"What about me?"

The impact of cystic fibrosis on parental differential treatment, sibling relationships and adjustment

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ABSTRACT

Cystic fibrosis (CF) is an inherited, multisystem disorder that primarily affects the respiratory and digestive systems. Currently, approximately half the individuals with CF in Australia, the United States and Canada are adults, which represents a remarkable change in the prognosis of this disease over the past 20 years. However, despite recent advances in diagnosis and treatment, CF shortens the life span, and the daily treatment regimen is complex, burdensome and time-consuming, taking 2-4 hours per day. The treatment regimen requires a high level of parental supervision in infancy, childhood and adolescence. The demands CF care places on parents raise the question of how time and attention are allocated when there are other, healthy siblings in the family. The overall objective of this study was to assess the extent of parental differential treatment in two types of families - those with a well child and a child with CF and those with two healthy children. In addition, the impact of differential treatment on the quality of the sibling relationship and the social and emotional adjustment of well siblings in families caring for a child with CF were examined. This study evaluated a cohort of 39 Australian children growing up with a younger sibling with CF and 29 comparison families with similarly aged, healthy children. Information on the amount of time parents spent in daily activities with the younger and older siblings was collected from both mothers and fathers using daily phone diaries. Data were collected from older siblings on a range of social and emotional variables and both younger and older siblings rated the quality of the sibling relationship. This study found evidence of parental differential treatment for fathers but not for mothers. Fathers spent more time with the younger child with CF than the older healthy sibling. High levels of fathers' differential treatment were consistently associated with older healthy siblings indicating a perception of parental bias towards the child with CF. Despite the limitations of cross-sectional research, a major strength of this study was the recruitment of fathers, whose contribution to family life is often neglected in studies of families caring for children with chronic health conditions. Clinical implications and recommendations for future research are discussed.

PREFACE

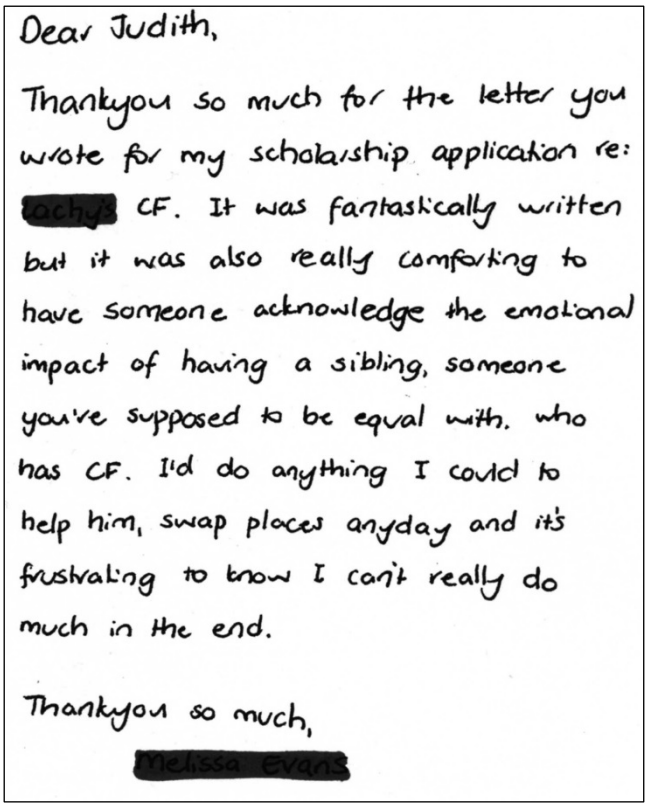
"Sibling relationships are usually complicated and yet also so taken for granted that unsophisticated participants are often unconscious of being caught in a spider's web of love and hate, rivalry and solidarity".

- Iris Murdoch, *The Black Prince*

For over 25 years, I have had the privilege of working with children with cystic fibrosis (CF). When I first started in my role as CF Clinical Nurse Consultant at The Royal Children's Hospital in Melbourne, Australia, I quickly realised that I was working with the family system, including other members of the family who were indirectly affected by this difficult and incurable disease. Family routines were skewed, treatment regimens were often an overwhelming priority and the relationships between parents and well children were strained in ways rarely experienced by other families.

I often found myself in conversations with distressed parents who were concerned about the focus on and time spent on treatments with their child with CF and the guilt they felt about its potentially negative impact on their other children.

My occasional interactions with the siblings of young children with CF highlighted the impact on brothers and sisters. The following letter of thanks I received for writing a reference for the sister of one of our patients provided one such example:



Dear Judith,

Thankyou so much for the letter you wrote for my scholarship application re: [redacted] CF. It was fantastically written but it was also really comforting to have someone acknowledge the emotional impact of having a sibling, someone you've supposed to be equal with, who has CF. I'd do anything I could to help him, swap places anyday and it's frustrating to know I can't really do much in the end.

