality

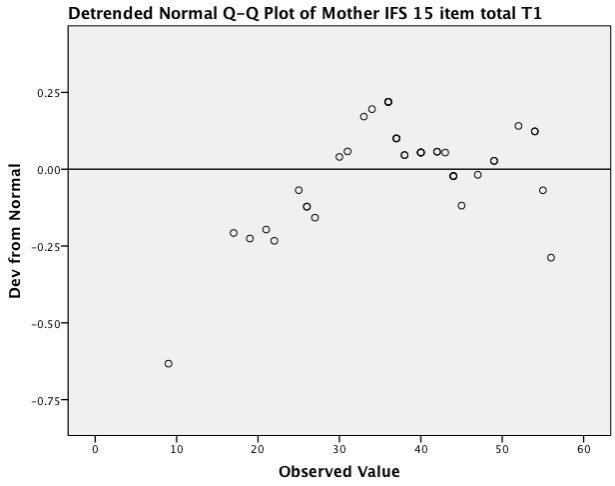
	Kolmogorov-Smirnov[a]		
	Statistic	df	Sig.
Mother IFS 15 item total Time 1	.136	42	.051
Mother IFS 15 item total Time 2	.126	36	.158
Father IFS 15 item total Time 1	.117	42	.162
Father IFS 15 item total Time 2	.097	37	.200*

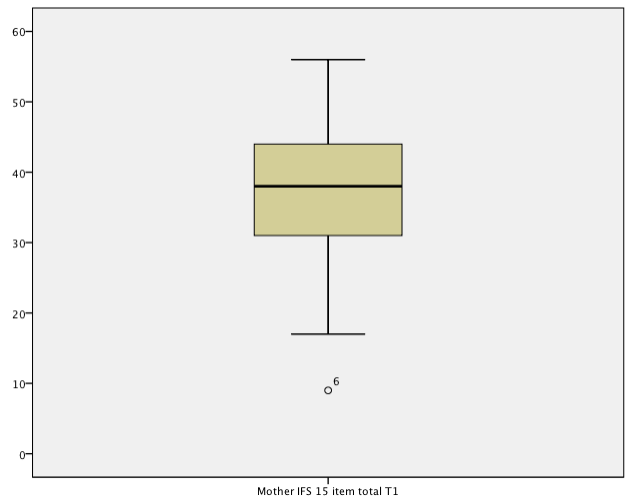
*. This is a lower bound of the true significance.
a. Lilliefors Significance Correction

Mother IFS 15 item total Time 1

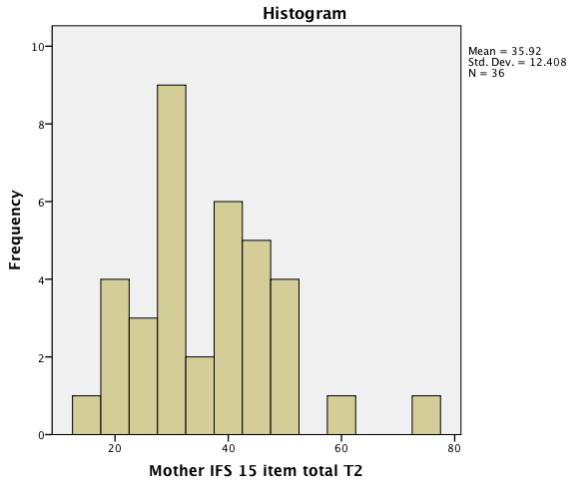


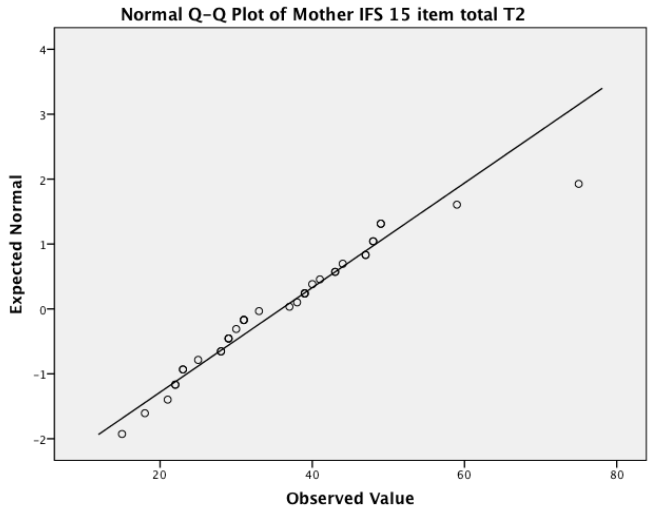


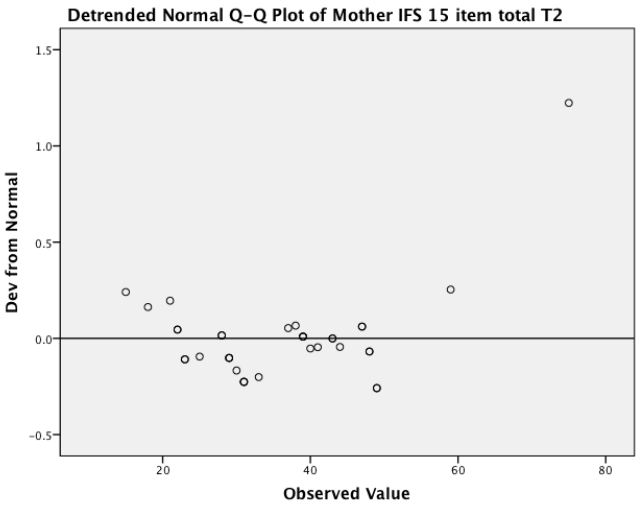


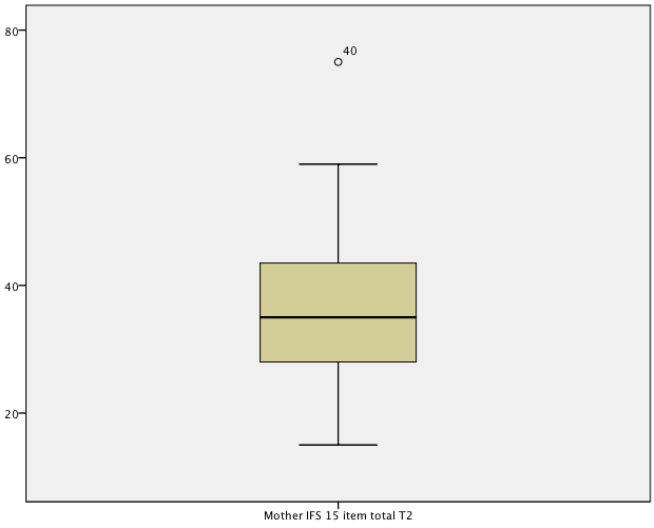


Mother IFS 15 item total Time 2

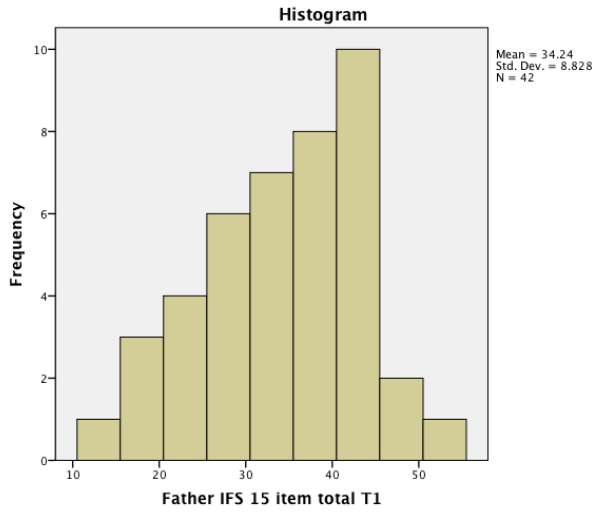


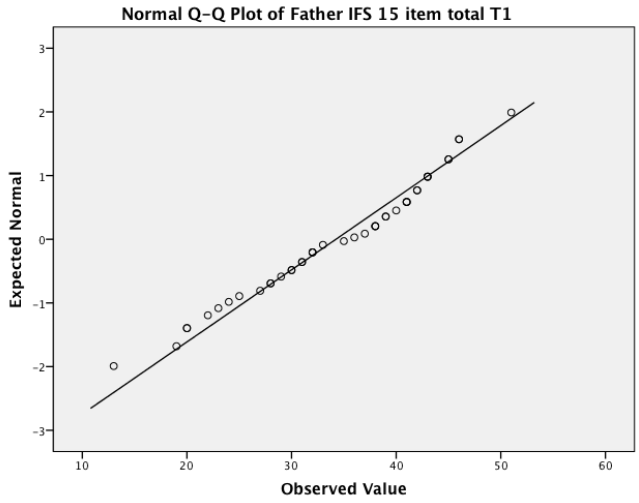


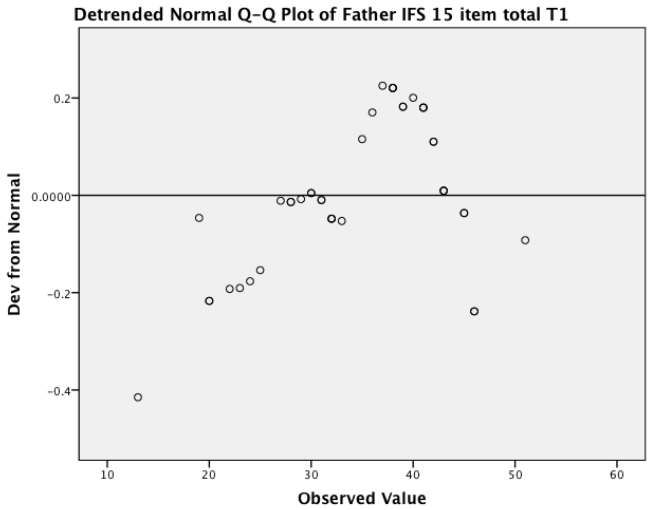


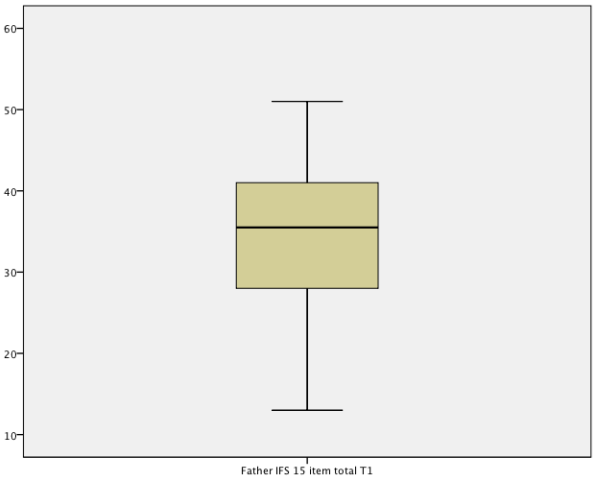


Father IFS 15 item total Time 1

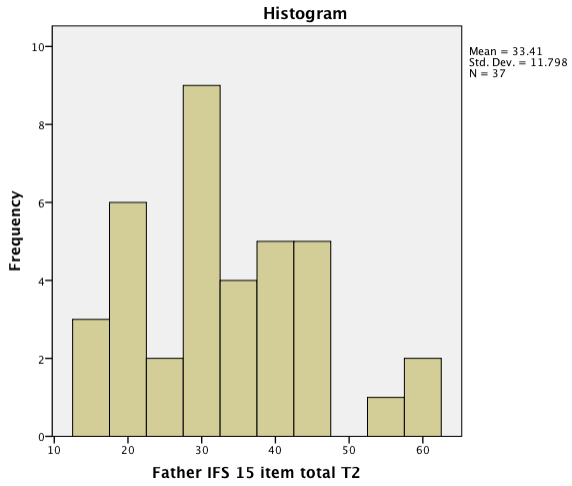


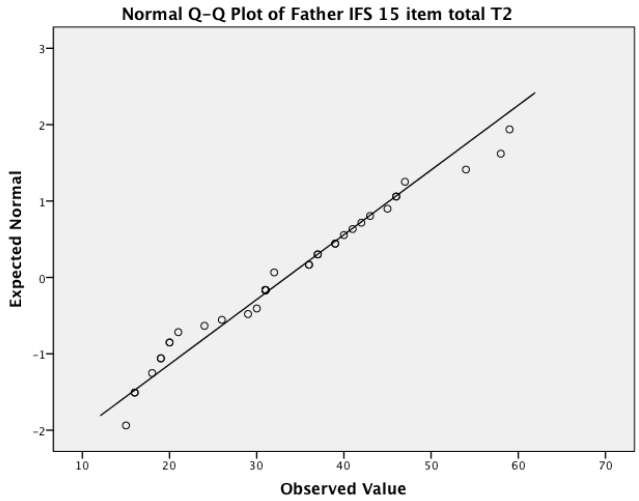


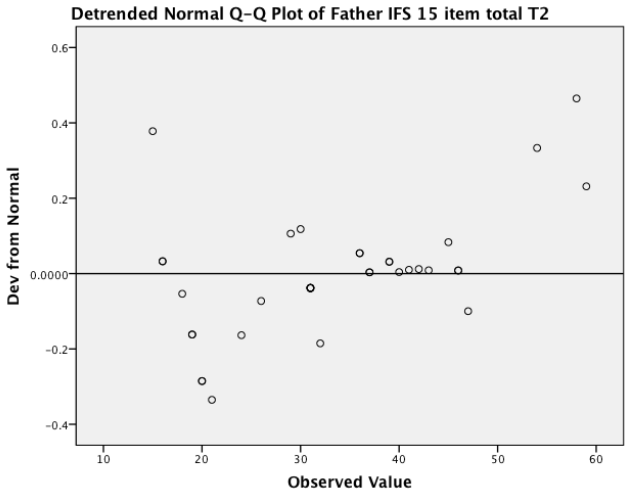


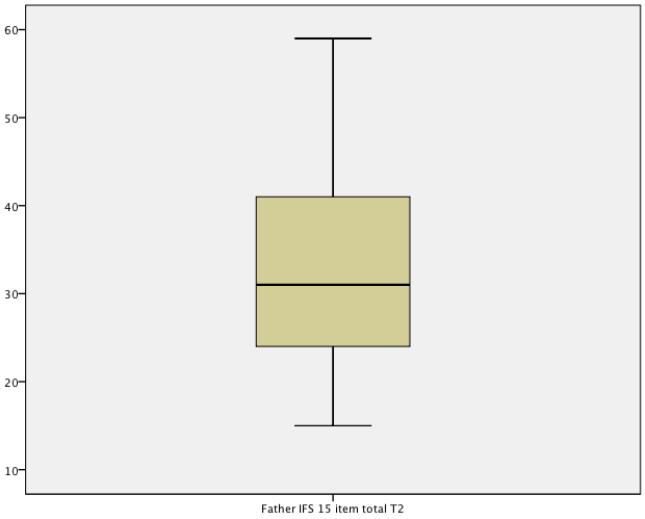


Father IFS 15 item total Time 2









IFS 19-item Total Scores Distribution and Normality

Case Processing Summary

	Cases					
	Valid		Missing		Total	
	N	Percent	N	Percent	N	Percent
Mother IFS 19 items total Time 1	42	100.0%	0	0.0%	42	100.0%
Mother IFS 19 items total Time 2	36	85.7%	6	14.3%	42	100.0%
Father IFS 19 items total Time 1	42	100.0%	0	0.0%	42	100.0%
Father IFS 19 items total Time 2	37	88.1%	5	11.9%	42	100.0%

Descriptives

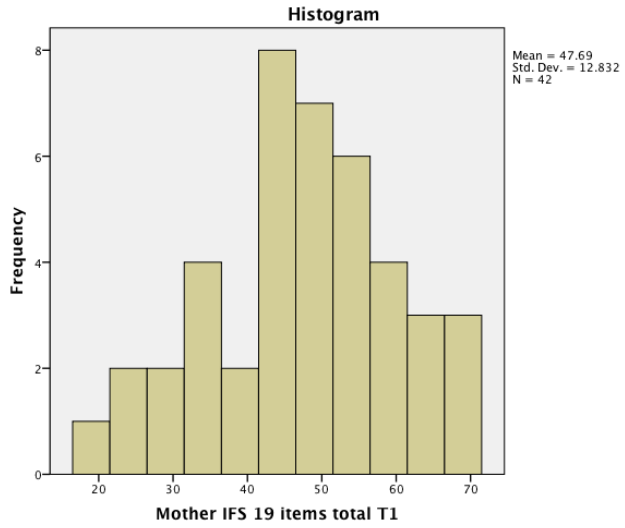
			Statistic	Std. Error
Mother IFS 19 items total Time 1	Mean		47.69	1.980
	95% Confidence Interval for Mean	Lower Bound	43.69	
		Upper Bound	51.69	
	5% Trimmed Mean		47.93	
	Median		49.00	
	Variance		164.658	
	Std. Deviation		12.832	
	Minimum		19	
	Maximum		71	
	Range		52	
	Interquartile Range		17	
	Skewness		-.276	.365
	Kurtosis		-.286	.717
Mother IFS 19 items total Time 2	Mean		45.61	2.498
	95% Confidence Interval for Mean	Lower Bound	40.54	
		Upper Bound	50.68	
	5% Trimmed Mean		44.89	
	Median		46.00	
	Variance		224.702	
	Std. Deviation		14.990	
	Minimum		21	
	Maximum		89	
	Range		68	
	Interquartile Range		22	
	Skewness		.569	.393
	Kurtosis		.658	.768
Father IFS 19 items total Time 1	Mean		44.07	1.717
	95% Confidence Interval for Mean	Lower Bound	40.60	
		Upper Bound	47.54	
	5% Trimmed Mean		44.22	
	Median		45.00	
	Variance		123.824	
	Std. Deviation		11.128	
	Minimum		22	
	Maximum		64	
	Range		42	
	Interquartile Range		18	
	Skewness		-.237	.365
	Kurtosis		-.859	.717
Father IFS 19 items total Time 2	Mean		42.68	2.350
	95% Confidence Interval for Mean	Lower Bound	37.91	
		Upper Bound	47.44	
	5% Trimmed Mean		42.25	
	Median		41.00	
	Variance		204.281	
	Std. Deviation		14.293	
	Minimum		19	
	Maximum		74	
	Range		55	
	Interquartile Range		25	
	Skewness		.203	.388
	Kurtosis		-.590	.759

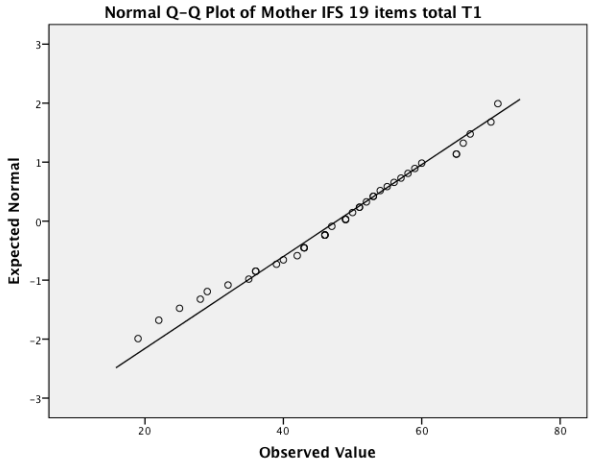
Tests of Normality

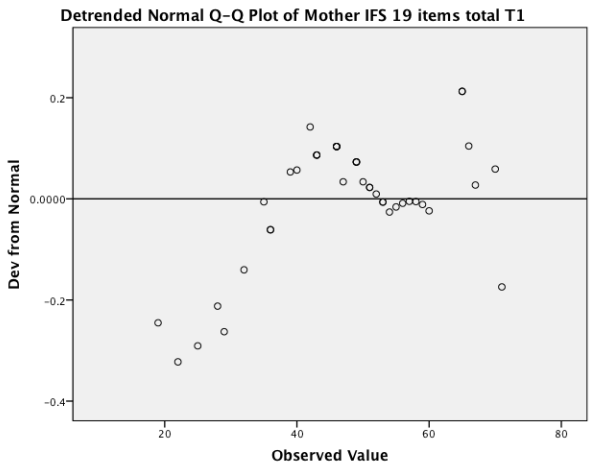
	Kolmogorov-Smirnov[a]		
	Statistic	df	Sig.
Mother IFS 19 items total Time 1	.090	42	.200*
Mother IFS 19 items total Time 2	.075	36	.200*
Father IFS 19 items total Time 1	.100	42	.200*
Father IFS 19 items total Time 2	.122	37	.184

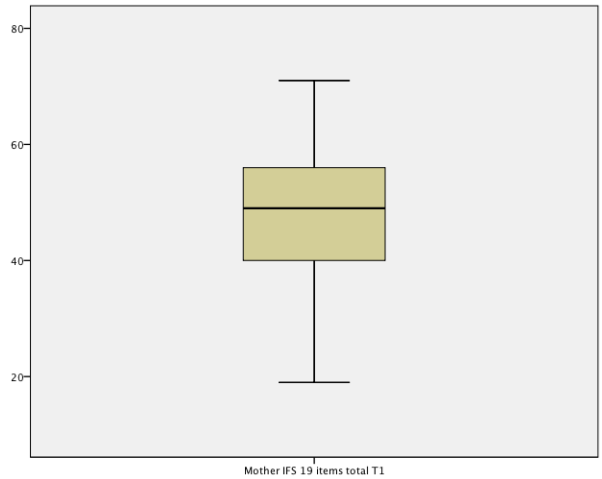
*. This is a lower bound of the true significance.
a. Lilliefors Significance Correction