Thankyou so much,

[redacted]

When I searched for resources and information about the impact of chronic illness, and specifically CF, on siblings, I found a paucity of research (let alone Australian research) and supportive programmes. This research is part of my effort to better understand the experience of CF from the perspective of siblings. I hope that the results of this study will be used to inform the care provided to families affected by CF at The Royal Children's Hospital and contribute to knowledge about the impact of chronic illness on siblings nationally and internationally.

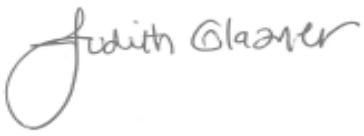
DECLARATION

This is to certify that

The thesis comprises only my original work towards the PhD.

Due acknowledgement has been made in the text of all other material used.

The thesis is less than 100,000 words in length, exclusive of tables, bibliographies, appendices and footnotes.



Judith Anne Glazner

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My gratitude is extended to Belinda Cerritelli for assisting me to set up the sibling database and for her help in determining which families were eligible for the study. I would also like to thank Louisa Salmon for her tireless work collecting the phone diary data.

To my wonderful work colleagues in the Department of Respiratory and Sleep Medicine at The Royal Children's Hospital, in particular Dr Anne-Marie Adams, I would like to thank you for your support, for cheering me on and believing I would get there.

Thank you too to the amazing staff in the J.W. Grieve Memorial Library at The Royal Children's Hospital, especially to Poh Chua for her assistance the whole way.

I would like to gratefully acknowledge the various sources of financial assistance that supported my travel throughout Victoria and NSW in order to collect data from families; that funded the employment of a research assistant to undertake the daily phone diaries; and that enabled me to present these findings at various national and international meetings.

This study would not have been possible without the support of and encouragement from the CF and comparison families, in particular the healthy brothers and sisters of the children with CF, many of whom shared with me how much they appreciated having

a voice through this research. Special thanks to Harry, the delightful young man who detailed a day in his life to assist the reader to understand the treatment burden of living with CF and the caring role of parents

To my childhood friend, Melissa Martin, thank you for your friendship and for teaching me about the relationships siblings share. I miss you and will never forget you.

To my wonderful husband Tony, I hope that you are proud I finally completed this labour of love. In the context of this research, it continues to amuse me that neither of us has siblings!

I would like to acknowledge my two beautiful daughters, Emma and Olivia. I hope that one day you develop a close sibling bond. Always remember how much I love you both and how proud I am of you. Particular thanks to my daughter Emma for her illustrations that have enhanced the many research PowerPoint presentations I have given along the way.

Finally, I dedicate this work to my parents, Pat and Len Glazner, in appreciation for the sacrifices they have made to give me the opportunities I have had, particularly my education.

ABBREVIATIONS

ANOVA: analysis of variance

AREST CF: Australian Respiratory Early Surveillance Team for Cystic Fibrosis

CF: cystic fibrosis

CDI: Child Depression Inventory

CFRD: cystic fibrosis related diabetes

CFTR: cystic fibrosis transmembrane conductance regulator

DPD: Daily Phone Diary

FEV_1: forced expiratory volume in the first second of a forced expiration

IRT: immunoreactive trypsinogen

km: kilometres

NBS: newborn screening

RCH: The Royal Children's Hospital, Melbourne, Australia

SPSS: Statistical Package for the Social Sciences

SRQ: Sibling Relationship Questionnaire

SSRS: Social Skills Rating System

TIDES: The International Depression/Anxiety Epidemiological Study

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PUBLISHED ABSTRACTS, CONFERENCE AND INVITED PRESENTATIONS

International

Glazner JA, Sawyer SM, Reece JE and Quittner AL (2006). What about me? The effect of having a sibling with CF on social and emotional wellbeing. Proceedings of the Twentieth North American Cystic Fibrosis Conference, Pediatric Pulmonology, 41(S29), 405 (poster).

Glazner JA, Sawyer SM and Quittner, AL (2005). Daddy day care or daddies don't care – What is the contribution of fathers in families where a child has CF? Proceedings of the Nineteenth North American Cystic Fibrosis Conference, Pediatric Pulmonology, 40(Suppl 28), 365 (poster and oral paper).

Glazner JA, Sawyer SM and Quittner, AL (2003). The Impact of CF on Sibling Relationships and Family Adjustment: Preliminary Findings from a Controlled Study. Seventeenth Annual North American Cystic Fibrosis Conference, Anaheim (oral paper).

Australasian

2007 Australasian Cystic Fibrosis Conference, Sydney
What about me? The impact of cystic fibrosis on parental differential treatment, sibling relationships and adjustment (oral paper).

2006 Australasian Cystic Fibrosis Nurses' Conference, Auckland
What about me? The effect of having a sibling with CF on social and emotional wellbeing (oral paper).

2005 Australasian Cystic Fibrosis Conference, Adelaide
The impact of cystic fibrosis on sibling relationships and adjustment (oral paper).

2004 Creating Connections – Siblings Australia Inc. First National Conference, Adelaide
The impact of cystic fibrosis on sibling relationships and adjustment: preliminary findings from a controlled study (oral paper).

2004 Australasian Cystic Fibrosis Nurses' Conference, Sydney
The impact of CF on sibling relationships and adjustment: preliminary findings from a controlled study (oral paper).

2003 Australasian Cystic Fibrosis Conference, Melbourne
What is the impact of CF on siblings? Early data from a controlled study (oral paper).

Invited presentations have also been given to the following organisations:

2006 Victorian Genetic Counsellors
Cystic Fibrosis Victoria – Biennial Conference
Australian Confederation of Paediatric and Child Health Nurses

2005 Australian Nursing Federation (Vic Branch) 13th Annual Delegates' Conference

2004 Victorian Visiting Teacher Service (Physically Disabled and Health Impaired)

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2005 Murdoch Children's Research Institute - Trainee Research Scholarship

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2002 Australian Nursing Federation Higher Education Research Award

2000 The Royal Children's Hospital's Travelling Scholarship for General Registered Nurses and Volunteers' Nursing Scholarship

INTRODUCTION

In 1964, social anthropologist Donald Irish drew attention to the scarcity of sibling research, describing it as a "neglected aspect of family life" (Irish, 1964, p. 279). Today, most of Irish's explanations for this deficit are no longer valid. These explanations included: a research focus on adults rather than children; the Freudian emphasis on the importance of infancy on development; and the focus of academics on romantic relationships and marital issues. While it can no longer be claimed that sibling research is a neglected topic, a number of research questions still warrant investigation.
The plan of this introductory chapter is as follows. First, the importance of sibling relationships will be considered. CF will then be described and its diagnosis, treatment and family care contextualised in the Australian setting. What is known about the impact of chronic illness and specifically, CF on sibling relationships and adjustment, and parental differential treatment will be discussed. Methodological approaches used in previous sibling studies will be described, highlighting the limitations of previous research. The chapter will conclude with the aims of this study.

Sibling relationships are one of the most important long-term relationships children have and serve as important models for future interaction with peers (Dunn & McGuire, 1992; Furman & Buhrmester, 1985; Lamb & Sutton-Smith, 2014). Siblings have intense and long-standing relationships that differ considerably from parent-child relationships. Healthy siblings must, at times, act as teachers, comforters, and protectors. Siblings generally develop a deep bond and experience a range of feelings towards their siblings, from anger and embarrassment to love and loyalty (Trahd, 1986). By living together, siblings learn strategies for conflict resolution and have significant effects on each other's development. The quality of the sibling relationship can affect a child's feelings of competence and attractiveness and contributes to their overall adjustment in later years (Dunn, Slomkowski, Bcardsall, & Rende, 1994). All siblings experience intense emotions including love, envy, empathy, and companionship; these are likely to be exaggerated when one sibling has a chronic illness such as CF (Eiser, 1993; Vermaes, van Susante, & van Bakel, 2012).

What is cystic fibrosis?

Dr Dorothy Anderson first described cystic fibrosis of the pancreas in the medical literature in 1938 and it was subsequently associated with respiratory infections and salt loss during a heat wave in New York (Davis, 2006). An ancient folk saying 'Woe to that child which when kissed on the forehead tastes salty. He is bewitched and soon must die' suggests that CF has existed since the Middle Ages (Orenstein, Rosenstein, & Stern, 2000).

CF is now appreciated as the most common life-limiting genetic disorder in Europe, North America, and Australia with a worldwide prevalence of 1 in 2500 live births (Elborn, 2016). In 2014, the most recent year for which data is available, the Australian CF Data Registry held records of 3,294 people with CF (Cystic Fibrosis Australia, 2016), while approximately 70,000 people live with CF across the world (Cutting, 2015).

An autosomal recessive disorder, CF is caused by mutation in a gene that encodes a chloride-conducting transmembrane channel called the cystic fibrosis transmembrane conductance regulator (CFTR). Dysfunction of CFTR mainly affects epithelial cells and causes problems with mucociliary clearance, chronic respiratory infections and gradual loss of lung function. CF affects multiple organ systems. In addition to the life-limiting impact on the respiratory system, epithelial cell dysfunction leads to comorbidities in the pancreas (causing malabsorption of fat-containing foods), liver (leading to cirrhosis), sweat glands (salt loss) and vas deferens resulting in infertility in males (Elborn, 2016). The disease phenotype varies in its features, age at onset, severity, and rate of progression (Zielenski, 2000).

CF is most commonly found in populations with northern European ancestry where the predominant mutation, p.F508del, was identified over 25 years ago (Kerem et al., 1989; Riordan et al., 1989; Rommens et al., 1989). Over 2000 variations of the CF gene have been identified since, although it is not clear that they all cause disease (Quon & Rowe, 2016). These gene mutations have different effects on the production and function of CFTR, and its stability at the cell membrane (Bell, De Boeck, & Amaral, 2015).