Mother IFS 19 items total Time 1

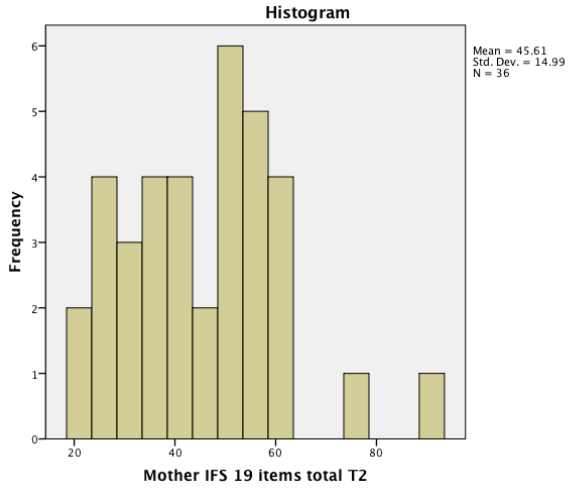


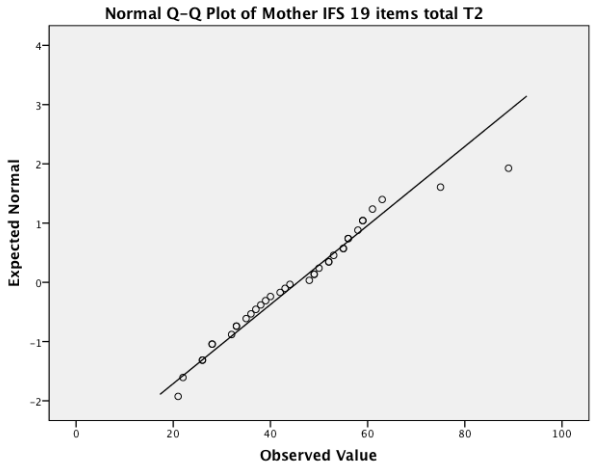


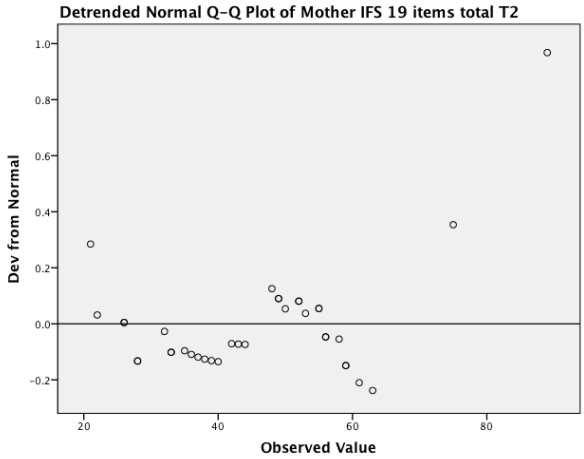


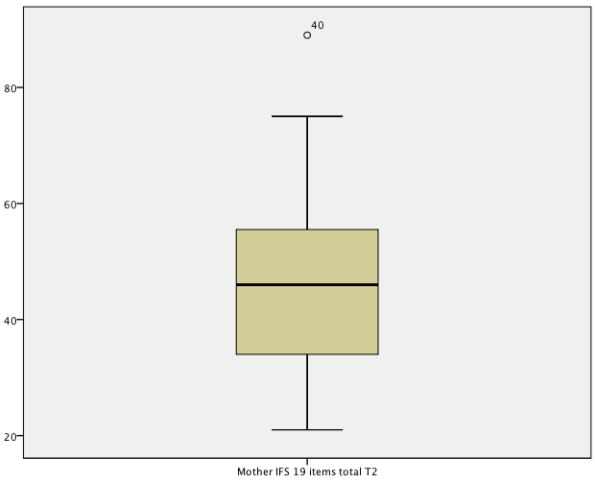


Mother IFS 19 items total Time 2

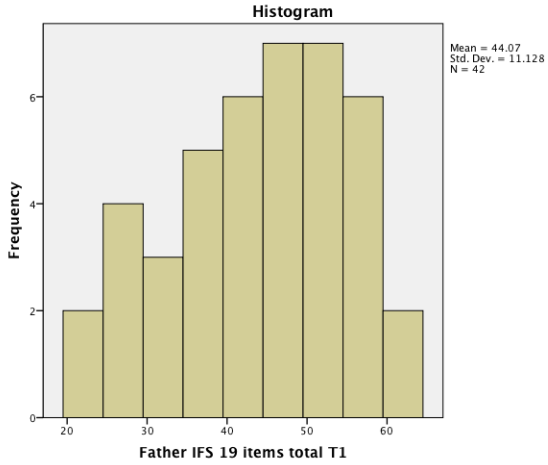


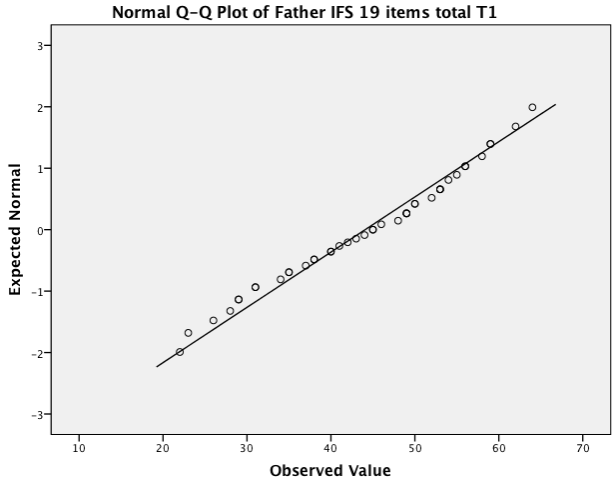


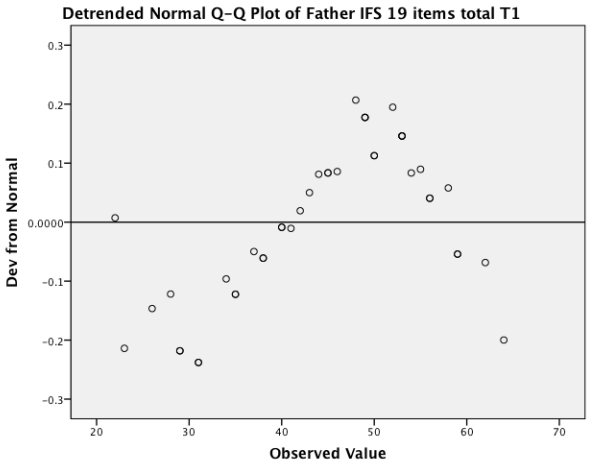


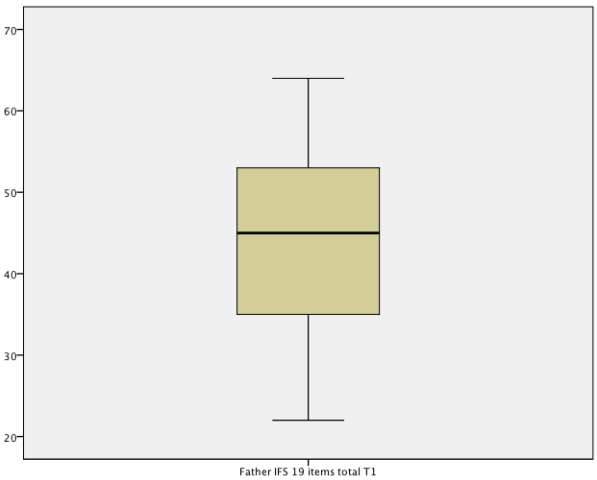


Father IFS 19 items total Time 1

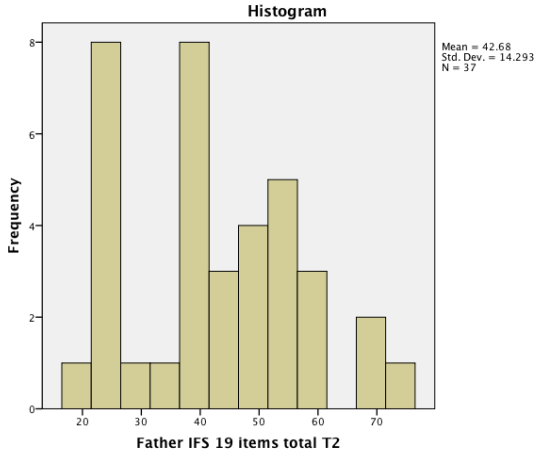


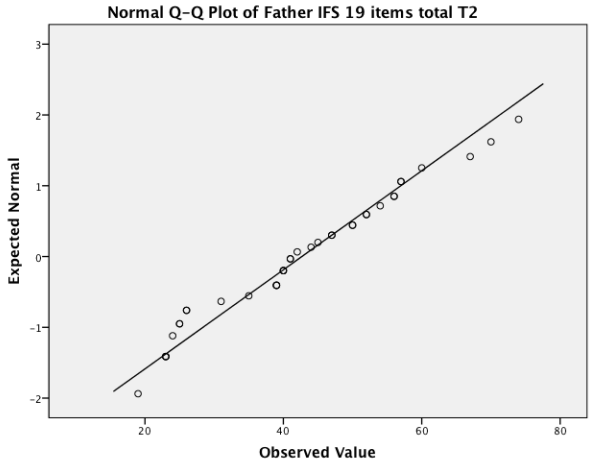


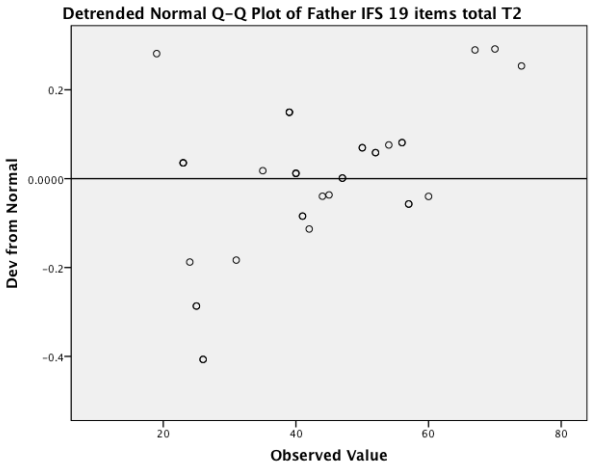


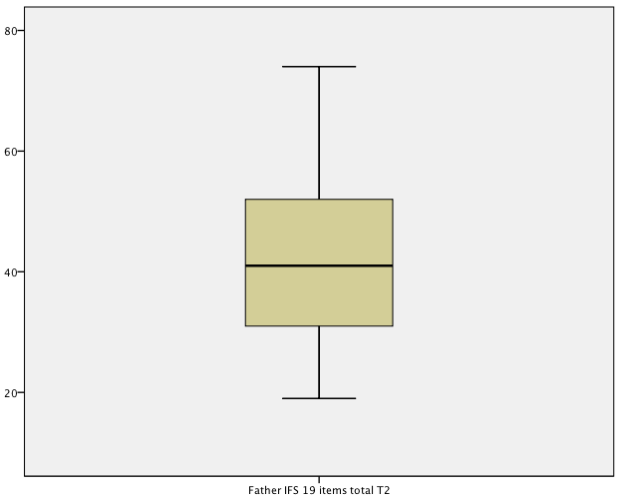


Father IFS 19 items total Time 2









DADS Scores Distribution and Normality

Case Processing Summary

	Cases					
	Valid		Missing		Total	
	N	Percent	N	Percent	N	Percent
Mother DADS Amount Time 1	42	100.0%	0	0.0%	42	100.0%
Mother DADS Helpfulness Time 1	42	100.0%	0	0.0%	42	100.0%
Mother DADS Amount Time 2	37	88.1%	5	11.9%	42	100.0%
Mother DADS Helpfulness Time 2	37	88.1%	5	11.9%	42	100.0%

Case Processing Summary

	Cases					
	Valid		Missing		Total	
	N	Percent	N	Percent	N	Percent
Father DADS Amount Time 1	42	100.0%	0	0.0%	42	100.0%
Father DADS Helpfulness Time 1	42	100.0%	0	0.0%	42	100.0%
Father DADS Amount Time 2	37	88.1%	5	11.9%	42	100.0%
Father DADS Helpfulness Time 2	37	88.1%	5	11.9%	42	100.0%

MOTHERS

Descriptives

			Statistic	Std. Error
Mother DADS Amount Time 1	Mean		73.7607	2.99840
	95% Confidence Interval for Mean	Lower Bound	67.7053	
		Upper Bound	79.8161	
	5% Trimmed Mean		73.6190	
	Median		72.5450	
	Variance		377.597	
	Std. Deviation		19.43186	
	Minimum		32.00	
	Maximum		120.00	
	Range		88.00	
	Interquartile Range		29.57	
	Skewness		.133	.365
	Kurtosis		-.435	.717
Mother DADS Helpfulness Time 1	Mean		76.1190	2.41131
	95% Confidence Interval for Mean	Lower Bound	71.2493	
		Upper Bound	80.9888	
	5% Trimmed Mean		76.5926	
	Median		75.5000	
	Variance		244.205	
	Std. Deviation		15.62706	
	Minimum		28.00	
	Maximum		106.00	
	Range		78.00	
	Interquartile Range		19.50	
	Skewness		-.614	.365
	Kurtosis		.949	.717
Mother DADS Amount Time 2	Mean		68.7400	3.83017
	95% Confidence Interval for Mean	Lower Bound	60.9721	
		Upper Bound	76.5079	
	5% Trimmed Mean		67.9559	
	Median		66.2900	
	Variance		542.798	
	Std. Deviation		23.29802	
	Minimum		30.00	
	Maximum		120.00	
	Range		90.00	
	Interquartile Range		32.12	
	Skewness		.495	.388
	Kurtosis		-.356	.759
Mother DADS Helpfulness Time 2	Mean		72.2703	2.81903
	95% Confidence Interval for Mean	Lower Bound	66.5530	
		Upper Bound	77.9875	
	5% Trimmed Mean		71.7568	
	Median		71.0000	
	Variance		294.036	
	Std. Deviation		17.14748	
	Minimum		43.00	
	Maximum		114.00	
	Range		71.00	
	Interquartile Range		23.00	
	Skewness		.392	.388
	Kurtosis		-.239	.759

FATHERS

Descriptives

			Statistic	Std. Error
Father DADS Amount Time 1	Mean		75.2821	2.59963
	95% Confidence Interval for Mean	Lower Bound	70.0321	
		Upper Bound	80.5322	
	5% Trimmed Mean		75.1728	
	Median		77.2850	
	Variance		283.839	
	Std. Deviation		16.84752	
	Minimum		42.00	
	Maximum		118.00	
	Range		76.00	
	Interquartile Range		23.66	
	Skewness		.006	.365
	Kurtosis		-.310	.717
Father DADS Helpfulness Time 1	Mean		72.1667	2.48828
	95% Confidence Interval for Mean	Lower Bound	67.1415	
		Upper Bound	77.1919	
	5% Trimmed Mean		71.4286	
	Median		69.5000	
	Variance		260.045	
	Std. Deviation		16.12590	
	Minimum		47.00	
	Maximum		120.00	
	Range		73.00	
	Interquartile Range		24.50	
	Skewness		.666	.365
	Kurtosis		.413	.717
Father DADS Amount Time 2	Mean		62.8859	2.43042
	95% Confidence Interval for Mean	Lower Bound	57.9568	
		Upper Bound	67.8151	
	5% Trimmed Mean		62.2485	
	Median		62.4000	
	Variance		218.557	
	Std. Deviation		14.78368	
	Minimum		36.00	
	Maximum		102.55	
	Range		66.55	
	Interquartile Range		22.81	
	Skewness		.529	.388
	Kurtosis		.196	.759
Father DADS Helpfulness Time 2	Mean		66.2038	3.06826
	95% Confidence Interval for Mean	Lower Bound	59.9811	
		Upper Bound	72.4265	
	5% Trimmed Mean		65.6123	
	Median		61.0000	
	Variance		348.327	
	Std. Deviation		18.66351	
	Minimum		29.00	
	Maximum		109.00	
	Range		80.00	
	Interquartile Range		31.00	
	Skewness		.655	.388
	Kurtosis		-.102	.759

Tests of Normality

	Kolmogorov-Smirnov[a]		
	Statistic	df	Sig.
Mother DADS Amount Time 1	.089	42	.200*
Mother DADS Helpfulness Time 1	.113	42	.200*
Mother DADS Amount Time 2	.092	37	.200*
Mother DADS Helpfulness Time 2	.099	37	.200*

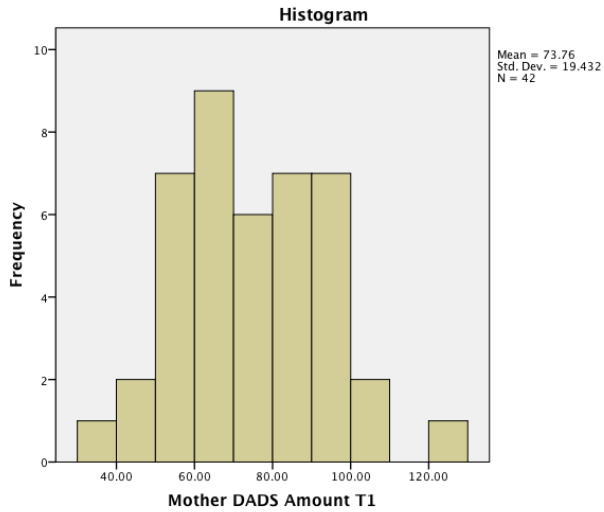
*. This is a lower bound of the true significance.
a. Lilliefors Significance Correction

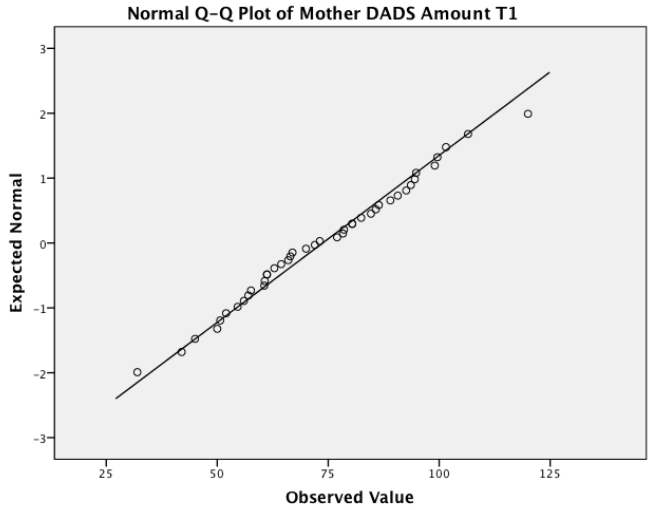
Tests of Normality

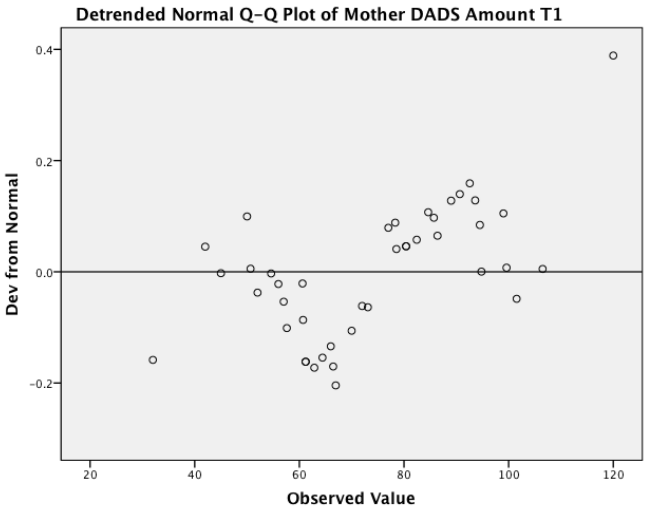
	Kolmogorov-Smirnov[a]		
	Statistic	df	Sig.
Father DADS Amount Time 1	.103	42	.200*
Father DADS Helpfulness Time 1	.102	42	.200*
Father DADS Amount Time 2	.094	37	.200*
Father DADS Helpfulness Time 2	.186	37	.002

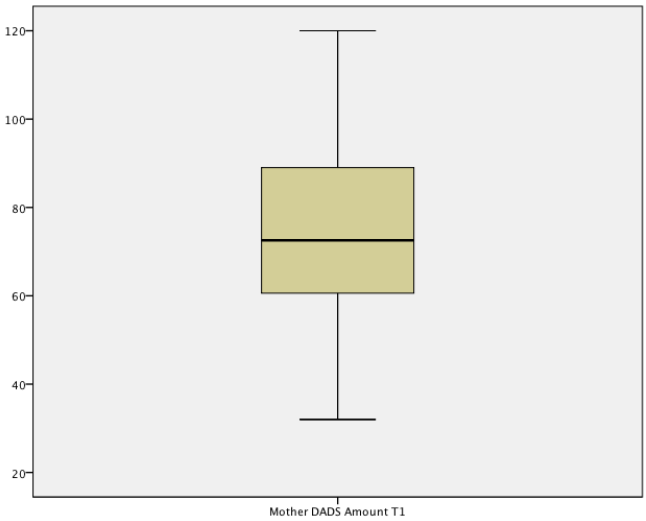
*. This is a lower bound of the true significance.
a. Lilliefors Significance Correction