In the 1950s and 1960s, clinical treatment programmes began to change CF from a devastating condition with a life expectancy of less than one year to a severe chronic disorder affecting children and adults (Orenstein et al., 2000). Despite still being incurable, the prognosis for people living with CF continues to improve. This has largely been due to earlier diagnosis through newborn screening programmes, a more aggressive approach to the detection and treatment of lung inflammation and infection, and the provision of care by multidisciplinary teams in specialised centres. More recently, both gene correctors and potentiators have been approved for individuals with CF depending on their primary genetic mutations (Davies et al., 2016; Davies et al., 2013; Ramsey et al., 2011; Wainwright et al., 2015); data on the efficacy of these new medications suggests that they will significantly reduce morbidity and decrease mortality. Median life expectancy has increased dramatically in the past 20 years with a median projected lifespan in the late 40s and early 50s (Burgel et al., 2015).

Cystic fibrosis health care in the 21st century

Newborn screening (NBS) for CF facilitates early diagnosis and access to genetic counselling for parents of affected infants (Massie, Curnow, Gaffney, Carlin, & Francis, 2010). NBS has been demonstrated to reduce disease severity as well as the cost and burden of CF care (Sims et al., 2007). Various NBS programmes for CF have now been implemented in a majority of countries that have a high prevalence of the disease (Mayell et al., 2009), although screening protocols differ by country.

In Victoria, Australia, NBS for CF was introduced in 1989. The Victorian paradigm for NBS is shown in Figure 1 (Curnow, 2017). All infants born in Victoria have a heel prick test on day 2-4 of life and are initially screened for elevated levels of serum trypsinogen in the blood by immunoreactive assay (IRT). In the early years of NBS in Victoria, a second IRT was requested at 4-6 weeks if the initial value was high and the diagnosis was then confirmed by a sweat test. The sweat test remains the gold standard diagnostic test for CF (Montgomery & Howenstine, 2009) and involves the measurement of chloride and sodium in sweat (Gibson & Cooke, 1959). Since 1991,

gene mutation analysis has been incorporated into the NBS programme. While initially only the most common CFTR gene mutation was assessed, p.F508del, since 2017, the Victorian NBS programme has included a panel of 38 gene mutations. Diagnosis is usually confirmed by the time the baby is 4-6 weeks old. An important exception is infants with meconium ileus, an intestinal obstruction that is largely a complication of CF which presents soon after birth, before the results of NBS are available.

Parents recall the period around the CF diagnosis as full of emotions and difficult thoughts (Havermans, Tack, Vertommen, Proesmans, & de Boeck, 2015). In a questionnaire study of parents of children with CF, de Monestrol (2011) found that parental experiences on receiving a CF diagnosis are intense and emotional with most parents experiencing anxiety or fear. Similarly in a postal survey of parents, Jedlicka-Kohler, Gotz, and Eichler (1996) found that the most frequent feelings at the time of diagnosis were fear and despair. Havermans et al. (2015) stressed the importance of CF teams tailoring the provision of the diagnosis to parents' needs, as it is the starting point of a long-term relationship.

The implementation of NBS in Australia enables a focus on preventative treatment of infants and children with CF. This has led to a paradigm shift from reactive treatment to proactive early disease surveillance and early therapeutic intervention (Branch-Smith, 2016). It also means that from early infancy when the diagnosis is made, parents are focused on the health of their child with CF, with the potential for wider impacts within the family system, including siblings.

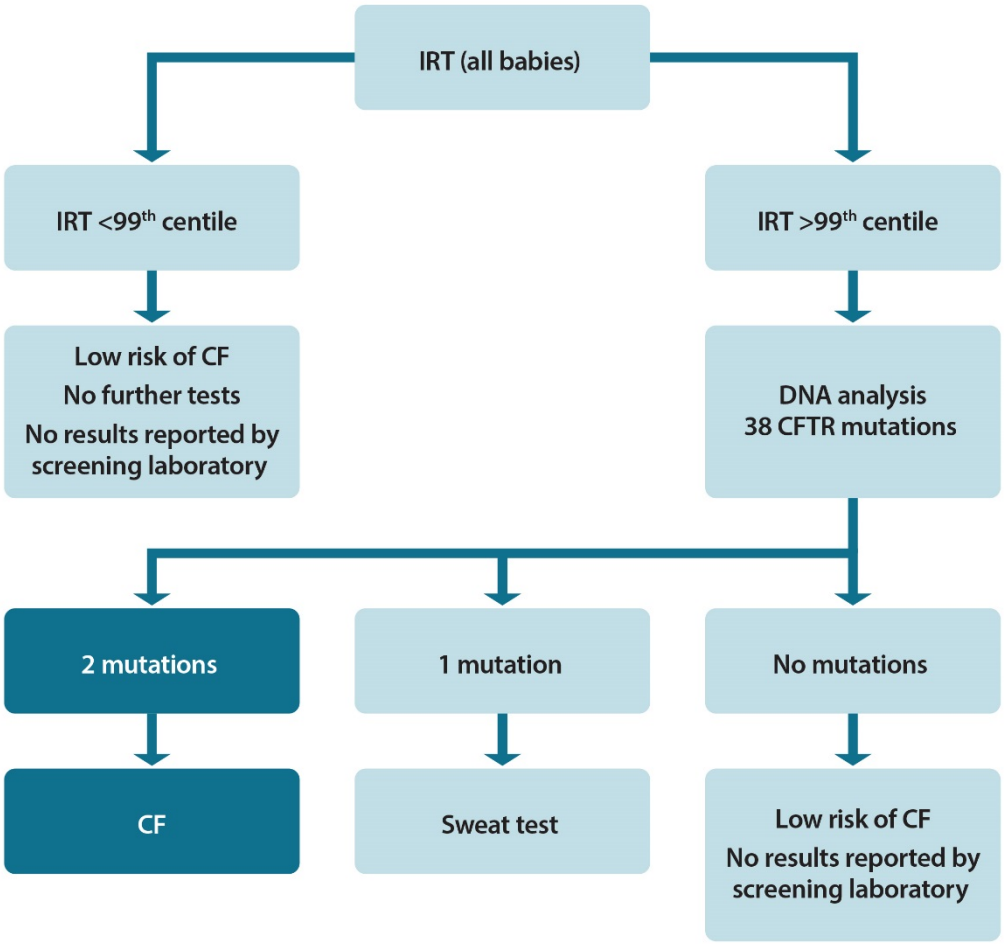

Figure 1. Victorian CF Newborn Screening Paradigm.

The aim of CF management is to improve the duration and quality of life for individuals who have the disease. In Australia, as in other developed countries, the clinical care of individuals with CF is provided by specialist multidisciplinary teams with the experience and expertise to ensure that the disease is well managed (Kerem & Webb, 2014). The development of national and international standards of care and clinical guidelines have enabled CF centres to develop standards to benchmark performance and outcomes, through quality improvement initiatives (Elborn, 2016). Until recently, CF guidelines have been largely medically focused, with less consideration of the psychosocial aspects of CF care. Although national standards of care advocate for a psychologist and social worker as part of the multidisciplinary team (Bell, Robinson, &

Fitzgerald, 2008), national consensus on how to assess the psychological health of patients and caregivers has only recently been addressed through the work of the International Committee on Mental Health in Cystic Fibrosis (Quittner et al., 2016). In line with these recommendations, the Australian and New Zealand CF Psychologists' Network is contributing a chapter on the standards of care for CF psychologists to the updated national standards of care.

The high level of transmissibility of respiratory pathogens in people with CF, such as *Pseudomonas aeruginosa* and *Mycobacterium abscessus* coupled with poor clinical outcomes associated with these bacterium, have led CF peak bodies and CF centres to implement stringent infection control guidelines in community, clinic and inpatient environments (Knibbs et al., 2014). A negative consequence of this essential clinical initiative has been the loss of informal and formal peer support that had previously been a feature of interactions between patients and families in various settings. Thus, infection control guidelines have reduced opportunities for CF families to socialise together.

Treatment interventions for CF place particular demands on parents, especially those of younger children. A daily treatment regimen is required due to the effects of abnormally thick, sticky secretions in the lungs. These secretions block small airways and can trap infection-causing bacteria. An important and arduous part of the daily treatment regimen is airway clearance or chest physiotherapy. In infants and young children this is performed by their parents. Older school-age children are taught independent physiotherapy techniques so that they can participate in activities such as school camps or sleep overs without their parents needing to be physically present. In addition to airway clearance, inhaled mucolytics such as Pulmozyme® and hypertonic saline are used to render the secretions less tenacious, making them easier to expectorate. Inhaled or oral antibiotics are used to prevent and treat infection.

In the digestive system, thick secretions block the passage of digestive enzymes into the small intestine, causing malabsorption. This requires the administration of pancreatic enzymes immediately prior to fat-containing meals and snacks. Newly diagnosed

infants are administered enzyme granules mixed with a small amount of apple puree. This is one of the first challenges faced by new parents, as they often comment about giving their baby “solids” at such an early age and need to establish this as part of their feeding routine. Children are often as young as two or three when they master the art of swallowing enzyme capsules. It is important to teach children with CF to do this prior to the commencement of school, as the ingestion of capsules is much easier for the classroom teacher to supervise and its absorption is also better than enzyme granules. Adolescents are often self-conscious about appearing different from their peers and need reminders to take their enzymes at school. In addition to taking pancreatic enzymes, people with CF require a high energy diet due to the increased metabolic rate associated with respiratory infections and the malabsorption of fat-containing foods.