Mother DADS Amount Time 1

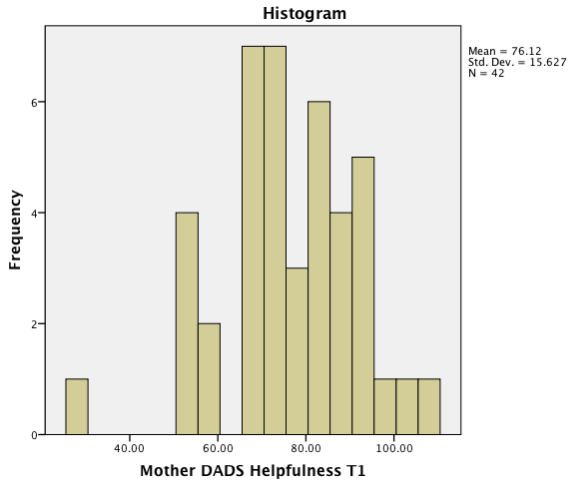


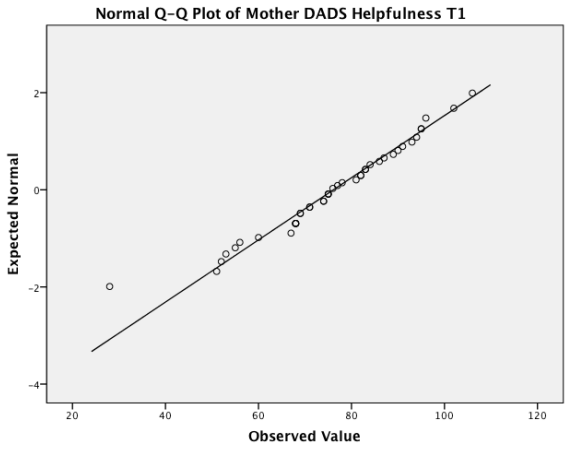


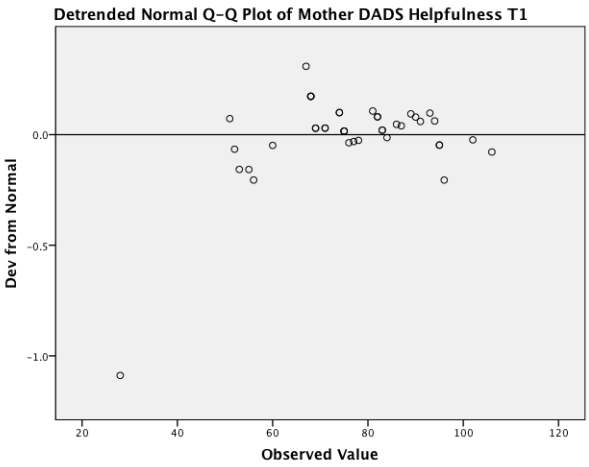


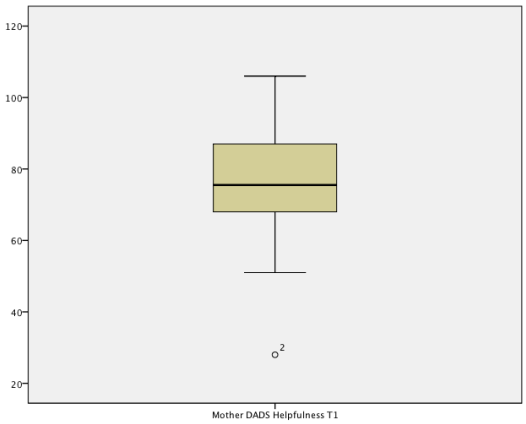


Mother DADS Helpfulness Time 1

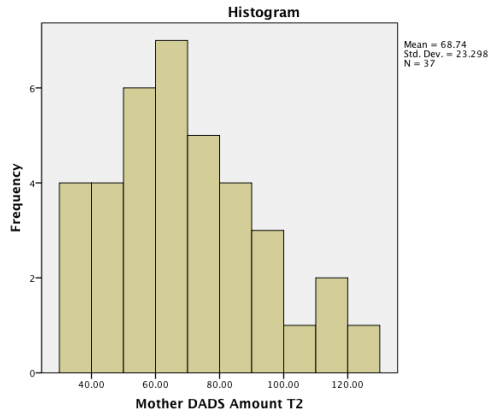


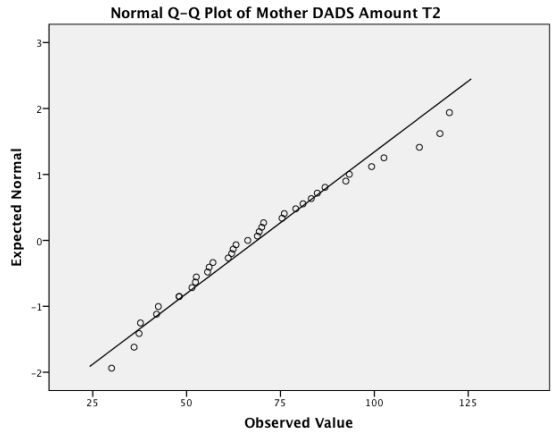


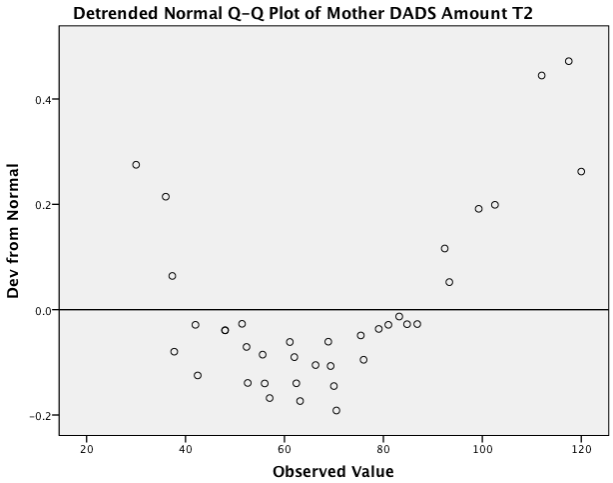


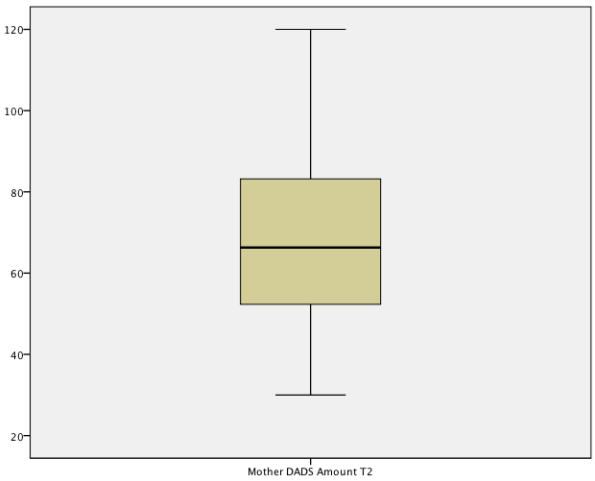


Mother DADS Amount Time 2









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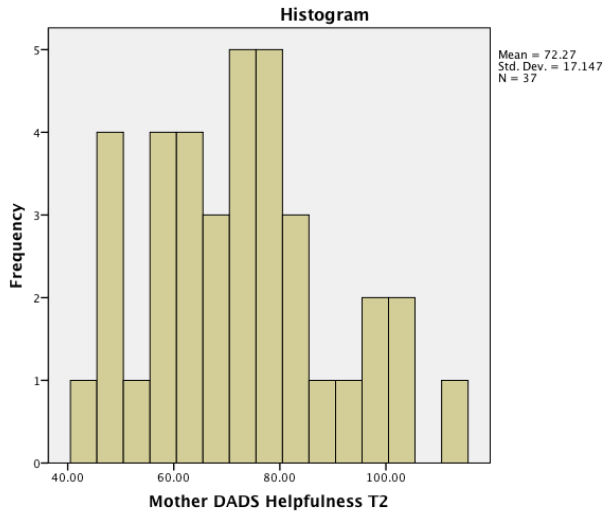
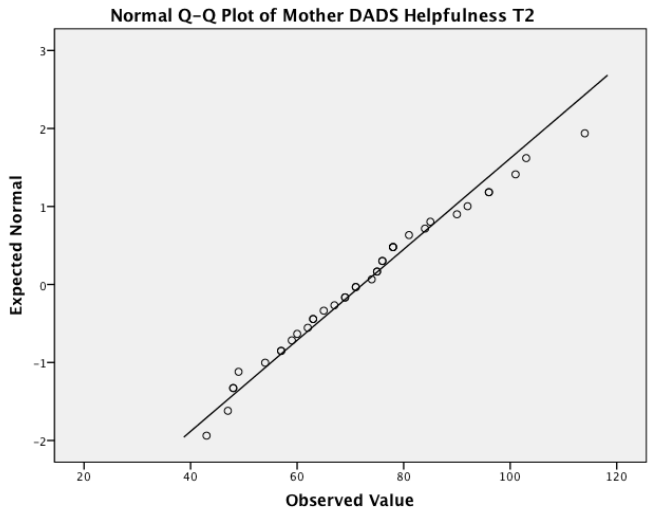
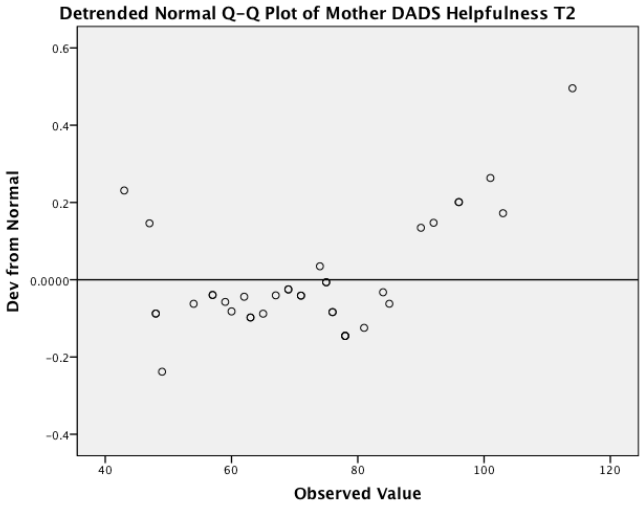
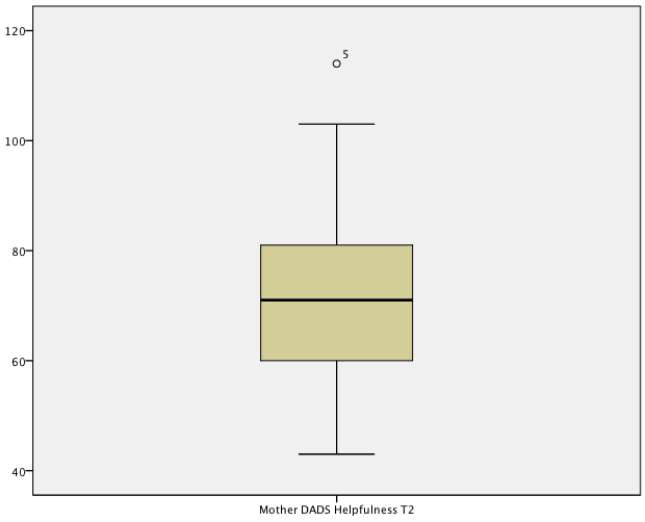
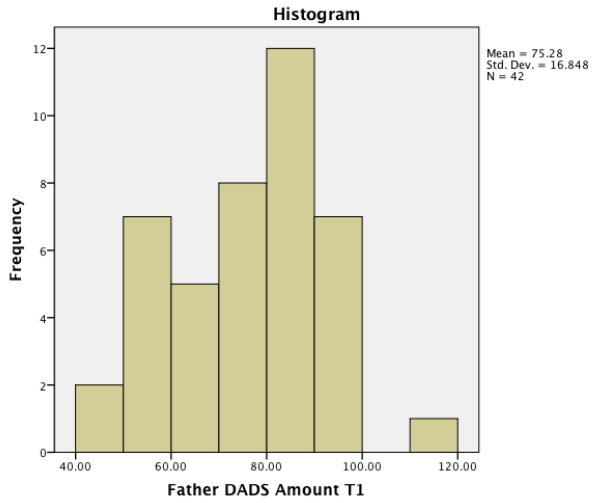


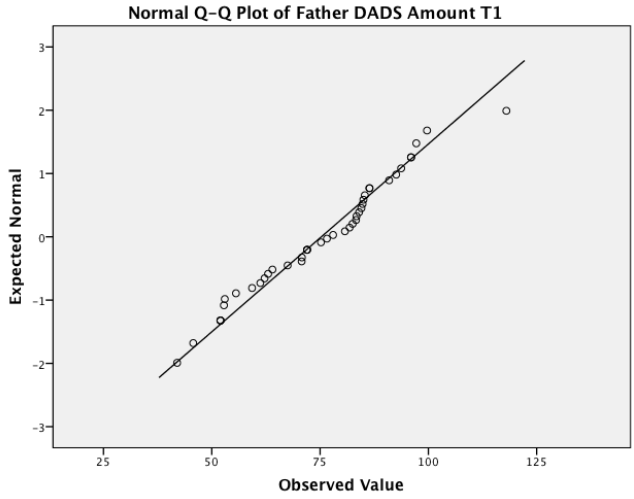


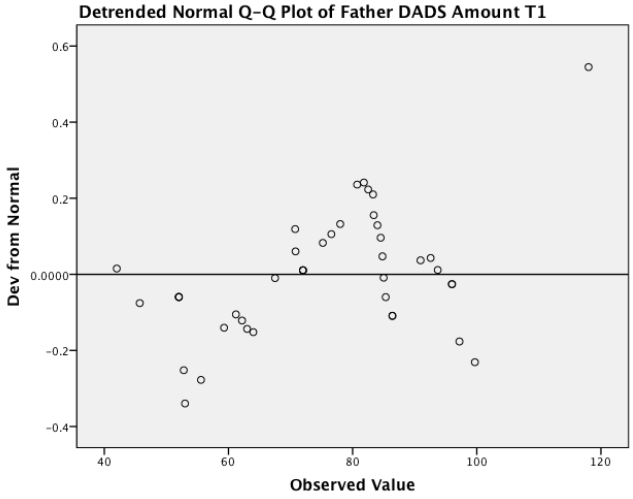


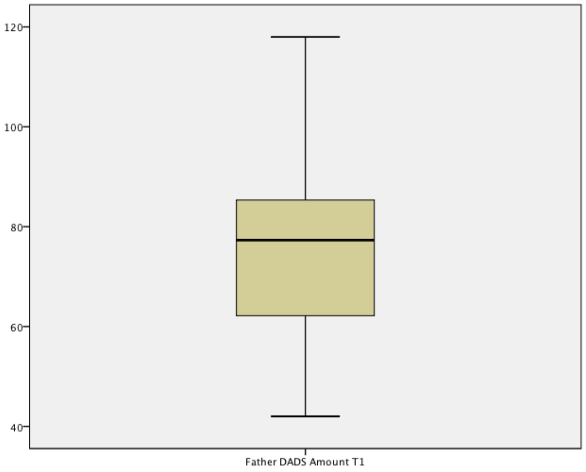


Father DADS Amount Time 1









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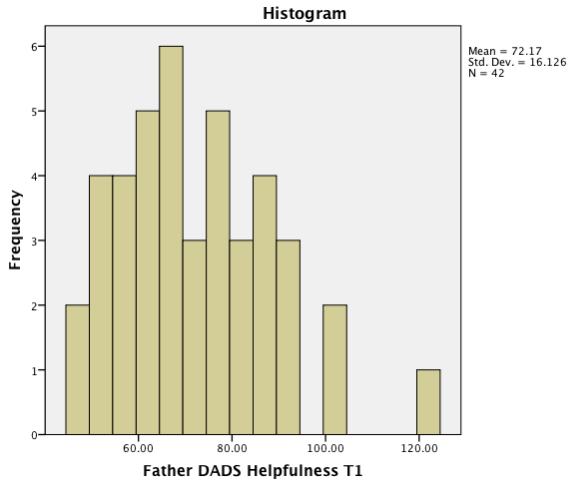
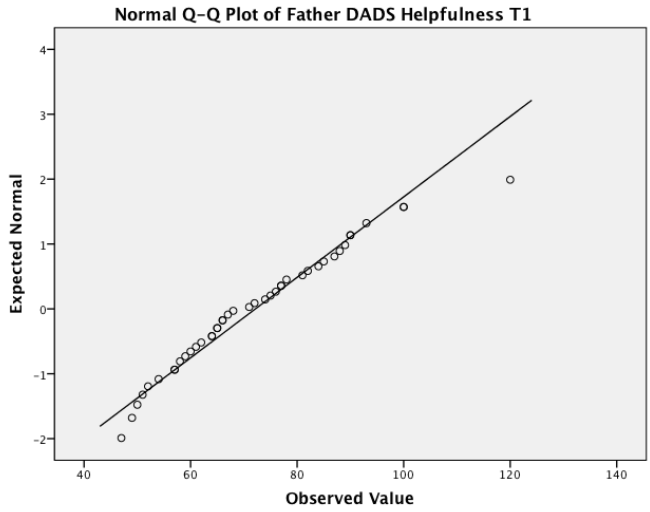
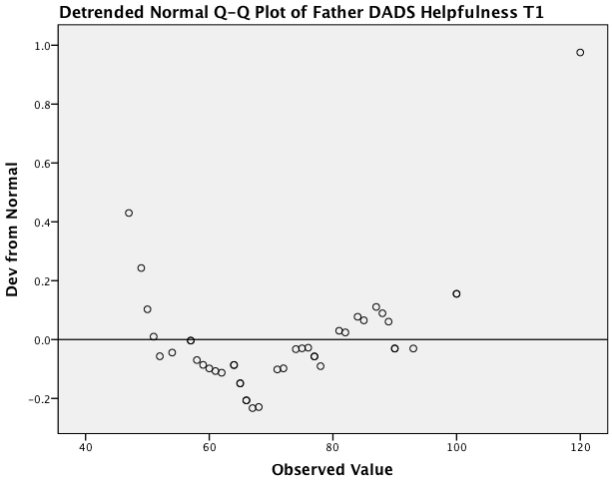
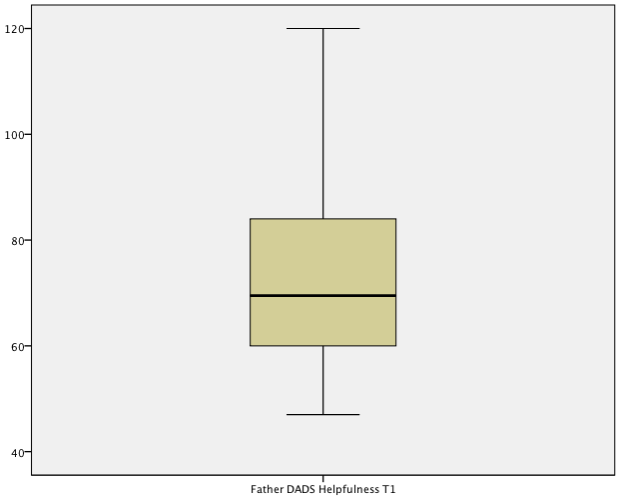
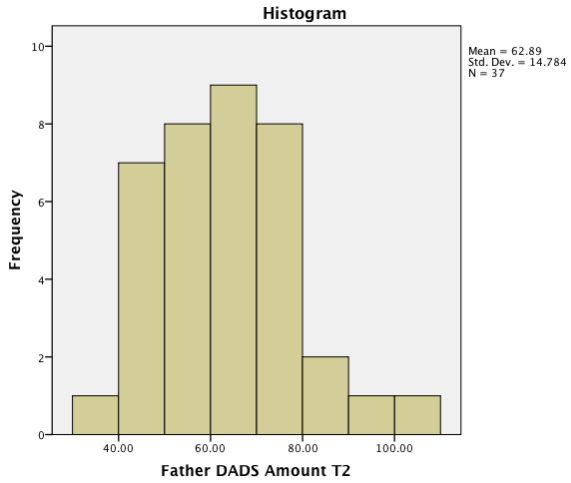