The dietitian in the CF team has an important role in educating parents about the nutritional management of their child with CF. In particular, parents need to gain a strong understanding of food groups (ie which food groups require pancreatic enzymes) and about the energy content of different food groups (ie providing high energy, high fat but nutritious foods). As children mature, they must also learn about their nutritional and enzyme requirements. The knowledge that body weight influences lung health results in parents becoming appropriately vigilant around maximising an energy-rich diet. While tensions around food and eating are a normal aspect of ordinary family life, these tensions can become much more problematic in families of children with CF at an early age. This is made even more challenging because healthy siblings are not required to eat the “special” energy-rich foods that are encouraged for children with CF such as chips and chocolate. In time it is likely that these nutritional recommendations will change with growing data on acquisition of CF-related diabetes in older teens and young adults (Moran, Pillay, Becker, & Acerini, 2014). However, early on, these difference in availability of “treats” can lead to feelings of envy in healthy siblings and concerns about differential treatment for parents.

Mealtime is one of the most frequently cited problems by families of children with CF. For example, in a study by Crist et al. (1994) parents reported that their children with CF took a long time to finish meals, delayed eating by talking and spat out food. The

parents of children with CF reported that they engaged in higher rates of ineffective mealtime strategies, such as coaxing their children to eat or making a second meal, as compared to parents of children without CF. A further example is the study by Stark et al. (2000) who found that while parents of children with CF engaged in similar mealtime management strategies to those used by parents of children without CF, they differed in that they kept their children longer at the table.

The daily treatment regimen for CF has been estimated to take between 2 to 4 hours (Sawicki et al., 2011). For young children, the burden of this care falls solely on parents. As the young person with CF matures, they require support from their family and the CF team to develop the knowledge, attitudes and skills that promote effective self-management of their disease (Sawyer & Aroni, 2005). At the same time parents are encouraged to gradually reduce the extent of direct involvement in their child's care, even though they are expected to remain actively engaged in a more supervisory capacity. In this way, parent responsibilities change from a direct or "hands on" caregiver role during infancy, to a more supervisory role in adolescence, to one that in time becomes more supportive of adults with CF. In addition to these changes in parent-child roles and responsibilities that change with age, the CF treatment regimen also changes according to pulmonary exacerbation events, and the onset of new CF complications (for example, CF-related diabetes (CFRD)) and life transitions (for example, commencement of primary and secondary school), as shown in Figure 2.