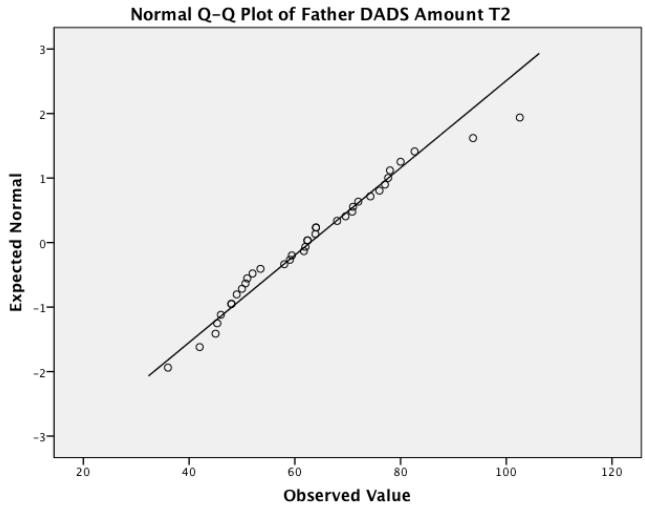


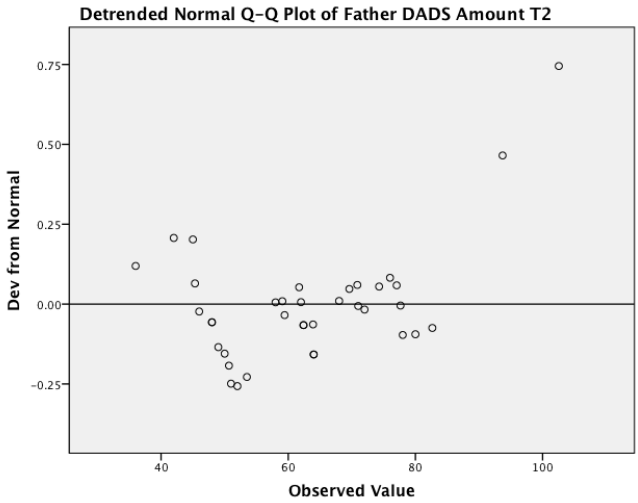


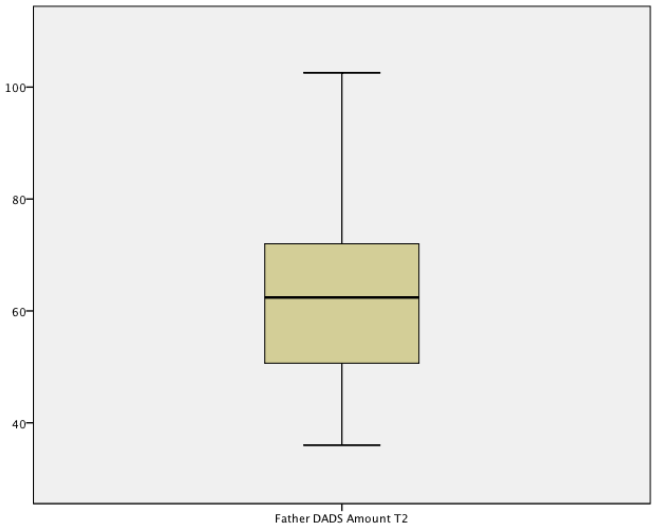


Father DADS Amount Time 2

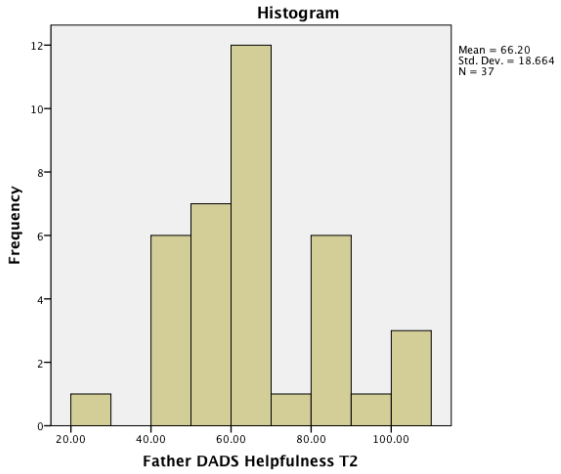


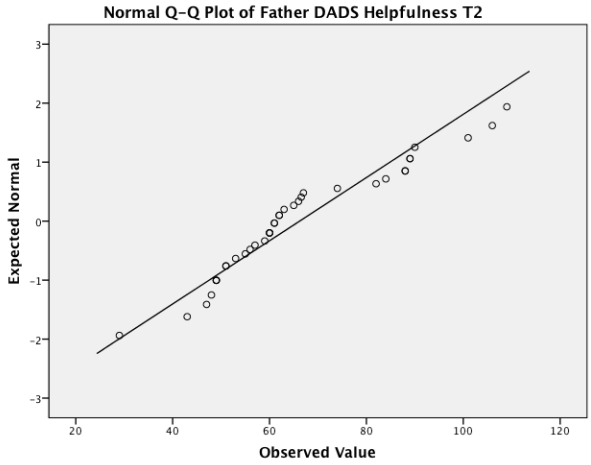


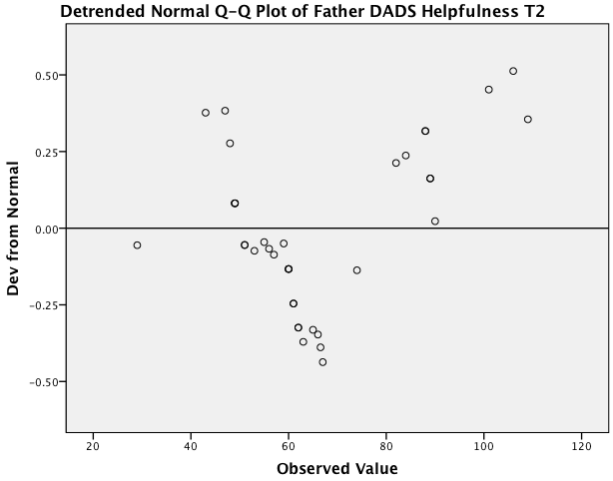


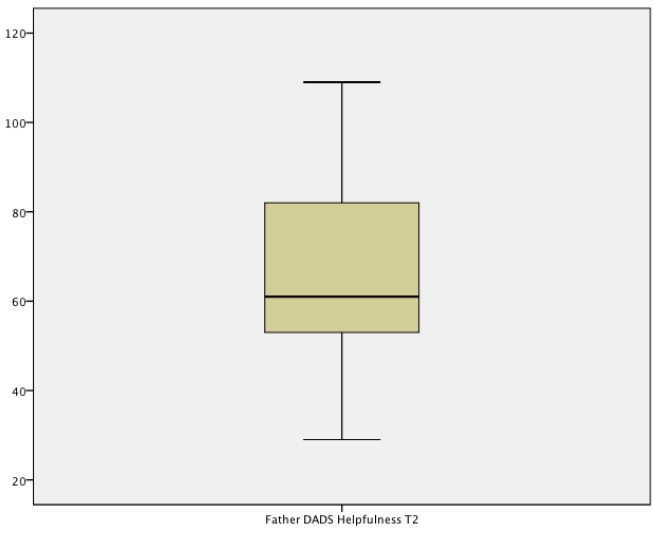


Father DADS Helpfulness Time 2









Appendix H
HYPOTHESIS 1

DASS SCORES

Case Processing Summary

	Cases					
	Valid		Missing		Total	
	N	Percent	N	Percent	N	Percent
Mother DASS Total Time 1	42	100.0%	0	0.0%	42	100.0%
Mother DASS Total Time 2	37	88.1%	5	11.9%	42	100.0%
Father DASS Total Time 1	42	100.0%	0	0.0%	42	100.0%
Father DASS Total Time 2	37	88.1%	5	11.9%	42	100.0%

Mothers' DASS Total raw scores

			Statistic	Std. Error
Mother DASS Total Time 1	Mean		23.14	3.373
	95% Confidence Interval for Mean	Lower Bound	16.33	
		Upper Bound	29.95	
	5% Trimmed Mean		20.97	
	Median		16.00	
	Variance		477.735	
	Std. Deviation		21.857	
	Minimum		0	
	Maximum		90	
	Range		90	
	Interquartile Range		21	
	Skewness		1.565	.365
	Kurtosis		2.085	.717
Mother DASS Total Time 2	Mean		22.46	2.822
	95% Confidence Interval for Mean	Lower Bound	16.74	
		Upper Bound	28.18	
	5% Trimmed Mean		21.58	
	Median		17.00	
	Variance		294.644	
	Std. Deviation		17.165	
	Minimum		0	
	Maximum		63	
	Range		63	
	Interquartile Range		26	
	Skewness		.794	.388
	Kurtosis		-.250	.759

Fathers' DASS Total raw scores

			Statistic	Std. Error
Father DASS Total Time 1	Mean		16.79	3.348
	95% Confidence Interval for Mean	Lower Bound	10.02	
		Upper Bound	23.55	
	5% Trimmed Mean		13.59	
	Median		12.00	
	Variance		470.904	
	Std. Deviation		21.700	
	Minimum		0	
	Maximum		126	
	Range		126	
	Interquartile Range		16	
	Skewness		3.456	.365
	Kurtosis		15.565	.717
Father DASS Total Time 2	Mean		13.24	1.966
	95% Confidence Interval for Mean	Lower Bound	9.26	
		Upper Bound	17.23	
	5% Trimmed Mean		12.39	
	Median		10.00	
	Variance		143.078	
	Std. Deviation		11.962	
	Minimum		0	
	Maximum		46	
	Range		46	
	Interquartile Range		18	
	Skewness		.904	.388
	Kurtosis		.153	.759

Mothers' DASS Total percentile scores

			Statistic	Std. Error
Mother DASS Total Percentile Time 1	Mean		57.29	4.391
	95% Confidence Interval for Mean	Lower Bound	48.42	
		Upper Bound	66.15	
	5% Trimmed Mean		57.90	
	Median		60.00	
	Variance		809.672	
	Std. Deviation		28.455	
	Minimum		5	
	Maximum		99	
	Range		94	
	Interquartile Range		43	
	Skewness		-.254	.365
	Kurtosis		-.949	.717
Mother DASS Total Percentile Time 2	Mean		59.76	4.642
	95% Confidence Interval for Mean	Lower Bound	50.34	
		Upper Bound	69.17	
	5% Trimmed Mean		60.81	
	Median		60.00	
	Variance		797.300	
	Std. Deviation		28.237	
	Minimum		5	
	Maximum		96	
	Range		91	
	Interquartile Range		49	
	Skewness		-.441	.388
	Kurtosis		-.997	.759

Fathers' DASS Total percentile scores

			Statistic	Std. Error
Father DASS Total Percentile Time 1	Mean		45.93	4.497
	95% Confidence Interval for Mean	Lower Bound	36.85	
		Upper Bound	55.01	
	5% Trimmed Mean		45.38	
	Median		47.50	
	Variance		849.385	
	Std. Deviation		29.144	
	Minimum		5	
	Maximum		99	
	Range		94	
	Interquartile Range		48	
	Skewness		.123	.365
	Kurtosis		-1.124	.717
Father DASS Total Percentile Time 2	Mean		42.76	4.828
	95% Confidence Interval for Mean	Lower Bound	32.97	
		Upper Bound	52.55	
	5% Trimmed Mean		42.22	
	Median		40.00	
	Variance		862.300	
	Std. Deviation		29.365	
	Minimum		5	
	Maximum		92	
	Range		87	
	Interquartile Range		50	
	Skewness		.197	.388
	Kurtosis		-1.585	.759

One-sample Wilcoxon Signed Rank Test:
DASS percentile scores compared with general population median score.

Time 1 *N* = 42, Time 2 *N* = 37. Population median score = 50.

Hypothesis Test Summary

	Null Hypothesis	Test	Sig.	Decision
1	The median of Mother DASS Total Percentile T1 equals 50.000.	One-Sample Wilcoxon Signed Rank Test	.095	Retain the null hypothesis.
2	The median of Mother DASS Total Percentile T2 equals 50.000.	One-Sample Wilcoxon Signed Rank Test	.048	Reject the null hypothesis.
3	The median of Father DASS Total Percentile T1 equals 50.000.	One-Sample Wilcoxon Signed Rank Test	.409	Retain the null hypothesis.
4	The median of Father DASS Total Percentile T2 equals 50.000.	One-Sample Wilcoxon Signed Rank Test	.074	Retain the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

Case Processing Summary

	Cases					
	Valid		Missing		Total	
	N	Percent	N	Percent	N	Percent
Mother FAD General Functioning Time 1	42	100.0%	0	0.0%	42	100.0%
Mother FAD General Functioning Time 2	37	88.1%	5	11.9%	42	100.0%
Father FAD General Functioning Time 1	42	100.0%	0	0.0%	42	100.0%
Father FAD General Functioning Time 2	37	88.1%	5	11.9%	42	100.0%

Mother FAD

			Statistic	Std. Error
Mother FAD General Functioning Time 1	Mean		1.5934	.05763
	95% Confidence Interval for Mean	Lower Bound	1.4770	
		Upper Bound	1.7098	
	5% Trimmed Mean		1.5837	
	Median		1.5000	
	Variance		.139	
	Std. Deviation		.37348	
	Minimum		1.00	
	Maximum		2.50	
	Range		1.50	
	Interquartile Range		.60	
	Skewness		.221	.365
	Kurtosis		-.703	.717
Mother FAD General Functioning Time 2	Mean		1.6078	.07207
	95% Confidence Interval for Mean	Lower Bound	1.4617	
		Upper Bound	1.7540	
	5% Trimmed Mean		1.5941	
	Median		1.5800	
	Variance		.192	
	Std. Deviation		.43841	
	Minimum		1.00	
	Maximum		2.50	
	Range		1.50	
	Interquartile Range		.67	
	Skewness		.471	.388
	Kurtosis		-.862	.759

Father FAD

			Statistic	Std. Error
Father FAD General Functioning Time 1	Mean		1.6117	.06336
	95% Confidence Interval for Mean	Lower Bound	1.4837	
		Upper Bound	1.7396	
	5% Trimmed Mean		1.5922	
	Median		1.5400	
	Variance		.169	
	Std. Deviation		.41063	
	Minimum		1.00	
	Maximum		2.75	
	Range		1.75	
	Interquartile Range		.59	
	Skewness		.509	.365
	Kurtosis		.025	.717
Father FAD General Functioning Time 2	Mean		1.6541	.06399
	95% Confidence Interval for Mean	Lower Bound	1.5243	
		Upper Bound	1.7838	
	5% Trimmed Mean		1.6443	
	Median		1.7500	
	Variance		.152	
	Std. Deviation		.38926	
	Minimum		1.00	
	Maximum		2.75	
	Range		1.75	
	Interquartile Range		.59	
	Skewness		.211	.388
	Kurtosis		.195	.759

One Sample T-Tests:
Comparisons of FAD scores against published cut-off score

One-Sample Statistics

	N	Mean	Std. Deviation	Std. Error Mean
Mother FAD General Functioning Time 1	42	1.5934	.37348	.05763
Mother FAD General Functioning Time 2	37	1.6078	.43841	.07207
Father FAD General Functioning Time 1	42	1.6117	.41063	.06336
Father FAD General Functioning Time 2	37	1.6541	.38926	.06399

One-Sample Test: Healthy cut-off score

	Test Value = 2.00					
	t	df	Sig. (2-tailed)	Mean Difference	95% Confidence Interval of the Difference	
					Lower	Upper
Mother FAD General Functioning Time 1	-7.056	41	.000	-.40662	-.5230	-.2902
Mother FAD General Functioning Time 2	-5.441	36	.000	-.39216	-.5383	-.2460
Father FAD General Functioning Time 1	-6.129	41	.000	-.38833	-.5163	-.2604
Father FAD General Functioning Time 2	-5.406	36	.000	-.34595	-.4757	-.2162

One-Sample Test: Medical mean

	Test Value = 1.89					
	t	df	Sig. (2-tailed)	Mean Difference	95% Confidence Interval of the Difference	
					Lower	Upper
Mother FAD General Functioning Time 1	-5.147	41	.000	-.29662	-.4130	-.1802
Father FAD General Functioning Time 1	-4.393	41	.000	-.27833	-.4063	-.1504
Mother FAD General Functioning Time 2	-3.915	36	.000	-.28216	-.4283	-.1360
Father FAD General Functioning Time 2	-3.687	36	.001	-.23595	-.3657	-.1062

IFS Scores

Case Processing Summary

	Cases					
	Valid		Missing		Total	
	N	Percent	N	Percent	N	Percent
Mother IFS 15 item total Time 1	42	100.0%	0	0.0%	42	100.0%
Mother IFS 15 item total Time 2	36	85.7%	6	14.3%	42	100.0%
Father IFS 15 item total Time 1	42	100.0%	0	0.0%	42	100.0%
Father IFS 15 item total Time 2	37	88.1%	5	11.9%	42	100.0%

Mothers' IFS 15-item total scores

			Statistic	Std. Error
Mother IFS 15 item total Time 1	Mean		37.50	1.676
	95% Confidence Interval for Mean	Lower Bound	34.11	
		Upper Bound	40.89	
	5% Trimmed Mean		37.85	
	Median		38.00	
	Variance		118.012	
	Std. Deviation		10.863	
	Minimum		9	
	Maximum		56	
	Range		47	
	Interquartile Range		13	
	Skewness		-.475	.365
	Kurtosis		.054	.717
Mother IFS 15 item total Time 2	Mean		35.92	2.068
	95% Confidence Interval for Mean	Lower Bound	31.72	
		Upper Bound	40.12	
	5% Trimmed Mean		35.23	
	Median		35.00	
	Variance		153.964	
	Std. Deviation		12.408	
	Minimum		15	
	Maximum		75	
	Range		60	
	Interquartile Range		16	
	Skewness		.831	.393
	Kurtosis		1.398	.768

Fathers' IFS 15-item total scores

			Statistic	Std. Error
Father IFS 15 item total Time 1	Mean		34.24	1.362
	95% Confidence Interval for Mean	Lower Bound	31.49	
		Upper Bound	36.99	
	5% Trimmed Mean		34.46	
	Median		35.50	
	Variance		77.942	
	Std. Deviation		8.828	
	Minimum		13	
	Maximum		51	
	Range		38	
	Interquartile Range		13	
	Skewness		-.385	.365
	Kurtosis		-.529	.717
Father IFS 15 item total Time 2	Mean		33.41	1.940
	95% Confidence Interval for Mean	Lower Bound	29.47	
		Upper Bound	37.34	
	5% Trimmed Mean		33.01	
	Median		31.00	
	Variance		139.192	
	Std. Deviation		11.798	
	Minimum		15	
	Maximum		59	
	Range		44	
	Interquartile Range		19	
	Skewness		.296	.388
	Kurtosis		-.490	.759

Mothers' IFS 19-item total scores

			Statistic	Std. Error
Mother IFS 19 items total Time 1	Mean		47.69	1.980
	95% Confidence Interval for Mean	Lower Bound	43.69	
		Upper Bound	51.69	
	5% Trimmed Mean		47.93	
	Median		49.00	
	Variance		164.658	
	Std. Deviation		12.832	
	Minimum		19	
	Maximum		71	
	Range		52	
	Interquartile Range		17	
	Skewness		-.276	.365
	Kurtosis		-.286	.717
Mother IFS 19 items total Time 2	Mean		45.61	2.498
	95% Confidence Interval for Mean	Lower Bound	40.54	
		Upper Bound	50.68	
	5% Trimmed Mean		44.89	
	Median		46.00	
	Variance		224.702	
	Std. Deviation		14.990	
	Minimum		21	
	Maximum		89	
	Range		68	
	Interquartile Range		22	
	Skewness		.569	.393
	Kurtosis		.658	.768

Fathers' IFS 19-item total scores

			Statistic	Std. Error
Father IFS 19 items total Time 1	Mean		44.07	1.717
	95% Confidence Interval for Mean	Lower Bound	40.60	
		Upper Bound	47.54	
	5% Trimmed Mean		44.22	
	Median		45.00	
	Variance		123.824	
	Std. Deviation		11.128	
	Minimum		22	
	Maximum		64	
	Range		42	
	Interquartile Range		18	
	Skewness		-.237	.365
	Kurtosis		-.859	.717
Father IFS 19 items total Time 2	Mean		42.68	2.350
	95% Confidence Interval for Mean	Lower Bound	37.91	
		Upper Bound	47.44	
	5% Trimmed Mean		42.25	
	Median		41.00	
	Variance		204.281	
	Std. Deviation		14.293	
	Minimum		19	
	Maximum		74	
	Range		55	
	Interquartile Range		25	
	Skewness		.203	.388
	Kurtosis		-.590	.759

One Sample T-Tests:
Comparisons of IFS scores against published mean scores

One-Sample Statistics: 19-item total score

	N	Mean	Std. Deviation	Std. Error Mean
Mother IFS 19 items total Time 1	42	47.6905	12.83191	1.98001
Mother IFS 19 items total Time 2	36	45.6111	14.99005	2.49834
Father IFS 19 items total Time 1	42	44.0714	11.12763	1.71703
Father IFS 19 items total Time 2	37	42.6757	14.29268	2.34970

One-Sample Test 19-item total score published mean, children with chronic illness

	Test Value = 48.03					
	t	df	Sig. (2-tailed)	Mean Difference	95% Confidence Interval of the Difference	
					Lower	Upper
Mother IFS 19 items total Time 1	-.171	41	.865	-.33952	-4.3382	3.6592
Mother IFS 19 items total Time 2	-.968	35	.340	-2.41889	-7.4908	2.6530
Father IFS 19 items total Time 1	-2.305	41	.026	-3.95857	-7.4262	-.4910
Father IFS 19 items total Time 2	-2.279	36	.029	-5.35432	-10.1197	-.5889

APPENDIX I
HYPOTHESIS 2

Mothers' DADS Scores Descriptive Statistics

Descriptives

			Statistic	Std. Error
Mother DADS Amount Time 1	Mean		72.0703	3.24509
	95% Confidence Interval for Mean	Lower Bound	65.4889	
		Upper Bound	78.6516	
	5% Trimmed Mean		71.7230	
	Median		66.9500	
	Variance		389.632	
	Std. Deviation		19.73910	
	Minimum		32.00	
	Maximum		120.00	
	Range		88.00	
	Interquartile Range		28.76	
	Skewness		.307	.388
	Kurtosis		-.290	.759
Mother DADS Helpfulness Time 1	Mean		74.6216	2.56048
	95% Confidence Interval for Mean	Lower Bound	69.4287	
		Upper Bound	79.8145	
	5% Trimmed Mean		75.1366	
	Median		75.0000	
	Variance		242.575	
	Std. Deviation		15.57482	
	Minimum		28.00	
	Maximum		106.00	
	Range		78.00	
	Interquartile Range		17.00	
	Skewness		-.608	.388
	Kurtosis		.996	.759
Mother DADS Amount Time 2	Mean		68.7400	3.83017
	95% Confidence Interval for Mean	Lower Bound	60.9721	
		Upper Bound	76.5079	
	5% Trimmed Mean		67.9559	
	Median		66.2900	
	Variance		542.798	
	Std. Deviation		23.29802	
	Minimum		30.00	
	Maximum		120.00	
	Range		90.00	
	Interquartile Range		32.12	
	Skewness		.495	.388
	Kurtosis		-.356	.759
Mother DADS Helpfulness Time 2	Mean		72.2703	2.81903
	95% Confidence Interval for Mean	Lower Bound	66.5530	
		Upper Bound	77.9875	
	5% Trimmed Mean		71.7568	
	Median		71.0000	
	Variance		294.036	
	Std. Deviation		17.14748	
	Minimum		43.00	
	Maximum		114.00	
	Range		71.00	
	Interquartile Range		23.00	
	Skewness		.392	.388
	Kurtosis		-.239	.759

Fathers' DADS Scores Descriptive Statistics

Descriptives

			Statistic	Std. Error
Father DADS Amount Time 1	Mean		72.8632	2.55141
	95% Confidence Interval for Mean	Lower Bound	67.6887	
		Upper Bound	78.0377	
	5% Trimmed Mean		73.0870	
	Median		75.2000	
	Variance		240.858	
	Std. Deviation		15.51961	
	Minimum		42.00	
	Maximum		99.69	
	Range		57.69	
	Interquartile Range		24.41	
	Skewness		-.244	.388
	Kurtosis		-.978	.759
Father DADS Helpfulness Time 1	Mean		70.0541	2.24041
	95% Confidence Interval for Mean	Lower Bound	65.5103	
		Upper Bound	74.5978	
	5% Trimmed Mean		69.7988	
	Median		67.0000	
	Variance		185.719	
	Std. Deviation		13.62788	
	Minimum		47.00	
	Maximum		100.00	
	Range		53.00	
	Interquartile Range		22.00	
	Skewness		.269	.388
	Kurtosis		-.751	.759
Father DADS Amount Time 2	Mean		62.8859	2.43042
	95% Confidence Interval for Mean	Lower Bound	57.9568	
		Upper Bound	67.8151	
	5% Trimmed Mean		62.2485	
	Median		62.4000	
	Variance		218.557	
	Std. Deviation		14.78368	
	Minimum		36.00	
	Maximum		102.55	
	Range		66.55	
	Interquartile Range		22.81	
	Skewness		.529	.388
	Kurtosis		.196	.759
Father DADS Helpfulness Time 2	Mean		66.2038	3.06826
	95% Confidence Interval for Mean	Lower Bound	59.9811	
		Upper Bound	72.4265	
	5% Trimmed Mean		65.6123	
	Median		61.0000	
	Variance		348.327	
	Std. Deviation		18.66351	
	Minimum		29.00	
	Maximum		109.00	
	Range		80.00	
	Interquartile Range		31.00	
	Skewness		.655	.388
	Kurtosis		-.102	.759

Mothers' DADS Scores as predictors of Mothers' DASS Scores

Simple linear regression:
Predictor variable Mothers' DADS Amount Time 1,
dependent variable Mothers' DASS Time 1

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Amount Time 1[b]	.	Enter

a. Dependent Variable: Mother DASS Total Percentile Time 1
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.101[a]	.010	-.015	28.662

a. Predictors: (Constant), Mother DADS Amount Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	336.916	1	336.916	.410	.526[b]
	Residual	32859.655	40	821.491		
	Total	33196.571	41			

a. Dependent Variable: Mother DASS Total Percentile Time 1
b. Predictors: (Constant), Mother DADS Amount Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	68.167	17.557		3.883	.000
	Mother DADS Amount Time 1	-.148	.230	-.101	-.640	.526

a. Dependent Variable: Mother DASS Total Percentile Time 1

Simple linear regression:
Predictor variable Mothers' DADS Helpfulness Time 1, dependent variable Mothers' DASS Time 1

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Helpfulness Time 1[b]	.	Enter

a. Dependent Variable: Mother DASS Total Percentile Time 1
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.234[a]	.055	.031	28.006

a. Predictors: (Constant), Mother DADS Helpfulness Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	1823.957	1	1823.957	2.326	.135[b]
	Residual	31372.615	40	784.315		
	Total	33196.571	41			

a. Dependent Variable: Mother DASS Total Percentile Time 1
b. Predictors: (Constant), Mother DADS Helpfulness Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	89.774	21.738		4.130	.000
	Mother DADS Helpfulness Time 1	-.427	.280	-.234	-1.525	.135

a. Dependent Variable: Mother DASS Total Percentile Time 1

Simple linear regression:
Predictor variable Mothers' DADS Amount Time 2, dependent variable Mothers' DASS Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Amount Time 2[b]	.	Enter

a. Dependent Variable: Mother DASS Total Percentile Time 2
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.067[a]	.004	-.024	28.573

a. Predictors: (Constant), Mother DADS Amount Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	127.264	1	127.264	.156	.695[b]
	Residual	28575.547	35	816.444		
	Total	28702.811	36			

a. Dependent Variable: Mother DASS Total Percentile Time 2
b. Predictors: (Constant), Mother DADS Amount Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	54.209	14.815		3.659	.001
	Mother DADS Amount Time 2	.081	.204	.067	.395	.695

a. Dependent Variable: Mother DASS Total Percentile Time 2

Simple linear regression:
Predictor variable Mothers' DADS Helpfulness Time 2, dependent variable Mothers' DASS Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Helpfulness Time 2[b]	.	Enter

a. Dependent Variable: Mother DASS Total Percentile Time 2
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.043[a]	.002	-.027	28.610

a. Predictors: (Constant), Mother DADS Helpfulness Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	53.646	1	53.646	.066	.799[b]
	Residual	28649.164	35	818.548		
	Total	28702.811	36			

a. Dependent Variable: Mother DASS Total Percentile Time 2
b. Predictors: (Constant), Mother DADS Helpfulness Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	64.902	20.640		3.144	.003
	Mother DADS Helpfulness Time 2	-.071	.278	-.043	-.256	.799

a. Dependent Variable: Mother DASS Total Percentile Time 2

Mothers' DADS Scores as predictors of Mothers' FAD Scores

Simple linear regression:
Predictor variable Mothers' DADS Amount Time 1,
dependent variable Mothers' FAD Time 1

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Amount Time 1[b]	.	Enter

a. Dependent Variable: Mother FAD General Functioning Time 1
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.071[a]	.005	-.020	.37717

a. Predictors: (Constant), Mother DADS Amount Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	.028	1	.028	.200	.657[b]
	Residual	5.690	40	.142		
	Total	5.719	41			

a. Dependent Variable: Mother FAD General Functioning Time 1
b. Predictors: (Constant), Mother DADS Amount Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	1.693	.231		7.329	.000
	Mother DADS Amount Time 1	-.001	.003	-.071	-.447	.657

a. Dependent Variable: Mother FAD General Functioning Time 1

Simple linear regression:
Predictor variable Mothers' DADS Helpfulness Time 1, dependent variable Mothers' FAD Time 1

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Helpfulness Time 1[b]	.	Enter

a. Dependent Variable: Mother FAD General Functioning Time 1
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.130[a]	.017	-.008	.37489

a. Predictors: (Constant), Mother DADS Helpfulness Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	.097	1	.097	.692	.411[b]
	Residual	5.622	40	.141		
	Total	5.719	41			

a. Dependent Variable: Mother FAD General Functioning Time 1
b. Predictors: (Constant), Mother DADS Helpfulness Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	1.831	.291		6.291	.000
	Mother DADS Helpfulness Time 1	-.003	.004	-.130	-.832	.411

a. Dependent Variable: Mother FAD General Functioning Time 1

Simple linear regression:
Predictor variable Mothers' DADS Amount Time 2, dependent variable Mothers' FAD Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Amount Time 2[b]	.	Enter

a. Dependent Variable: Mother FAD General Functioning Time 2
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.094[a]	.009	-.020	.44268

a. Predictors: (Constant), Mother DADS Amount Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	.061	1	.061	.309	.582[b]
	Residual	6.859	35	.196		
	Total	6.919	36			

a. Dependent Variable: Mother FAD General Functioning Time 2
b. Predictors: (Constant), Mother DADS Amount Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	1.729	.230		7.532	.000
	Mother DADS Amount Time 2	-.002	.003	-.094	-.556	.582

a. Dependent Variable: Mother FAD General Functioning Time 2

Simple linear regression:
Predictor variable Mothers' DADS Helpfulness Time 2, dependent variable Mothers' FAD Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Helpfulness Time 2[b]	.	Enter

a. Dependent Variable: Mother FAD General Functioning Time 2
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.097[a]	.009	-.019	.44255

a. Predictors: (Constant), Mother DADS Helpfulness Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	.065	1	.065	.330	.569[b]
	Residual	6.855	35	.196		
	Total	6.919	36			

a. Dependent Variable: Mother FAD General Functioning Time 2
b. Predictors: (Constant), Mother DADS Helpfulness Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	1.786	.319		5.595	.000
	Mother DADS Helpfulness Time 2	-.002	.004	-.097	-.574	.569

a. Dependent Variable: Mother FAD General Functioning Time 2

Mothers' DADS Scores as predictors of Mothers' IFS Scores

Simple linear regression:
Predictor variable Mothers' DADS Amount Time 1,
dependent variable Mothers' IFS Time 1

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Amount Time 1[b]	.	Enter

a. Dependent Variable: Mother IFS 15 item total Time 1
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.153[a]	.023	-.001	10.870

a. Predictors: (Constant), Mother DADS Amount Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	112.558	1	112.558	.953	.335[b]
	Residual	4725.942	40	118.149		
	Total	4838.500	41			

a. Dependent Variable: Mother IFS 15 item total Time 1
b. Predictors: (Constant), Mother DADS Amount Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	43.789	6.658		6.577	.000
	Mother DADS Amount Time 1	-.085	.087	-.153	-.976	.335

a. Dependent Variable: Mother IFS 15 item total Time 1

Simple linear regression:
Predictor variable Mothers' DADS Helpfulness Time 1, dependent variable Mothers' IFS Time 1

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Helpfulness Time 1[b]	.	Enter

a. Dependent Variable: Mother IFS 15 item total Time 1
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.136[a]	.018	-.006	10.896

a. Predictors: (Constant), Mother DADS Helpfulness Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	89.286	1	89.286	.752	.391[b]
	Residual	4749.214	40	118.730		
	Total	4838.500	41			

a. Dependent Variable: Mother IFS 15 item total Time 1
b. Predictors: (Constant), Mother DADS Helpfulness Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	44.688	8.458		5.284	.000
	Mother DADS Helpfulness Time 1	-.094	.109	-.136	-.867	.391

a. Dependent Variable: Mother IFS 15 item total Time 1

Simple linear regression:
Predictor variable Mothers' DADS Amount Time 2, dependent variable Mothers' IFS Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Amount Time 2[b]	.	Enter

a. Dependent Variable: Mother IFS 15 item total Time 2
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.217[a]	.047	.019	12.290

a. Predictors: (Constant), Mother DADS Amount Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	253.123	1	253.123	1.676	.204[b]
	Residual	5135.627	34	151.048		
	Total	5388.750	35			

a. Dependent Variable: Mother IFS 15 item total Time 2
b. Predictors: (Constant), Mother DADS Amount Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	43.846	6.459		6.789	.000
	Mother DADS Amount Time 2	-.115	.089	-.217	-1.295	.204

a. Dependent Variable: Mother IFS 15 item total Time 2

Simple linear regression:
Predictor variable Mothers' DADS Helpfulness Time 2, dependent variable Mothers' IFS Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Helpfulness Time 2[b]	.	Enter

a. Dependent Variable: Mother IFS 15 item total Time 2
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.001[a]	.000	-.029	12.589

a. Predictors: (Constant), Mother DADS Helpfulness Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	.005	1	.005	.000	.996[b]
	Residual	5388.745	34	158.493		
	Total	5388.750	35			

a. Dependent Variable: Mother IFS 15 item total Time 2
b. Predictors: (Constant), Mother DADS Helpfulness Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	35.964	9.092		3.956	.000
	Mother DADS Helpfulness Time 2	-.001	.123	-.001	-.005	.996

a. Dependent Variable: Mother IFS 15 item total Time 2

Fathers' DADS Scores as predictors of Fathers' DASS Scores

Simple linear regression:
Predictor variable fathers' DADS Amount Time 1,
dependent variable fathers' DASS Time 1

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Amount Time 1[b]	.	Enter

a. Dependent Variable: Father DASS Total Percentile Time 1
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.112[a]	.013	-.012	29.319

a. Predictors: (Constant), Father DADS Amount Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	439.827	1	439.827	.512	.479[b]
	Residual	34384.959	40	859.624		
	Total	34824.786	41			

a. Dependent Variable: Father DASS Total Percentile Time 1
b. Predictors: (Constant), Father DADS Amount Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	31.293	20.955		1.493	.143
	Father DADS Amount Time 1	.194	.272	.112	.715	.479

a. Dependent Variable: Father DASS Total Percentile Time 1

Simple linear regression:
Predictor variable fathers' DADS Helpfulness Time 1, dependent variable fathers' DASS Time 1

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Helpfulness Time 1[b]	.	Enter

a. Dependent Variable: Father DASS Total Percentile Time 1
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.135[a]	.018	-.006	29.238

a. Predictors: (Constant), Father DADS Helpfulness Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	630.871	1	630.871	.738	.395[b]
	Residual	34193.915	40	854.848		
	Total	34824.786	41			

a. Dependent Variable: Father DASS Total Percentile Time 1
b. Predictors: (Constant), Father DADS Helpfulness Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	28.374	20.927		1.356	.183
	Father DADS Helpfulness Time 1	.243	.283	.135	.859	.395

a. Dependent Variable: Father DASS Total Percentile Time 1

Simple linear regression:
Predictor variable fathers' DADS Amount Time 2, dependent variable fathers' DASS Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Amount Time 2[b]	.	Enter

a. Dependent Variable: Father DASS Total Percentile Time 2
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.035[a]	.001	-.027	29.764

a. Predictors: (Constant), Father DADS Amount Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	37.257	1	37.257	.042	.839[b]
	Residual	31005.554	35	885.873		
	Total	31042.811	36			

a. Dependent Variable: Father DASS Total Percentile Time 2
b. Predictors: (Constant), Father DADS Amount Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	38.429	21.661		1.774	.085
	Father DADS Amount Time 2	.069	.336	.035	.205	.839

a. Dependent Variable: Father DASS Total Percentile Time 2

Simple linear regression:
Predictor variable fathers' DADS Helpfulness Time 2, dependent variable fathers' DASS Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Helpfulness Time 2[b]	.	Enter

a. Dependent Variable: Father DASS Total Percentile Time 2
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.148[a]	.022	-.006	29.452

a. Predictors: (Constant), Father DADS Helpfulness Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	683.743	1	683.743	.788	.381[b]
	Residual	30359.068	35	867.402		
	Total	31042.811	36			

a. Dependent Variable: Father DASS Total Percentile Time 2
b. Predictors: (Constant), Father DADS Helpfulness Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	27.298	18.073		1.510	.140
	Father DADS Helpfulness Time 2	.234	.263	.148	.888	.381

a. Dependent Variable: Father DASS Total Percentile Time 2

Fathers' DADS Scores as predictors of Fathers' FAD Scores

Simple linear regression:
Predictor variable fathers' DADS Amount Time 1,
dependent variable fathers' FAD Time 1

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Amount T1[b]	.	Enter

a. Dependent Variable: Father FAD General Functioning T1
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.223[a]	.050	.026	.40527

a. Predictors: (Constant), Father DADS Amount T1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	.344	1	.344	2.092	.156[b]
	Residual	6.570	40	.164		
	Total	6.913	41			

a. Dependent Variable: Father FAD General Functioning T1
b. Predictors: (Constant), Father DADS Amount T1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	2.021	.290		6.976	.000
	Father DADS Amount T1	-.005	.004	-.223	-1.446	.156

a. Dependent Variable: Father FAD General Functioning T1

Simple linear regression:
Predictor variable fathers' DADS Helpfulness Time 1, dependent variable fathers' FAD Time 1

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Helpfulness T1[b]	.	Enter

a. Dependent Variable: Father FAD General Functioning T1
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.062[a]	.004	-.021	.41493

a. Predictors: (Constant), Father DADS Helpfulness T1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	.026	1	.026	.153	.698[b]
	Residual	6.887	40	.172		
	Total	6.913	41			

a. Dependent Variable: Father FAD General Functioning T1
b. Predictors: (Constant), Father DADS Helpfulness T1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	1.725	.297		5.809	.000
	Father DADS Helpfulness T1	-.002	.004	-.062	-.391	.698

a. Dependent Variable: Father FAD General Functioning T1

Simple linear regression: Predictor variable fathers' DADS Amount Time 2, dependent variable fathers' FAD Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Amount Time 2[b]	.	Enter

a. Dependent Variable: Father FAD General Functioning Time 2
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.367[a]	.135	.110	.36721

a. Predictors: (Constant), Father DADS Amount Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	.735	1	.735	5.453	.025[b]
	Residual	4.720	35	.135		
	Total	5.455	36			

a. Dependent Variable: Father FAD General Functioning Time 2
b. Predictors: (Constant), Father DADS Amount Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	2.262	.267		8.464	.000
	Father DADS Amount Time 2	-.010	.004	-.367	-2.335	.025

a. Dependent Variable: Father FAD General Functioning Time 2

Simple linear regression:
Predictor variable fathers' DADS Helpfulness Time 2, dependent variable fathers' FAD Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Helpfulness Time 2[b]	.	Enter

a. Dependent Variable: Father FAD General Functioning Time 2
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.216[a]	.047	.020	.38543

a. Predictors: (Constant), Father DADS Helpfulness Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	.255	1	.255	1.719	.198[b]
	Residual	5.200	35	.149		
	Total	5.455	36			

a. Dependent Variable: Father FAD General Functioning Time 2
b. Predictors: (Constant), Father DADS Helpfulness Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	1.953	.237		8.256	.000
	Father DADS Helpfulness Time 2	-.005	.003	-.216	-1.311	.198

a. Dependent Variable: Father FAD General Functioning Time 2

Fathers' DADS Scores as predictors of Fathers' IFS Scores

Simple linear regression:
Predictor variable fathers' DADS Amount Time 1,
dependent variable fathers' IFS Time 1

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Amount T1[b]	.	Enter

a. Dependent Variable: Father IFS 15 item total T1
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.014[a]	.000	-.025	8.937

a. Predictors: (Constant), Father DADS Amount T1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	.606	1	.606	.008	.931[b]
	Residual	3195.013	40	79.875		
	Total	3195.619	41			

a. Dependent Variable: Father IFS 15 item total T1
b. Predictors: (Constant), Father DADS Amount T1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	34.781	6.388		5.445	.000
	Father DADS Amount T1	-.007	.083	-.014	-.087	.931

a. Dependent Variable: Father IFS 15 item total T1

Simple linear regression:
Predictor variable fathers' DADS Helpfulness Time 1, dependent variable fathers' IFS Time 1

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Helpfulness T1[b]	.	Enter

a. Dependent Variable: Father IFS 15 item total T1
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.151[a]	.023	-.002	8.835

a. Predictors: (Constant), Father DADS Helpfulness T1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	73.239	1	73.239	.938	.339[b]
	Residual	3122.380	40	78.059		
	Total	3195.619	41			

a. Dependent Variable: Father IFS 15 item total T1
b. Predictors: (Constant), Father DADS Helpfulness T1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	40.219	6.324		6.360	.000
	Father DADS Helpfulness T1	-.083	.086	-.151	-.969	.339

a. Dependent Variable: Father IFS 15 item total T1

Simple linear regression:
Predictor variable fathers' DADS Amount Time 2, dependent variable fathers' IFS Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Amount Time 2[b]	.	Enter

a. Dependent Variable: Father IFS 15 item total Time 2
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.320[a]	.102	.076	11.338

a. Predictors: (Constant), Father DADS Amount Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	511.853	1	511.853	3.982	.054[b]
	Residual	4499.066	35	128.545		
	Total	5010.919	36			

a. Dependent Variable: Father IFS 15 item total Time 2
b. Predictors: (Constant), Father DADS Amount Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	17.366	8.251		2.105	.043
	Father DADS Amount Time 2	.255	.128	.320	1.995	.054

a. Dependent Variable: Father IFS 15 item total Time 2

Simple linear regression:
Predictor variable fathers' DADS Helpfulness Time 2, dependent variable fathers' IFS Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Helpfulness Time 2[b]	.	Enter

a. Dependent Variable: Father IFS 15 item total Time 2
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.284[a]	.081	.054	11.474

a. Predictors: (Constant), Father DADS Helpfulness Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	403.459	1	403.459	3.065	.089[b]
	Residual	4607.460	35	131.642		
	Total	5010.919	36			

a. Dependent Variable: Father IFS 15 item total Time 2
b. Predictors: (Constant), Father DADS Helpfulness Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.
		B	Std. Error	Beta		
1	(Constant)	21.530	7.041		3.058	.004
	Father DADS Helpfulness Time 2	.179	.102	.284	1.751	.089

a. Dependent Variable: Father IFS 15 item total Time 2

APPENDIX J
HYPOTHESIS 3

Building the Multiple Regression Model: Demographic Variables (Mothers)

Predictor variables: demographic variables

Dependent variable: mothers' CBCL Total Problem T score

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	First born child or later, Socio Economic Index, Mother's Age[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary[b]

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.461[a]	.213	.141	10.718

a. Predictors: (Constant), First born child or later, Socio Economic Index, Mother's Age
b. Dependent Variable: Mother CBCL Total Problems T-Score

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	1023.110	3	341.037	2.969	.046[b]
	Residual	3791.160	33	114.884		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), First born child or later, Socio Economic Index, Mother's Age

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	71.496	11.366		6.290	.000	48.371	94.622
	Socio Economic Index	-.144	.076	-.304	-1.893	.067	-.299	.011
	Mother's Age	-.476	.320	-.260	-1.486	.147	-1.128	.176
	First born child or later	1.561	4.143	.064	.377	.709	-6.869	9.991

a. Dependent Variable: Mother CBCL Total Problems T-Score

Building the Multiple Regression Model: Demographic Variables (Fathers)

Predictor variables: demographic variables

Dependent variable: fathers' CBCL Total Problem T score

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	First born child or later, Socio Economic Index, Father's Age[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary[b]

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.462[a]	.213	.142	10.932

a. Predictors: (Constant), First born child or later, Socio Economic Index, Father's Age
b. Dependent Variable: Father CBCL Total Problems T-Score

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	1069.145	3	356.382	2.982	.045[b]
	Residual	3943.773	33	119.508		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), First born child or later, Socio Economic Index, Father's Age

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	64.348	11.167		5.762	.000	41.629	87.067
	Socio Economic Index	-.192	.077	-.398	-2.499	.018	-.348	-.036
	Father's Age	-.119	.291	-.071	-.408	.686	-.711	.473
	First born child or later	-5.232	4.225	-.210	-1.238	.224	-13.829	3.364

a. Dependent Variable: Father CBCL Total Problems T-Score

Building the Multiple Regression Model: Illness Variables (Mothers)

Predictor variables: illness variables Time 1

Dependent variable: mothers' CBCL Total Problems T scores at Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Sqrt transformation ratio ADM T1, Biliary atresia or other severe liver disease, Sqrt transformation ratio OPD T1, Liver transplant at Time 1 or not[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary[b]