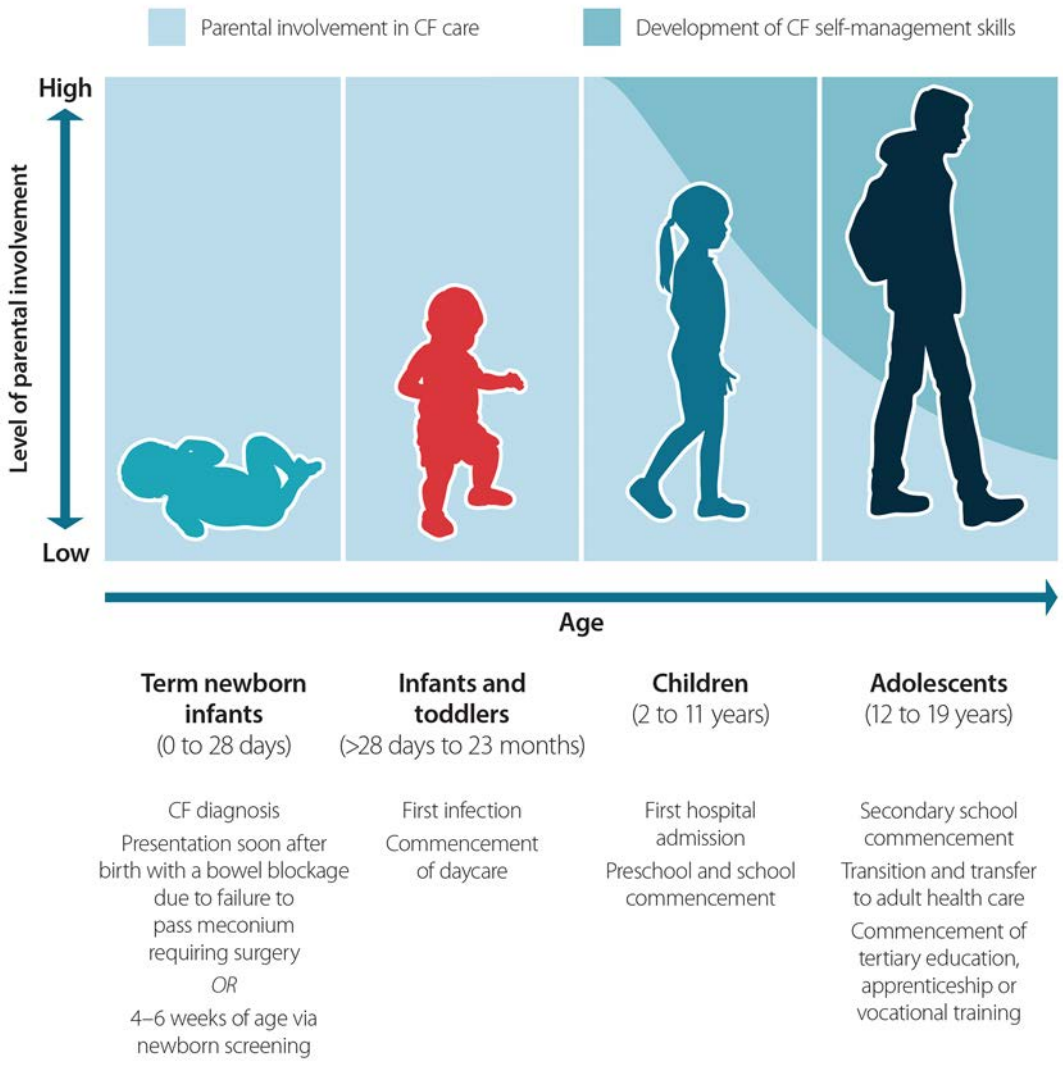

Figure 2. Major transitions and influences on CF care from birth to adolescence.

The example of Harry, a 13 year old boy with CF, shown in Figure 3, exemplifies the daily burden of care that is required, especially for older children and adolescents with CF. It also highlights that parents are still intimately involved in monitoring their child's health. Beyond the immediate supervisory and treatment monitoring roles, parents are also required to prepare feeds, put out medication for children to take, arrange a continuous supply of medication to be available (for example, prescription management), take children to their medical outpatient appointments and spend time with them in hospital when admission is required. Thus, a focus on daily treatment of

the child with CF continues to be a feature of family life which is experienced, one way or another, by all family members, including siblings.

The development of CF complications can change and challenge established roles within the family, at least in the short term, which brings renewed focus to the child with CF. For example, CFRD is the most common co-morbidity associated with CF. While it can occur at any age, its prevalence increases with age, especially during adolescence (Moran et al., 2014). All CF patients over the age of 10 years who do not have CFRD are screened annually using an oral glucose tolerance test. Insulin therapy is the mainstay of treatment and it is recommended that blood glucose levels are monitored four times a day. The care required for CFRD adds to the daily established treatment burden of CF care for patients and is often perceived by young people as yet "another" diagnosis to deal with. It is an emotional time that is often associated with feelings of shock and uncertainty (Collins & Reynolds, 2008).

The combination of early diagnosis of CF from NBS and the many advances made in the care of children, now results in most adolescents transferring to adult care in good physical condition with a good quality of life (Duff & Oxley, 2016). Contemporary cohorts of adults now constitute the majority of the CF population in Australia, Canada, the United States and the United Kingdom. Many have full-time or part-time jobs, are completing further education and have partners and children of their own. While increasing numbers of people with CF are reaching adulthood with milder disease, this is often coupled with a complex and time-consuming daily treatment regimen, where optimal levels of adherence compete with the activities of daily living (Duff & Oxley, 2016; Modi et al., 2010; Quittner, Zhang, et al., 2014). These changing roles can become more conflictual in the context of poor adherence with treatment regimens, which are a feature of all chronic conditions including CF (Barker & Quittner, 2016; Quittner, Zhang, et al., 2014). These more negative aspects of family life (for example, when parents argue with their child with CF about remembering to take medication or engage in airway clearance) can also be experienced by all family members including siblings. Notwithstanding the tension and even conflictual emotions that can

characterise parent-child communication around treatment adherence, the focus of parent attention is still on the child with CF rather than the well sibling.

Due to the chronic, progressive nature of CF, adults in the 21st century can experience longer periods of poor health and hospitalisation than their paediatric counterparts, and have to address the reality of declining health and the complications associated with living longer. Growing older with CF brings with it new challenges of juggling declining health and the increased time required for CF care, work and family life.