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.354[a]	.125	.016	11.473

a. Predictors: (Constant), Sqrt transformation ratio ADM T1, Biliary atresia or other severe liver disease, Sqrt transformation ratio OPD T1, Liver transplant at Time 1 or not
b. Dependent Variable: Mother CBCL Total Problems T-Score

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	602.073	4	150.518	1.143	.354[b]
	Residual	4212.198	32	131.631		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Sqrt transformation ratio ADM T1, Biliary atresia or other severe liver disease, Sqrt transformation ratio OPD T1, Liver transplant at Time 1 or not

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	41.484	6.352		6.530	.000	28.544	54.423
	Biliary atresia or other severe liver disease	-5.505	4.122	-.239	-1.335	.191	-13.901	2.892
	Liver transplant at Time 1 or not	3.632	5.503	.131	.660	.514	-7.577	14.841
	Sqrt transformation ratio OPD T1	24.865	33.140	.135	.750	.459	-42.640	92.369
	Sqrt transformation ratio ADM T1	7.120	9.818	.148	.725	.474	-12.878	27.118

a. Dependent Variable: Mother CBCL Total Problems T-Score

Predictor variables: illness variables Time 2

Dependent variable: mothers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Sqrt transformation ratio ADM T2, Biliary atresia or other severe liver disease, Sqrt transformation ratio OPD T2, Liver transplant at Time 2 or not[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary[b]

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.543[a]	.295	.207	10.298

a. Predictors: (Constant), Sqrt transformation ratio ADM T2, Biliary atresia or other severe liver disease, Sqrt transformation ratio OPD T2, Liver transplant at Time 2 or not
b. Dependent Variable: Mother CBCL Total Problems T-Score

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	1420.947	4	355.237	3.350	.021[b]
	Residual	3393.323	32	106.041		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Sqrt transformation ratio ADM T2, Biliary atresia or other severe liver disease, Sqrt transformation ratio OPD T2, Liver transplant at Time 2 or not

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	29.588	7.049		4.198	.000	15.231	43.946
	Biliary atresia or other severe liver disease	-7.690	3.800	-.334	-2.024	.051	-15.430	.050
	Liver transplant at Time 2 or not	-3.071	6.691	-.135	-.459	.649	-16.699	10.557
	Sqrt transformation ratio OPD T2	140.138	48.081	.719	2.915	.006	42.199	238.077
	Sqrt transformation ratio ADM T2	-9.245	16.374	-.148	-.565	.576	-42.597	24.108

a. Dependent Variable: Mother CBCL Total Problems T-Score

Building the Multiple Regression Model: Illness Variables (Fathers)

Predictor variables: illness variables Time 1

Dependent variable: fathers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Sqrt transformation ratio ADM T1, Biliary atresia or other severe liver disease, Sqrt transformation ratio OPD T1, Liver transplant at Time 1 or not[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary[b]

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.525[a]	.276	.186	10.649

a. Predictors: (Constant), Sqrt transformation ratio ADM T1, Biliary atresia or other severe liver disease, Sqrt transformation ratio OPD T1, Liver transplant at Time 1 or not
b. Dependent Variable: Father CBCL Total Problems T-Score

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	1384.295	4	346.074	3.052	.031[b]
	Residual	3628.624	32	113.394		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Sqrt transformation ratio ADM T1, Biliary atresia or other severe liver disease, Sqrt transformation ratio OPD T1, Liver transplant at Time 1 or not

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	42.727	5.896		7.247	.000	30.718	54.737
	Biliary atresia or other severe liver disease	-8.644	3.826	-.368	-2.259	.031	-16.437	-.851
	Liver transplant at Time 1 or not	8.846	5.108	.313	1.732	.093	-1.558	19.250
	Sqrt transformation ratio OPD T1	31.169	30.759	.166	1.013	.319	-31.485	93.823
	Sqrt transformation ratio ADM T1	1.958	9.112	.040	.215	.831	-16.603	20.519

a. Dependent Variable: Father CBCL Total Problems T-Score

Predictor variables: illness variables Time 2

Dependent variable: fathers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Sqrt transformation ratio ADM T2, Biliary atresia or other severe liver disease, Sqrt transformation ratio OPD T2, Liver transplant at Time 2 or not[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary[b]

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.540[a]	.292	.204	10.531

a. Predictors: (Constant), Sqrt transformation ratio ADM T2, Biliary atresia or other severe liver disease, Sqrt transformation ratio OPD T2, Liver transplant at Time 2 or not
b. Dependent Variable: Father CBCL Total Problems T-Score

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	1463.948	4	365.987	3.300	.023[b]
	Residual	3548.971	32	110.905		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Sqrt transformation ratio ADM T2, Biliary atresia or other severe liver disease, Sqrt transformation ratio OPD T2, Liver transplant at Time 2 or not

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	39.401	7.208		5.466	.000	24.718	54.084
	Biliary atresia or other severe liver disease	-11.280	3.886	-.480	-2.903	.007	-19.195	-3.364
	Liver transplant at Time 2 or not	4.889	6.842	.210	.714	.480	-9.049	18.826
	Sqrt transformation ratio OPD T2	76.592	49.172	.385	1.558	.129	-23.568	176.751
	Sqrt transformation ratio ADM T2	-10.607	16.745	-.167	-.633	.531	-44.716	23.501

a. Dependent Variable: Father CBCL Total Problems T-Score

Building the Multiple Regression Model: Study Measures Variables (Mothers)

Simple linear regression

Predictor variable mothers' DASS Time 1

Dependent variable mothers' CBCL Total Problems score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DASS Total Percentile Time 1[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.188[a]	.035	.008	11.519

a. Predictors: (Constant), Mother DASS Total Percentile Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	169.901	1	169.901	1.280	.266[b]
	Residual	4644.369	35	132.696		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Mother DASS Total Percentile Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	42.404	4.309		9.840	.000	33.655	51.152
	Mother DASS Total Percentile Time 1	.077	.068	.188	1.132	.266	-.061	.215

a. Dependent Variable: Mother CBCL Total Problems T-Score

Simple linear regression

Predictor variable mothers' DASS Time 2

Dependent variable mothers' CBCL Total Problems score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DASS Total Percentile Time 2[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.293[a]	.086	.060	11.214

a. Predictors: (Constant), Mother DASS Total Percentile Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	412.533	1	412.533	3.280	.079[b]
	Residual	4401.737	35	125.764		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Mother DASS Total Percentile Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	39.620	4.364		9.079	.000	30.760	48.479
	Mother DASS Total Percentile Time 2	.120	.066	.293	1.811	.079	-.014	.254

a. Dependent Variable: Mother CBCL Total Problems T-Score

Simple linear regression

Predictor variable mothers' FAD Time 1

Dependent variable mothers' CBCL Total Problems score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother FAD General Functioning Time 1[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.024[a]	.001	-.028	11.725

a. Predictors: (Constant), Mother FAD General Functioning Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	2.869	1	2.869	.021	.886[b]
	Residual	4811.402	35	137.469		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Mother FAD General Functioning Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	48.041	8.916		5.388	.000	29.942	66.141
	Mother FAD General Functioning Time 1	-.787	5.450	-.024	-.144	.886	-11.852	10.278

a. Dependent Variable: Mother CBCL Total Problems T-Score

Simple linear regression

Predictor variable mothers' FAD Time 2

Dependent variable mothers' CBCL Total Problems score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother FAD General Functioning Time 2[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.023[a]	.001	-.028	11.725

a. Predictors: (Constant), Mother FAD General Functioning Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	2.492	1	2.492	.018	.894[b]
	Residual	4811.778	35	137.479		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Mother FAD General Functioning Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	45.819	7.422		6.174	.000	30.752	60.885
	Mother FAD General Functioning Time 2	.600	4.457	.023	.135	.894	-8.449	9.649

a. Dependent Variable: Mother CBCL Total Problems T-Score

Simple linear regression

Predictor variable: mothers' IFS score Time 1

Dependent variable: mothers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother IFS 15 item total Time 1[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.450[a]	.203	.180	10.471

a. Predictors: (Constant), Mother IFS 15 item total Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	976.490	1	976.490	8.905	.005[b]
	Residual	3837.781	35	109.651		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Mother IFS 15 item total Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	29.179	6.145		4.748	.000	16.703	41.655
	Mother IFS 15 item total Time 1	.476	.160	.450	2.984	.005	.152	.800

a. Dependent Variable: Mother CBCL Total Problems T-Score

Simple linear regression

Predictor variable: mothers' IFS score Time 2

Dependent variable: mothers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother IFS 15 item total Time 2[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.432[a]	.187	.163	10.713

a. Predictors: (Constant), Mother IFS 15 item total Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	897.079	1	897.079	7.816	.008[b]
	Residual	3902.476	34	114.779		
	Total	4799.556	35			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Mother IFS 15 item total Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	32.235	5.538		5.821	.000	20.981	43.488
	Mother IFS 15 item total Time 2	.408	.146	.432	2.796	.008	.111	.705

a. Dependent Variable: Mother CBCL Total Problems T-Score

Simple linear regression

Predictor variable: mothers' DADS Amount score Time 1

Dependent variable: mothers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Amount Time 1[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.011[a]	.000	-.028	11.727

a. Predictors: (Constant), Mother DADS Amount Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	.579	1	.579	.004	.949[b]
	Residual	4813.691	35	137.534		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Mother DADS Amount Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	47.247	7.392		6.391	.000	32.240	62.254
	Mother DADS Amount Time 1	-.006	.099	-.011	-.065	.949	-.207	.195

a. Dependent Variable: Mother CBCL Total Problems T-Score

Simple linear regression

Predictor variable: mothers' DADS Amount score Time 2

Dependent variable: mothers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Amount Time 2[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.147[a]	.021	-.006	11.601

a. Predictors: (Constant), Mother DADS Amount Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	103.482	1	103.482	.769	.387[b]
	Residual	4710.788	35	134.594		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Mother DADS Amount Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	51.786	6.015		8.609	.000	39.574	63.998
	Mother DADS Amount Time 2	-.073	.083	-.147	-.877	.387	-.241	.096

a. Dependent Variable: Mother CBCL Total Problems T-Score

Simple linear regression

Predictor variable: mothers' DADS Helpfulness score Time 1

Dependent variable: mothers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Helpfulness Time 1[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.106[a]	.011	-.017	11.662

a. Predictors: (Constant), Mother DADS Helpfulness Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	53.893	1	53.893	.396	.533[b]
	Residual	4760.377	35	136.011		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Mother DADS Helpfulness Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	52.646	9.508		5.537	.000	33.344	71.948
	Mother DADS Helpfulness Time 1	-.079	.125	-.106	-.629	.533	-.332	.175

a. Dependent Variable: Mother CBCL Total Problems T-Score

Simple linear regression

Predictor variable: mothers' DADS Helpfulness score Time 2

Dependent variable: mothers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Mother DADS Helpfulness Time 2[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.114[a]	.013	-.015	11.652

a. Predictors: (Constant), Mother DADS Helpfulness Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	62.571	1	62.571	.461	.502[b]
	Residual	4751.699	35	135.763		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Mother DADS Helpfulness Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	52.340	8.406		6.227	.000	35.276	69.405
	Mother DADS Helpfulness Time 2	-.077	.113	-.114	-.679	.502	-.307	.153

a. Dependent Variable: Mother CBCL Total Problems T-Score

Building the Multiple Regression Model: Study Measures Variables (Fathers)

Simple linear regression

Predictor variable: fathers' DASS score Time 1

Dependent variable: fathers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DASS Total Percentile Time 1[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.280[a]	.079	.052	11.487

a. Predictors: (Constant), Father DASS Total Percentile Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	394.367	1	394.367	2.989	.093[b]
	Residual	4618.552	35	131.959		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Father DASS Total Percentile Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	40.948	3.678		11.132	.000	33.481	48.416
	Father DASS Total Percentile Time 1	.118	.068	.280	1.729	.093	-.021	.256

a. Dependent Variable: Father CBCL Total Problems T-Score

Simple linear regression

Predictor variable: fathers' DASS score Time 2

Dependent variable: fathers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DASS Total Percentile Time 2[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.268[a]	.072	.045	11.530

a. Predictors: (Constant), Father DASS Total Percentile Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	360.147	1	360.147	2.709	.109[b]
	Residual	4652.772	35	132.936		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Father DASS Total Percentile Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	41.800	3.380		12.368	.000	34.939	48.661
	Father DASS Total Percentile Time 2	.108	.065	.268	1.646	.109	-.025	.241

a. Dependent Variable: Father CBCL Total Problems T-Score

Simple linear regression

Predictor variable: fathers' FAD score Time 1

Dependent variable: fathers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father FAD General Functioning Time 1[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.136[a]	.019	-.009	11.856

a. Predictors: (Constant), Father FAD General Functioning Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	93.204	1	93.204	.663	.421[b]
	Residual	4919.715	35	140.563		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Father FAD General Functioning Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	39.791	8.354		4.763	.000	22.832	56.750
	Father FAD General Functioning Time 1	4.049	4.973	.136	.814	.421	-6.046	14.145

a. Dependent Variable: Father CBCL Total Problems T-Score

Simple linear regression

Predictor variable: fathers' FAD score Time 2

Dependent variable: fathers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father FAD General Functioning Time 2[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.085[a]	.007	-.021	11.925

a. Predictors: (Constant), Father FAD General Functioning Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	35.927	1	35.927	.253	.618[b]
	Residual	4976.992	35	142.200		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Father FAD General Functioning Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	42.161	8.670		4.863	.000	24.560	59.761
	Father FAD General Functioning Time 2	2.566	5.106	.085	.503	.618	-7.799	12.932

a. Dependent Variable: Father CBCL Total Problems T-Score

Simple linear regression

Predictor variable: fathers' IFS score Time 1

Dependent variable: fathers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father IFS 15 item total Time 1[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.435[a]	.189	.166	10.775

a. Predictors: (Constant), Father IFS 15 item total Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	949.124	1	949.124	8.174	.007[b]
	Residual	4063.795	35	116.108		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Father IFS 15 item total Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	26.668	7.127		3.742	.001	12.199	41.137
	Father IFS 15 item total Time 1	.576	.201	.435	2.859	.007	.167	.985

a. Dependent Variable: Father CBCL Total Problems T-Score

Simple linear regression

Predictor variable: fathers' IFS score Time 2

Dependent variable: fathers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father IFS 15 item total Time 2[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.429[a]	.184	.161	10.810

a. Predictors: (Constant), Father IFS 15 item total Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	923.274	1	923.274	7.902	.008[b]
	Residual	4089.645	35	116.847		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Father IFS 15 item total Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	32.066	5.402		5.936	.000	21.100	43.033
	Father IFS 15 item total Time 2	.429	.153	.429	2.811	.008	.119	.739

a. Dependent Variable: Father CBCL Total Problems T-Score

Simple linear regression

Predictor variable: fathers' DADS Amount score Time 1

Dependent variable: fathers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Amount Time 1[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.035[a]	.001	-.027	11.960

a. Predictors: (Constant), Father DADS Amount Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	6.299	1	6.299	.044	.835[b]
	Residual	5006.620	35	143.046		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Father DADS Amount Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	48.369	9.563		5.058	.000	28.955	67.783
	Father DADS Amount Time 1	-.027	.128	-.035	-.210	.835	-.288	.234

a. Dependent Variable: Father CBCL Total Problems T-Score

Simple linear regression

Predictor variable: fathers' DADS Amount score Time 2

Dependent variable: fathers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Amount Time 2[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.065[a]	.004	-.024	11.942

a. Predictors: (Constant), Father DADS Amount Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	21.469	1	21.469	.151	.700[b]
	Residual	4991.450	35	142.613		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Father DADS Amount Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	43.120	8.691		4.961	.000	25.477	60.764
	Father DADS Amount Time 2	.052	.135	.065	.388	.700	-.221	.326

a. Dependent Variable: Father CBCL Total Problems T-Score

Simple linear regression

Predictor variable: fathers' DADS Helpfulness score Time 1

Dependent variable: fathers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Helpfulness Time 1[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.244[a]	.059	.032	11.607

a. Predictors: (Constant), Father DADS Helpfulness Time 1

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	297.437	1	297.437	2.208	.146[b]
	Residual	4715.482	35	134.728		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Father DADS Helpfulness Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	31.630	10.126		3.124	.004	11.073	52.186
	Father DADS Helpfulness Time 1	.211	.142	.244	1.486	.146	-.077	.499

a. Dependent Variable: Father CBCL Total Problems T-Score

Simple linear regression

Predictor variable: fathers' DADS Helpfulness score Time 2

Dependent variable: fathers' CBCL Total Problems T score Time 2

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Father DADS Helpfulness Time 2[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.125[a]	.016	-.012	11.874

a. Predictors: (Constant), Father DADS Helpfulness Time 2

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	78.501	1	78.501	.557	.461[b]
	Residual	4934.418	35	140.983		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Father DADS Helpfulness Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	41.167	7.286		5.650	.000	26.376	55.959
	Father DADS Helpfulness Time 2	.079	.106	.125	.746	.461	-.136	.294

a. Dependent Variable: Father CBCL Total Problems T-Score

Hierarchical Multiple Regression: Mothers

Significant demographic and illness predictors (Block 1), mothers' IFS scores at Time 1 (Block 2), and mothers' DASS scores at Time 2 (Block 3)

Descriptive Statistics

	Mean	Std. Deviation	N
Mother CBCL Total Problems T-Score	46.78	11.564	37
Socio Economic Index	62.305	24.4339	37
Biliary atresia or other severe liver disease	.57	.502	37
Sqrt transformation ratio OPD Time 2	.1859	.05937	37
Mother IFS 15 item total Time 1	36.97	10.938	37
Mother DASS Total Percentile Time 2	59.76	28.237	37

Correlations

		Mother CBCL Total Problems T-Score	Socio Economic Index	Biliary atresia or other severe liver disease	Sqrt transformation ratio OPD Time 2	Mother IFS 15 item total Time 1	Mother DASS Total Percentile Time 2
Pearson Correlation	Mother CBCL Total Problems T-Score	1.000	-.362	-.179	.386	.450	.293
	Socio Economic Index	-.362	1.000	.235	-.062	-.329	-.290
	Biliary atresia or other severe liver disease	-.179	.235	1.000	.368	.266	-.182
	Sqrt transformation ratio OPD Time 2	.386	-.062	.368	1.000	.516	-.112
	Mother IFS 15 item total Time 1	.450	-.329	.266	.516	1.000	.132
	Mother DASS Total Percentile Time 2	.293	-.290	-.182	-.112	.132	1.000
Sig. (1-tailed)	Mother CBCL Total Problems T-Score	.	.014	.144	.009	.003	.039
	Socio Economic Index	.014	.	.081	.359	.023	.041
	Biliary atresia or other severe liver disease	.144	.081	.	.012	.056	.141
	Sqrt transformation ratio OPD Time 2	.009	.359	.012	.	.001	.255
	Mother IFS 15 item total Time 1	.003	.023	.056	.001	.	.218
	Mother DASS Total Percentile Time 2	.039	.041	.141	.255	.218	.
N	Mother CBCL Total Problems T-Score	37	37	37	37	37	37
	Socio Economic Index	37	37	37	37	37	37
	Biliary atresia or other severe liver disease	37	37	37	37	37	37
	Sqrt transformation ratio OPD Time 2	37	37	37	37	37	37
	Mother IFS 15 item total Time 1	37	37	37	37	37	37
	Mother DASS Total Percentile Time 2	37	37	37	37	37	37

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Sqrt transformation ratio OPD Time 2, Socio Economic Index, Biliary atresia or other severe liver disease[b]	.	Enter
2	Mother IFS 15 item total Time 1[b]	.	Enter
3	Mother DASS Total Percentile Time 2[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary[d]

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate	Change Statistics					Durbin-Watson
					R Square Change	F Change	df1	df2	Sig. F Change	
1	.577[a]	.333	.272	9.868	.333	5.480	3	33	.004	
2	.628[b]	.394	.318	9.547	.062	3.259	1	32	.080	
3	.657[c]	.431	.340	9.396	.037	2.034	1	31	.164	2.328

a. Predictors: (Constant), Sqrt transformation ratio OPD Time 2, Socio Economic Index, Biliary atresia or other severe liver disease
b. Predictors: (Constant), Sqrt transformation ratio OPD Time 2, Socio Economic Index, Biliary atresia or other severe liver disease, Mother IFS 15 item total Time 1
c. Predictors: (Constant), Sqrt transformation ratio OPD Time 2, Socio Economic Index, Biliary atresia or other severe liver disease, Mother IFS 15 item total Time 1, Mother DASS Total Percentile Time 2
d. Dependent Variable: Mother CBCL Total Problems T-Score

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	1600.815	3	533.605	5.480	.004[b]
	Residual	3213.455	33	97.377		
	Total	4814.270	36			
2	Regression	1897.795	4	474.449	5.206	.002[c]
	Residual	2916.475	32	91.140		
	Total	4814.270	36			
3	Regression	2077.347	5	415.469	4.706	.003[d]
	Residual	2736.923	31	88.288		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Sqrt transformation ratio OPD Time 2, Socio Economic Index, Biliary atresia or other severe liver disease
c. Predictors: (Constant), Sqrt transformation ratio OPD Time 2, Socio Economic Index, Biliary atresia or other severe liver disease, Mother IFS 15 item total Time 1
d. Predictors: (Constant), Sqrt transformation ratio OPD Time 2, Socio Economic Index, Biliary atresia or other severe liver disease, Mother IFS 15 item total Time 1, Mother DASS Total Percentile Time 2

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B		Correlations			Collinearity Statistics	
		B	Std. Error	Beta			Lower Bound	Upper Bound	Zero-order	Partial	Part	Tolerance	VIF
1	(Constant)	41.097	7.169		5.732	.000	26.510	55.683					
	Socio Economic Index	-.125	.070	-.264	-1.779	.084	-.268	.018	-.362	-.296	-.253	.919	1.088
	Biliary atresia or other severe liver disease	-6.747	3.667	-.293	-1.840	.075	-14.207	.712	-.179	-.305	-.262	.798	1.254
	Sqrt transformation ratio OPD Time 2	93.056	30.206	.478	3.081	.004	31.601	154.512	.386	.473	.438	.841	1.189
2	(Constant)	31.122	8.868		3.509	.001	13.059	49.185					
	Socio Economic Index	-.073	.074	-.154	-.985	.332	-.223	.078	-.362	-.172	-.136	.778	1.285
	Biliary atresia or other severe liver disease	-8.144	3.631	-.354	-2.243	.032	-15.539	-.749	-.179	-.369	-.309	.761	1.313
	Sqrt transformation ratio OPD Time 2	66.921	32.613	.344	2.052	.048	.491	133.351	.386	.341	.282	.675	1.481
	Mother IFS 15 item total Time 1	.335	.185	.317	1.805	.080	-.043	.712	.450	.304	.248	.616	1.624
3	(Constant)	24.612	9.850		2.499	.018	4.523	44.701					
	Socio Economic Index	-.052	.074	-.110	-.700	.489	-.203	.099	-.362	-.125	-.095	.748	1.337
	Biliary atresia or other severe liver disease	-7.595	3.594	-.330	-2.113	.043	-14.925	-.265	-.179	-.355	-.286	.753	1.329
	Sqrt transformation ratio OPD Time 2	74.045	32.485	.380	2.279	.030	7.792	140.298	.386	.379	.309	.659	1.517
	Mother IFS 15 item total Time 1	.295	.185	.279	1.595	.121	-.082	.671	.450	.275	.216	.601	1.663
	Mother DASS Total Percentile Time 2	.085	.059	.207	1.426	.164	-.036	.206	.293	.248	.193	.873	1.145

a. Dependent Variable: Mother CBCL Total Problems T-Score

Excluded Variables[a]

Model		Beta In	t	Sig.	Partial Correlation	Collinearity Statistics		
						Tolerance	VIF	Minimum Tolerance
1	Mother IFS 15 item total Time 1	.317[b]	1.805	.080	.304	.616	1.624	.616
	Mother DASS Total Percentile Time 2	.242[b]	1.649	.109	.280	.894	1.119	.793
2	Mother DASS Total Percentile Time 2	.207[c]	1.426	.164	.248	.873	1.145	.601

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors in the Model: (Constant), Sqrt transformation ratio OPD Time 2, Socio Economic Index, Biliary atresia or other severe liver disease
c. Predictors in the Model: (Constant), Sqrt transformation ratio OPD Time 2, Socio Economic Index, Biliary atresia or other severe liver disease, Mother IFS 15 item total Time 1

Collinearity Diagnostics[a]

Model	Dimension	Eigenvalue	Condition Index	Variance Proportions					
				(Constant)	Socio Economic Index	Biliary atresia or other severe liver disease	Sqrt transformation ratio OPD Time 2	Mother IFS 15 item total Time 1	Mother DASS Total Percentile Time 2
1	1	3.542	1.000	.00	.01	.02	.01		
	2	.309	3.388	.02	.03	.86	.01		
	3	.118	5.478	.01	.57	.01	.26		
	4	.031	10.640	.97	.39	.11	.72		
2	1	4.460	1.000	.00	.00	.01	.00	.00	
	2	.321	3.730	.01	.01	.85	.00	.01	
	3	.157	5.327	.00	.42	.00	.04	.07	
	4	.042	10.296	.06	.01	.04	.95	.29	
	5	.021	14.700	.93	.56	.10	.00	.63	
3	1	5.249	1.000	.00	.00	.01	.00	.00	.00
	2	.396	3.641	.00	.00	.58	.00	.00	.10
	3	.176	5.454	.00	.36	.15	.00	.01	.17
	4	.123	6.522	.00	.06	.19	.15	.07	.45
	5	.037	11.951	.02	.00	.00	.82	.55	.13
	6	.018	16.947	.97	.58	.07	.03	.36	.15

a. Dependent Variable: Mother CBCL Total Problems T-Score

Residuals Statistics[a]

	Minimum	Maximum	Mean	Std. Deviation	N
Predicted Value	25.00	62.22	46.78	7.596	37
Residual	-17.070	20.804	.000	8.719	37
Std. Predicted Value	-2.868	2.032	.000	1.000	37
Std. Residual	-1.817	2.214	.000	.928	37

a. Dependent Variable: Mother CBCL Total Problems T-Score

Hierarchical Multiple Regression Best-Fit Model: Mothers

Descriptive Statistics

	Mean	Std. Deviation	N
Mother CBCL Total Problems T-Score	46.78	11.564	37
Biliary atresia or other severe liver disease	.57	.502	37
Sqrt transformation ratio OPD Time 2	.1859	.05937	37
Mother IFS 15 item total Time 1	36.97	10.938	37

Correlations

		Mother CBCL Total Problems T-Score	Biliary atresia or other severe liver disease	Sqrt transformation ratio OPD Time 2	Mother IFS 15 item total Time 1
Pearson Correlation	Mother CBCL Total Problems T-Score	1.000	-.179	.386	.450
	Biliary atresia or other severe liver disease	-.179	1.000	.368	.266
	Sqrt transformation ratio OPD Time 2	.386	.368	1.000	.516
	Mother IFS 15 item total Time 1	.450	.266	.516	1.000
Sig. (1-tailed)	Mother CBCL Total Problems T-Score	.	.144	.009	.003
	Biliary atresia or other severe liver disease	.144	.	.012	.056
	Sqrt transformation ratio OPD Time 2	.009	.012	.	.001
	Mother IFS 15 item total Time 1	.003	.056	.001	.
N	Mother CBCL Total Problems T-Score	37	37	37	37
	Biliary atresia or other severe liver disease	37	37	37	37
	Sqrt transformation ratio OPD Time 2	37	37	37	37
	Mother IFS 15 item total Time 1	37	37	37	37

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Sqrt transformation ratio OPD Time 2, Biliary atresia or other severe liver disease[b]	.	Enter
2	Mother IFS 15 item total Time 1[b]	.	Enter

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary[c]

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate	Change Statistics					Durbin-Watson
					R Square Change	F Change	df1	df2	Sig. F Change	
1	.518[a]	.269	.225	10.177	.269	6.240	2	34	.005	
2	.613[b]	.376	.319	9.542	.107	5.675	1	33	.023	2.166

a. Predictors: (Constant), Sqrt transformation ratio OPD Time 2, Biliary atresia or other severe liver disease
b. Predictors: (Constant), Sqrt transformation ratio OPD Time 2, Biliary atresia or other severe liver disease, Mother IFS 15 item total Time 1
c. Dependent Variable: Mother CBCL Total Problems T-Score

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	1292.656	2	646.328	6.240	.005[b]
	Residual	3521.614	34	103.577		
	Total	4814.270	36			
2	Regression	1809.398	3	603.133	6.624	.001[c]
	Residual	3004.872	33	91.057		
	Total	4814.270	36			

a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors: (Constant), Sqrt transformation ratio OPD Time 2, Biliary atresia or other severe liver disease
c. Predictors: (Constant), Sqrt transformation ratio OPD Time 2, Biliary atresia or other severe liver disease, Mother IFS 15 item total Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B		Correlations			Collinearity Statistics	
		B	Std. Error	Beta			Lower Bound	Upper Bound	Zero-order	Partial	Part	Tolerance	VIF
1	(Constant)	32.706	5.568		5.873	.000	21.389	44.022					
	Biliary atresia or other severe liver disease	-8.560	3.633	-.372	-2.356	.024	-15.942	-1.177	-.179	-.375	-.346	.864	1.157
	Sqrt transformation ratio OPD Time 2	101.869	30.731	.523	3.315	.002	39.416	164.323	.386	.494	.486	.864	1.157
2	(Constant)	24.856	6.174		4.026	.000	12.294	37.417					
	Biliary atresia or other severe liver disease	-9.336	3.421	-.405	-2.729	.010	-16.297	-2.374	-.179	-.429	-.375	.857	1.167
	Sqrt transformation ratio OPD Time 2	65.680	32.574	.337	2.016	.052	-.592	131.951	.386	.331	.277	.676	1.479
	Mother IFS 15 item total Time 1	.406	.171	.384	2.382	.023	.059	.753	.450	.383	.328	.727	1.375

a. Dependent Variable: Mother CBCL Total Problems T-Score

Excluded Variables[a]

Model		Beta In	t	Sig.	Partial Correlation	Collinearity Statistics		
						Tolerance	VIF	Minimum Tolerance
1	Mother IFS 15 item total Time 1	.384[b]	2.382	.023	.383	.727	1.375	.676

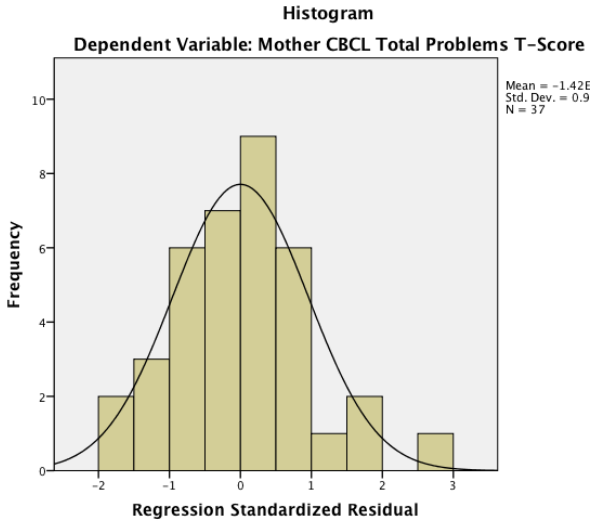
a. Dependent Variable: Mother CBCL Total Problems T-Score
b. Predictors in the Model: (Constant), Sqrt transformation ratio OPD Time 2, Biliary atresia or other severe liver disease

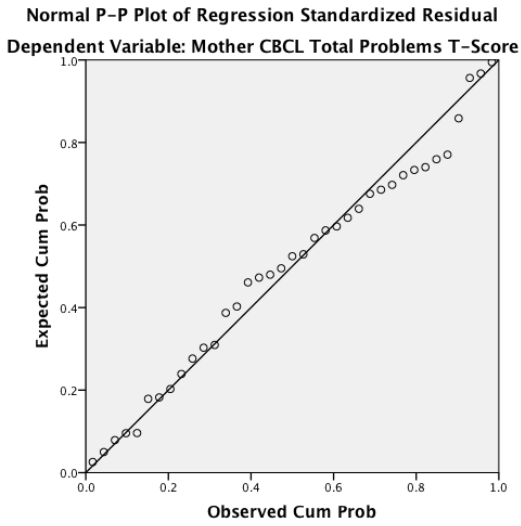
Collinearity Diagnostics[a]

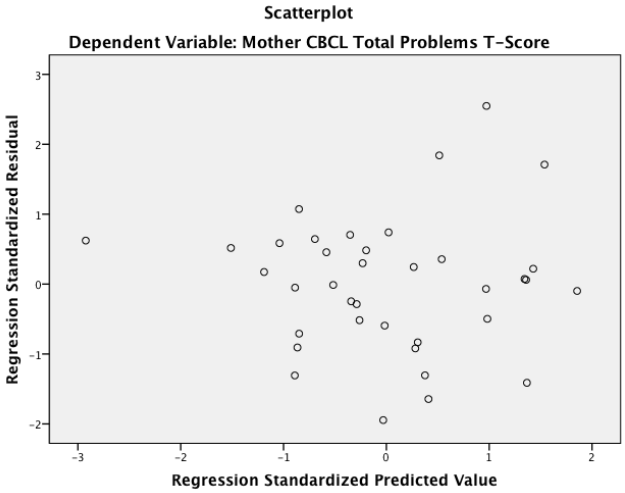
Model	Dimension	Eigenvalue	Condition Index	Variance Proportions			
				(Constant)	Biliary atresia or other severe liver disease	Sqrt transformation ratio OPD Time 2	Mother IFS 15 item total Time 1
1	1	2.669	1.000	.01	.04	.01	
	2	.287	3.049	.06	.90	.03	
	3	.044	7.785	.92	.06	.96	
2	1	3.603	1.000	.00	.02	.00	.00
	2	.315	3.384	.02	.92	.01	.01
	3	.044	9.044	.62	.06	.79	.00
	4	.039	9.666	.36	.00	.20	.98

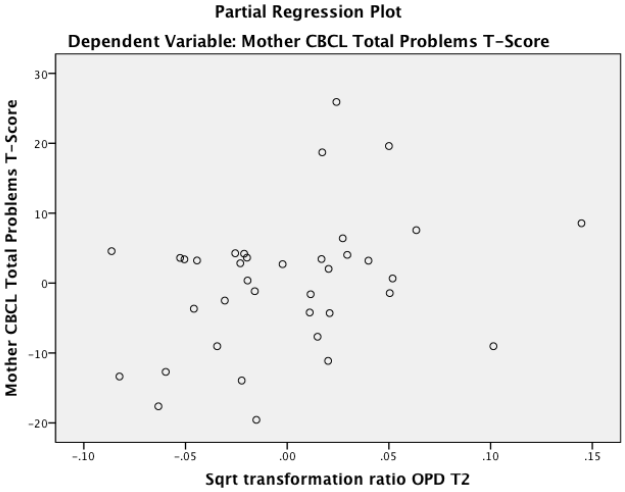
a. Dependent Variable: Mother CBCL Total Problems T-Score

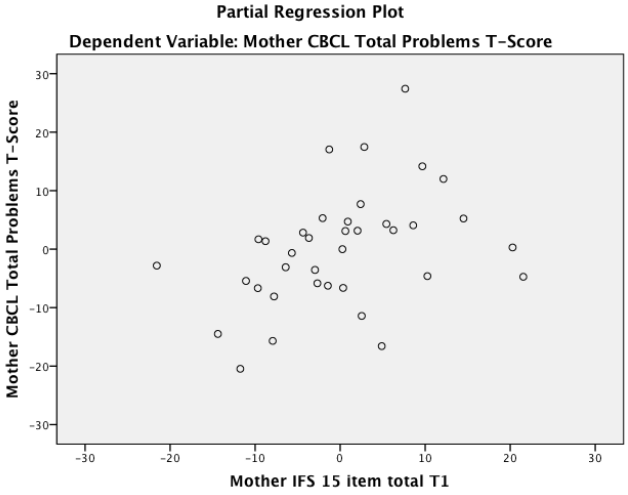
Hierarchical Multiple Regression Best Fit Model: Mothers' Charts











Hierarchical Multiple Regression: Fathers

Significant demographic and illness predictors (Block 1), fathers' IFS scores at Time 1 (Block 2), and fathers' DASS scores at Time 1 (Block 3)

Descriptive Statistics

	Mean	Std. Deviation	N
Father CBCL Total Problems T-Score	46.41	11.800	37
Socio Economic Index	62.305	24.4339	37
Biliary atresia or other severe liver disease	.57	.502	37
Liver transplant at Time 1 or not	.22	.417	37
Father IFS 15 item total Time 1	34.27	8.915	37
Father DASS Total Percentile Time 1	46.35	28.113	37

Correlations

		Father CBCL Total Problems T-Score	Socio Economic Index	Biliary atresia or other severe liver disease	Liver transplant at Time 1 or not	Father IFS 15 item total Time 1	Father DASS Total Percentile Time 1
Pearson Correlation	Father CBCL Total Problems T-Score	1.000	-.420	-.335	.393	.435	.280
	Socio Economic Index	-.420	1.000	.235	-.085	-.115	-.094
	Biliary atresia or other severe liver disease	-.335	.235	1.000	-.072	.157	.021
	Liver transplant at Time 1 or not	.393	-.085	-.072	1.000	.185	.266
	Father IFS 15 item total Time 1	.435	-.115	.157	.185	1.000	.452
	Father DASS Total Percentile Time 1	.280	-.094	.021	.266	.452	1.000
Sig. (1-tailed)	Father CBCL Total Problems T-Score	.	.005	.021	.008	.004	.046
	Socio Economic Index	.005	.	.081	.309	.250	.289
	Biliary atresia or other severe liver disease	.021	.081	.	.337	.177	.451
	Liver transplant at Time 1 or not	.008	.309	.337	.	.136	.056
	Father IFS 15 item total Time 1	.004	.250	.177	.136	.	.003
	Father DASS Total Percentile Time 1	.046	.289	.451	.056	.003	.
N	Father CBCL Total Problems T-Score	37	37	37	37	37	37
	Socio Economic Index	37	37	37	37	37	37
	Biliary atresia or other severe liver disease	37	37	37	37	37	37
	Liver transplant at Time 1 or not	37	37	37	37	37	37
	Father IFS 15 item total Time 1	37	37	37	37	37	37
	Father DASS Total Percentile Time 1	37	37	37	37	37	37

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index[b]	.	Enter
2	Father IFS 15 item total Time 1[b]	.	Enter
3	Father DASS Total Percentile Time 1[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary[d]

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate	Change Statistics					Durbin-Watson
					R Square Change	F Change	df1	df2	Sig. F Change	
1	.596[a]	.356	.297	9.893	.356	6.073	3	33	.002	
2	.710[b]	.504	.442	8.816	.148	9.552	1	32	.004	
3	.710[c]	.504	.424	8.957	.000	.004	1	31	.952	2.481

a. Predictors: (Constant), Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index
b. Predictors: (Constant), Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index, Father IFS 15 item total Time 1
c. Predictors: (Constant), Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index, Father IFS 15 item total Time 1, Father DASS Total Percentile Time 1
d. Dependent Variable: Father CBCL Total Problems T-Score

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	1783.142	3	594.381	6.073	.002[b]
	Residual	3229.776	33	97.872		
	Total	5012.919	36			
2	Regression	2525.582	4	631.395	8.123	.000[c]
	Residual	2487.337	32	77.729		
	Total	5012.919	36			
3	Regression	2525.883	5	505.177	6.297	.000[d]
	Residual	2487.036	31	80.227		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index
c. Predictors: (Constant), Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index, Father IFS 15 item total Time 1
d. Predictors: (Constant), Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index, Father IFS 15 item total Time 1, Father DASS Total Percentile Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B		Correlations			Collinearity Statistics	
		B	Std. Error	Beta			Lower Bound	Upper Bound	Zero-order	Partial	Part	Tolerance	VIF
1	(Constant)	57.471	4.768		12.054	.000	47.770	67.171					
	Socio Economic Index	-.162	.070	-.336	-2.332	.026	-.304	-.021	-.420	-.376	-.326	.940	1.064
	Biliary atresia or other severe liver disease	-5.432	3.382	-.231	-1.606	.118	-12.314	1.450	-.335	-.269	-.224	.942	1.062
	Liver transplant at Time 1 or not	9.851	3.971	.348	2.481	.018	1.772	17.929	.393	.396	.347	.990	1.010
2	(Constant)	39.017	7.328		5.324	.000	24.090	53.945					
	Socio Economic Index	-.134	.063	-.277	-2.134	.041	-.262	-.006	-.420	-.353	-.266	.920	1.087
	Biliary atresia or other severe liver disease	-7.369	3.079	-.314	-2.393	.023	-13.641	-1.098	-.335	-.390	-.298	.903	1.107
	Liver transplant at Time 1 or not	7.717	3.605	.273	2.140	.040	.373	15.061	.393	.354	.267	.954	1.049
	Father IFS 15 item total Time 1	.532	.172	.402	3.091	.004	.181	.883	.435	.479	.385	.916	1.092
3	(Constant)	39.014	7.446		5.240	.000	23.828	54.199					
	Socio Economic Index	-.134	.064	-.277	-2.098	.044	-.264	-.004	-.420	-.353	-.265	.919	1.088
	Biliary atresia or other severe liver disease	-7.364	3.129	-.313	-2.353	.025	-13.746	-.982	-.335	-.389	-.298	.902	1.108
	Liver transplant at Time 1 or not	7.671	3.740	.271	2.051	.049	.043	15.298	.393	.346	.259	.915	1.093
	Father IFS 15 item total Time 1	.527	.193	.398	2.738	.010	.135	.920	.435	.441	.346	.756	1.322
	Father DASS Total Percentile Time 1	.004	.061	.009	.061	.952	-.120	.128	.280	.011	.008	.760	1.316

a. Dependent Variable: Father CBCL Total Problems T-Score

Excluded Variables[a]

Model		Beta In	t	Sig.	Partial Correlation	Collinearity Statistics		
						Tolerance	VIF	Minimum Tolerance
1	Father IFS 15 item total Time 1	.402[b]	3.091	.004	.479	.916	1.092	.903
	Father DASS Total Percentile Time 1	.175[b]	1.209	.235	.209	.921	1.086	.921
2	Father DASS Total Percentile Time 1	.009[c]	.061	.952	.011	.760	1.316	.756

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors in the Model: (Constant), Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index
c. Predictors in the Model: (Constant), Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index, Father IFS 15 item total Time 1

Collinearity Diagnostics[a]

Model	Dimension	Eigenvalue	Condition Index	Variance Proportions					
				(Constant)	Socio Economic Index	Biliary atresia or other severe liver disease	Liver transplant at Time 1 or not	Father IFS 15 item total Time 1	Father DASS Total Percentile Time 1
1	1	2.882	1.000	.01	.01	.04	.03		
	2	.765	1.941	.00	.01	.05	.87		
	3	.288	3.165	.06	.07	.91	.06		
	4	.065	6.658	.92	.91	.00	.04		
2	1	3.792	1.000	.00	.01	.02	.02	.00	
	2	.767	2.223	.00	.00	.04	.86	.00	
	3	.310	3.499	.01	.03	.91	.08	.01	
	4	.107	5.947	.01	.68	.00	.04	.18	
	5	.024	12.524	.97	.28	.02	.01	.80	
3	1	4.581	1.000	.00	.00	.01	.01	.00	.01
	2	.771	2.437	.00	.01	.05	.78	.00	.00
	3	.340	3.668	.00	.00	.78	.16	.01	.09
	4	.200	4.783	.01	.21	.14	.05	.00	.50
	5	.083	7.426	.06	.53	.00	.00	.20	.35
	6	.023	14.003	.92	.24	.02	.00	.79	.04

a. Dependent Variable: Father CBCL Total Problems T-Score

Residuals Statistics[a]

	Minimum	Maximum	Mean	Std. Deviation	N
Predicted Value	31.67	64.59	46.41	8.376	37
Residual	-15.264	14.494	.000	8.312	37
Std. Predicted Value	-1.759	2.171	.000	1.000	37
Std. Residual	-1.704	1.618	.000	.928	37

a. Dependent Variable: Father CBCL Total Problems T-Score

Hierarchical Multiple Regression Best Fit Model: Fathers

Descriptive Statistics

	Mean	Std. Deviation	N
Father CBCL Total Problems T-Score	46.41	11.800	37
Socio Economic Index	62.305	24.4339	37
Biliary atresia or other severe liver disease	.57	.502	37
Liver transplant at Time 1 or not	.2162	.41734	37
Father IFS 15 item total Time 1	34.27	8.915	37

Correlations

		Father CBCL Total Problems T-Score	Socio Economic Index	Biliary atresia or other severe liver disease	Liver transplant at Time 1 or not	Father IFS 15 item total Time 1
Pearson Correlation	Father CBCL Total Problems T-Score	1.000	-.420	-.335	.393	.435
	Socio Economic Index	-.420	1.000	.235	-.085	-.115
	Biliary atresia or other severe liver disease	-.335	.235	1.000	-.072	.157
	Liver transplant at Time 1 or not	.393	-.085	-.072	1.000	.185
	Father IFS 15 item total Time 1	.435	-.115	.157	.185	1.000
Sig. (1-tailed)	Father CBCL Total Problems T-Score	.	.005	.021	.008	.004
	Socio Economic Index	.005	.	.081	.309	.250
	Biliary atresia or other severe liver disease	.021	.081	.	.337	.177
	Liver transplant at Time 1 or not	.008	.309	.337	.	.136
	Father IFS 15 item total Time 1	.004	.250	.177	.136	.
N	Father CBCL Total Problems T-Score	37	37	37	37	37
	Socio Economic Index	37	37	37	37	37
	Biliary atresia or other severe liver disease	37	37	37	37	37
	Liver transplant at Time 1 or not	37	37	37	37	37
	Father IFS 15 item total Time 1	37	37	37	37	37

Variables Entered/Removed[a]

Model	Variables Entered	Variables Removed	Method
1	Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index[b]	.	Enter
2	Father IFS 15 item total Time 1[b]	.	Enter

a. Dependent Variable: Father CBCL Total Problems T-Score
b. All requested variables entered.

Model Summary[c]

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate	Change Statistics					Durbin-Watson
					R Square Change	F Change	df1	df2	Sig. F Change	
1	.596[a]	.356	.297	9.893	.356	6.073	3	33	.002	
2	.710[b]	.504	.442	8.816	.148	9.552	1	32	.004	2.476

a. Predictors: (Constant), Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index
b. Predictors: (Constant), Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index, Father IFS 15 item total Time 1
c. Dependent Variable: Father CBCL Total Problems T-Score

ANOVA[a]

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	1783.142	3	594.381	6.073	.002[b]
	Residual	3229.776	33	97.872		
	Total	5012.919	36			
2	Regression	2525.582	4	631.395	8.123	.000[c]
	Residual	2487.337	32	77.729		
	Total	5012.919	36			

a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors: (Constant), Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index
c. Predictors: (Constant), Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index, Father IFS 15 item total Time 1

Coefficients[a]

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B		Correlations			Collinearity Statistics	
		B	Std. Error	Beta			Lower Bound	Upper Bound	Zero-order	Partial	Part	Tolerance	VIF
1	(Constant)	57.471	4.768		12.054	.000	47.770	67.171					
	Socio Economic Index	-.162	.070	-.336	-2.332	.026	-.304	-.021	-.420	-.376	-.326	.940	1.064
	Biliary atresia or other severe liver disease	-5.432	3.382	-.231	-1.606	.118	-12.314	1.450	-.335	-.269	-.224	.942	1.062
	Liver transplant at Time 1 or not	9.851	3.971	.348	2.481	.018	1.772	17.929	.393	.396	.347	.990	1.010
2	(Constant)	39.017	7.328		5.324	.000	24.090	53.945					
	Socio Economic Index	-.134	.063	-.277	-2.134	.041	-.262	-.006	-.420	-.353	-.266	.920	1.087
	Biliary atresia or other severe liver disease	-7.369	3.079	-.314	-2.393	.023	-13.641	-1.098	-.335	-.390	-.298	.903	1.107
	Liver transplant at Time 1 or not	7.717	3.605	.273	2.140	.040	.373	15.061	.393	.354	.267	.954	1.049
	Father IFS 15 item total Time 1	.532	.172	.402	3.091	.004	.181	.883	.435	.479	.385	.916	1.092

a. Dependent Variable: Father CBCL Total Problems T-Score

Excluded Variables[a]

Model		Beta In	t	Sig.	Partial Correlation	Collinearity Statistics		
						Tolerance	VIF	Minimum Tolerance
1	Father IFS 15 item total Time 1	.402[b]	3.091	.004	.479	.916	1.092	.903

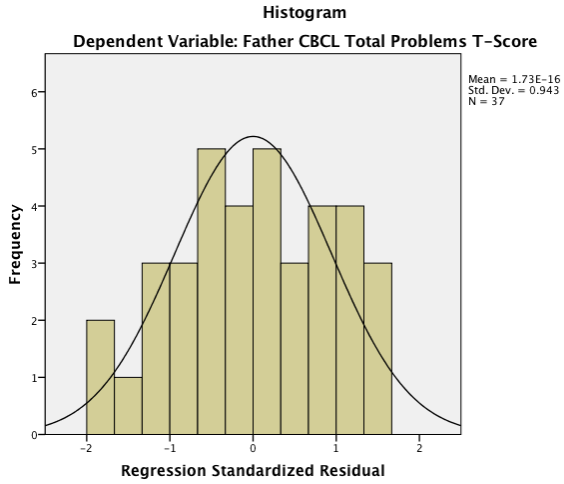
a. Dependent Variable: Father CBCL Total Problems T-Score
b. Predictors in the Model: (Constant), Liver transplant at Time 1 or not, Biliary atresia or other severe liver disease, Socio Economic Index

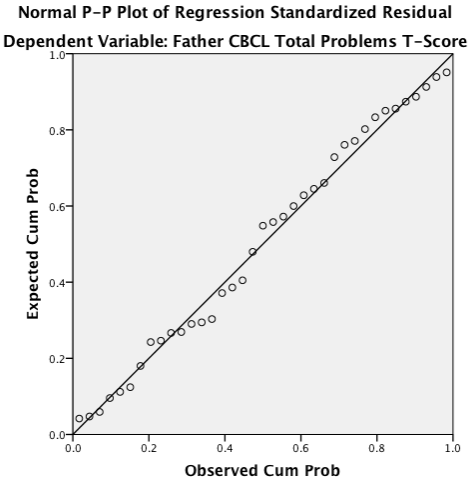
Collinearity Diagnostics[a]

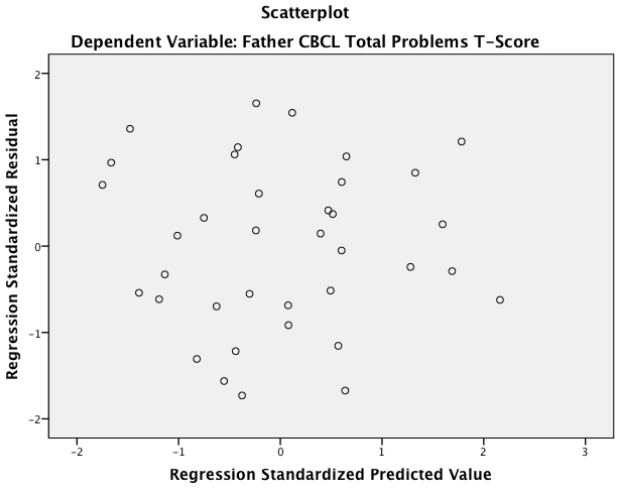
Model	Dimension	Eigenvalue	Condition Index	Variance Proportions				
				(Constant)	Socio Economic Index	Biliary atresia or other severe liver disease	Liver transplant at Time 1 or not	Father IFS 15 item total Time 1
1	1	2.882	1.000	.01	.01	.04	.03	
	2	.765	1.941	.00	.01	.05	.87	
	3	.288	3.165	.06	.07	.91	.06	
	4	.065	6.658	.92	.91	.00	.04	
2	1	3.792	1.000	.00	.01	.02	.02	.00
	2	.767	2.223	.00	.00	.04	.86	.00
	3	.310	3.499	.01	.03	.91	.08	.01
	4	.107	5.947	.01	.68	.00	.04	.18
	5	.024	12.524	.97	.28	.02	.01	.80

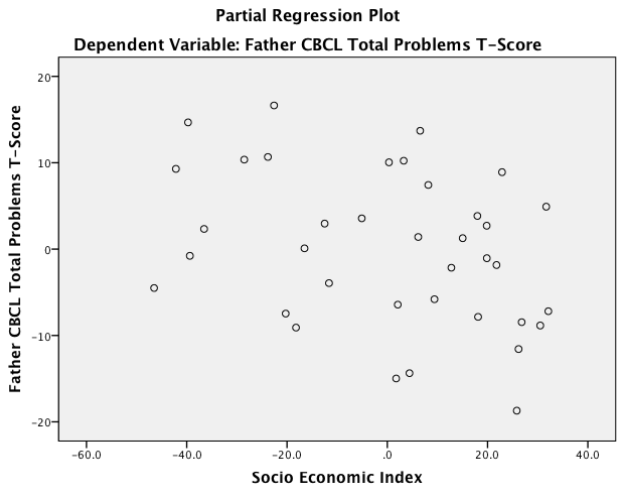
a. Dependent Variable: Father CBCL Total Problems T-Score

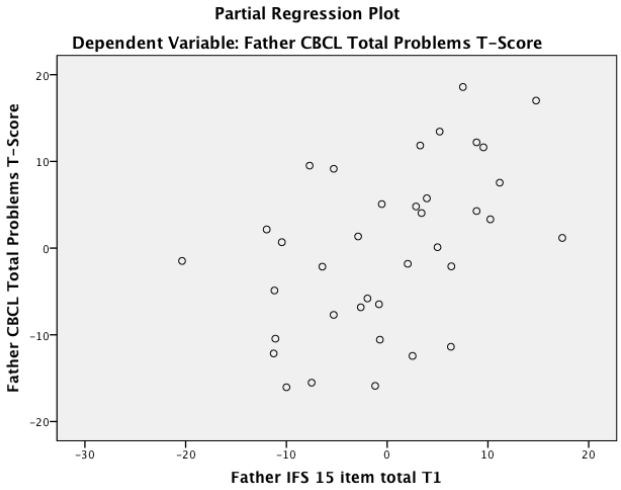
Hierarchical Multiple Regression Best Fit Model: Fathers' Charts











Appendix K
Families Included in the Qualitative Analysis

Parent Names[a]	Infant Name[a]	Infant Gender	Infant Diagnosis	Reason for Inclusion in Qualitative Analysis	Details
Adam and Abigail	Amelia	Female	Biliary Atresia	Parents highly distressed at Time 1	Parents reported they were distressed at Time 1, doing well at Time 2.
Ben and Brooke	Becky	Female	Biliary Atresia	Change in family reports of coping over time	Parents reported they were doing well emotionally at Time 1, father distressed at Time 2.
Charlie and Christine	Chloe	Female	Alagille Syndrome	Marked family disruption	Family made major change to their plans due to infant's illness.
David and Diane	Dani	Female	Biliary Atresia	Well supported family	Family have good social support available. Father reported feeling stressed, mother coping well.
Ewan and Elizabeth	Ebony	Female	Biliary Atresia	Infant's illness severe	Infant was in hospital for an extended period of time. Parents identified multiple additional stressors.
Fraser and Fiona	Felicity	Female	Autoimmune Hepatitis	Family with multiple additional stressors	Parents had two children with the same illness and reported multiple stressors at Time 1, but no additional stressors at Time 2.

Parent Names[a]	Infant Name[a]	Infant Gender	Infant Diagnosis	Reason for Inclusion in Qualitative Analysis	Details
Gary and Gretel	Gordon	Male	Alagille Syndrome	Family bereavement	Family experienced loss of both grandmothers during the study period.
Heath and Hollie	Hayley	Female	Biliary Atresia	Parent mental health treatment	Mother had mental health treatment at Time 1, father had mental health treatment at Time 2.
Ian and Imogen	Isaac	Male	Alpha-1 Antitrypsin Deficiency	Low impact of infant's illness on the family	Parents reporting low impact of the illness on the family: both parents scored below group mean on IFS at both time points.
Jason and Janice	Jasmine	Female	Biliary Atresia	Change in family functioning over time	Parents' scores on FAD changed from both below the group mean at Time 1 to both above the group mean at Time 2.
Kane and Karen	Kristy	Female	Citrullinemia	Stable family functioning over time	Both parents' scores on the FAD were above the group mean at both time points.
Luke and Lucy	Leonard	Male	Biliary Atresia	Difference between parent scores	Mother's DASS scores reduced between Time 1 and Time 2, father's did not. Mother's score on FAD below the mean, father above the mean at both time points.

Parent Names[a]	Infant Name[a]	Infant Gender	Infant Diagnosis	Reason for Inclusion in Qualitative Analysis	Details
Max and Marie	Madeline	Female	Cryptogenic Hepatitis	Sustained high scores for both parents across study measures	Parent scores above the mean on DASS, FAD and IFS at both time points, except father's score on FAD at Time 1. No additional stressors at either time point.
Neil and Nicole	Nick	Male	Biliary Atresia	Difference between scores on different family measures	Parent scores above the group mean on FAD at both time points. Parent scores below the group mean on IFS at both time points.
Orlando and Odette	Olivia	Female	Autoimmune Hepatitis	Change in parent scores over time	Both parents' scores on DASS changed from below the group mean at Time 1 to above the group mean at Time 2.
Peter and Penny	Philip	Male	Biliary Atresia	Difference between parent scores	Mother above the group mean on FAD, father below the group mean at both time points. Both parents above the group mean on IFS both time points.
Richard and Rhonda	Robert	Male	Biliary Atresia	Change in one parent's scores over time; and ensuring adequacy of representation of recruitment hospital	Parents both above group mean on FAD at Time 1, mother above group mean and father below group mean on FAD at Time 2.

Parent Names[a]	Infant Name[a]	Infant Gender	Infant Diagnosis	Reason for Inclusion in Qualitative Analysis	Details
Shannon and Simone	Suzie	Female	Autoimmune Hepatitis	Difference between parent scores	Mother scored CBCL > 1SD below the group mean, while father scored > 1SD above the mean.
Tony and Trish	Tina	Female	Cryptogenic Hepatitis	Agreement in parent scores; and infant characteristics	Both parents scored CBCL > 1SD above the group mean. Infant had emotional problems requiring psychological treatment.
Victor and Valerie	Vivienne	Female	Biliary Atresia	Change in parent scores over time	Parent DASS scores were above the group mean at Time 1 and below the group mean at Time 2.
Warren and Winona	Whitney	Female	Biliary Atresia	Difference between parent scores	Mother scored CBCL above group mean, father scored below group mean. Both parents scored above group mean on IFS at both time points.

Parent Names[a]	Infant Name[a]	Infant Gender	Infant Diagnosis	Reason for Inclusion in Qualitative Analysis	Details
Alan and Alicia	Andrew	Male	Biliary Atresia	Change in parent scores over time; and difference between parent scores	Mother's DASS changed from below the group mean at Time 1 to above the group mean at Time 2. Mother's IFS changed from above the group mean at Time 1 to below the group mean at Time 2. Father's DASS and IFS below the mean at both time points. Difference between parent scores on CBCL: mother above study group mean, father below group mean.
Barry and Barbara	Blake	Male	Alagille Syndrome	Ensuring adequacy of represent-ation	Ensuring adequacy of representation of recruitment hospital and infant gender. Both parents scored DADS below the mean for amount and helpfulness.

Parent Names[a]	Infant Name[a]	Infant Gender	Infant Diagnosis	Reason for Inclusion in Qualitative Analysis	Details
Clarke and Charlotte	Caitlyn	Female	Alpha-1 Antitrypsin Deficiency	Ensuring adequacy of represent-ation	Ensuring adequate representation of parent scores below the mean, infant diagnosis and recruitment hospital. Both parents scored DASS, FAD and IFS below the group mean at both time points. Both parents scored DADS above the mean (greater amount, more helpful).
Darren and Donna	Debbie	Female	Biliary Atresia	New codes identified	New codes identified during final check of non-coded transcripts

[a]All names are pseudonyms

DASS: Depression Anxiety Stress Scales
FAD: Family Assessment Device
IFS: Impact on Family Scale
DADS: Dads' Active Disease Support Scale
CBCL: Child Behavior Checklist

Appendix L
Structure of the codes used in the qualitative analysis

Parent interview transcripts were coded according to the content of the discussion. Codes were organised into related categories.

Category	Codes
Emotions	Negative emotions Positive emotions
External Circumstances	Living issues Stressors Work
Hospital Experience	Infant's progress Diagnosis Father Hospital Illness management Treating team
Infant	Infant's development Infant's personality Infant's response to illness
Physical aspects of the Infant's Illness	Complications Feeding problems Genetics Infection Physical symptoms Urgency
Relationships	External relationships Family relationships Relationships with the infant Relationships and adversity Relationships changing or not Wider family relationships
Response of the Family to the Illness	Change Coping mechanisms Planning Practicalities
Social Support	External support Family support

Appendix M
Characteristics of Families Included in the Qualitative and Integrated Analyses in Comparison with the Total Sample

Questionnaire Responses

Characteristic	Total Sample Time 1 N = 42 (%)[a]	Families in Qualitative Analysis Time 1 N = 25 (%)[a]	Total Sample Time 2 N = 37 (%)[a]	Families in Qualitative Analysis Time 2 N = 25 (%)[a]
Both parents below the mean on the DASS	13 (31)	8 (32)	9 (24)	4 (16)
One parent above the mean on the DASS	15 (36)	8 (32)	17 (46)	13 (52)
Both parents above the mean on the DASS	14 (33)	9 (36)	11 (30)	8 (32)
Both parents below the mean on the FAD	17 (41)	9 (36)	12 (32)	6 (24)
One parent above the mean on the FAD	11 (26)	6 (24)	13 (35)	8 (32)
Both parents above the mean on the FAD	14 (33)	10 (40)	12 (32)	11 (44)
Both parents below the mean on the IFS	13 (31)	7 (28)	14 (39)[b]	9 (38)[c]
One parent above the mean on the IFS	14 (33)	7 (28)	9 (25)[b]	6 (25)[c]
Both parents above the mean on the IFS	15 (36)	11 (44)	13 (36)[b]	9 (38)[c]

[a] Some percentages do not add to 100 due to rounding
[b] N = 36: one mother did not complete the IFS at Time 2
[c] N = 24: one mother did not complete the IFS at Time 2
DASS: Depression, Anxiety, Stress Scales
FAD: Family Assessment Device
IFS: Impact on Family Scale
DADS: Dads' Active Disease Support Scale
CBCL: Child Behavior Checklist

Characteristic	Total Sample Time 1 N = 42 (%)[a]	Families in Qualitative Analysis Time 1 N = 25 (%)[a]	Total Sample Time 2 N = 37 (%)[a]	Families in Qualitative Analysis Time 2 N = 25 (%)[a]
Both parents below the mean on the DADS Amount	14 (33)	10 (40)	14 (38)	12 (48)
One parent above the mean on the DADS Amount	14 (33)	9 (36)	11 (30)	8 (32)
Both parents above the mean on the DADS Amount	14 (33)	6 (24)	12 (32)	5 (20)
Both parents below the mean on the DADS Helpfulness	14 (33)	12 (48)	15 (41)	12 (48)
One parent above the mean on the DADS Helpfulness	17 (41)	9 (36)	13 (35)	8 (32)
Both parents above the mean on the DADS Helpfulness	11 (26)	4 (16)	9 (24)	5 (20)
Both parents below the mean on the CBCL	-	-	15 (41)	12 (48)
One parent above the mean on the CBCL	-	-	9 (24)	6 (24)
Both parents above the mean on the CBCL	-	-	13 (35)	7 (28)

[a] Some percentages do not add to 100 due to rounding
[b] N = 36: one mother did not complete the IFS at Time 2
[c] N = 24: one mother did not complete the IFS at Time 2
DASS: Depression, Anxiety, Stress Scales
FAD: Family Assessment Device
IFS: Impact on Family Scale
DADS: Dads' Active Disease Support Scale
CBCL: Child Behavior Checklist

Additional Stressors and Adequacy of Support

Characteristic	Total Sample Time 1 N = 42 (%)	Families in Qualitative Analysis Time 1 N = 25 (%)	Total Sample Time 2 N = 34[a] (%)	Families in Qualitative Analysis Time 2 N = 25 (%)
At least one additional stressor	30 (71)	18 (72)	25 (74)	20 (80)
No additional stressors	12 (29)	7 (28)	9 (26)	5 (20)
At least one additional stressor at both Time 1 and Time 2	-	-	19 (56)	16 (64)
No additional stressors at both Time 1 and Time 2	-	-	3 (9)	3 (12)
Enough social support	34 (81)	19 (76)	27 (79)	19 (76)
Not enough social support	8 (19)	6 (24)	7 (21)	6 (24)
Enough social support at both Time 1 and Time 2	-	-	22 (65)	16 (64)
Not enough social support at both Time 1 and Time 2	-	-	3 (9)	3 (12)

[a] N = 34: Data were available for the total of 34 families that completed the interview at Time 2