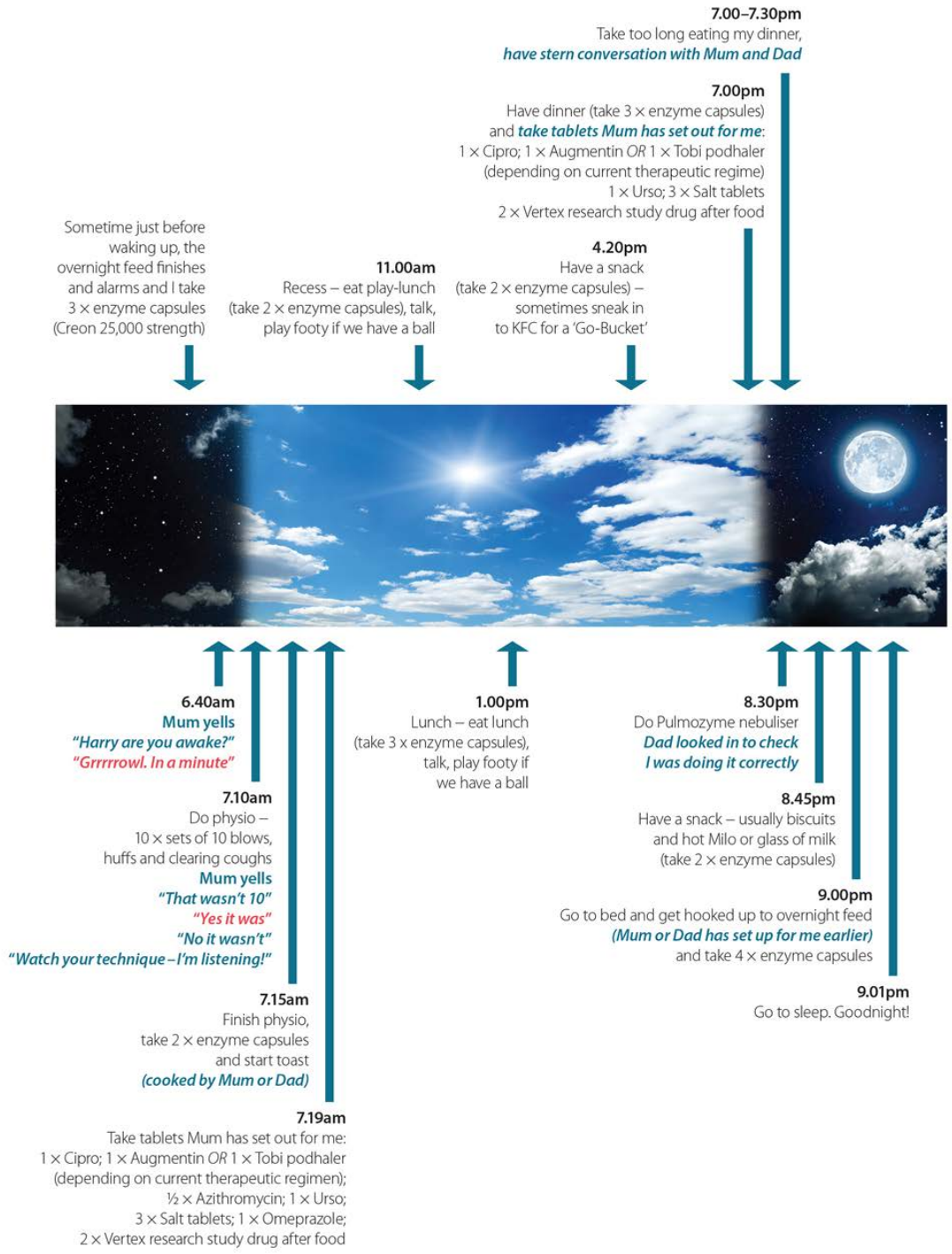

Figure 3. A day in the life of Harry, a 13 year old boy with CF

Note: diary provided for the purpose of this thesis, chosen as his mother was a member of the CF Family Advisory Committee.

New therapies for cystic fibrosis

Until recently, CF treatments had been aimed at managing the complications of defective CFTR. This included pancreatic enzyme replacement therapy, using antibiotics to treat respiratory infections, and chest physiotherapy to help clear the tenacious sputum produced by people with CF (Pittman & Ferkol, 2015). Gradually, more CF-specific treatments such as inhaled recombinant human DNase (Pulmozyme®) and TOBI® (inhaled Tobramycin) have been developed, although as Shanthikumar and Massie point out (Shanthikumar & Massie, 2017), these treatments still do not address the problem of altered CFTR function. These treatments have slowed down the inevitable decline in lung function but also add to the treatment burden of people with CF (Sawicki, Sellers, & Robinson, 2009).

There is great interest in treatments that target the restoration of CFTR function, as these could potentially correct the underlying defect, arrest lung function decline and potentially reduce treatment burden. Innovative and transformational therapies that target the basic defect in CF have recently been developed, such as Kalydeco® (Davies et al., 2016; Davies et al., 2013; Ramsey et al., 2011) and Orkambi® (Wainwright et al., 2015) and have been shown to improve lung function and reduce respiratory exacerbations. Further small molecule and gene-based therapies are being developed to restore CFTR function, which promise further to improve lung function in people with CF (Elborn, 2016).

Gene therapy is another novel approach to restoring CFTR function. This involves delivering artificial CFTR to the lungs, either by inhalation or stem cell transplantation. The major benefit of gene therapy is that it would help all patients regardless of CFTR genotype. However, a limitation of inhaled gene therapy is that it would not target the non-pulmonary complications of CF, such as pancreatic defects (Shanthikumar & Massie, 2017). While there is ongoing gene therapy research being conducted, it is not routine clinical practice.

As CF is a chronic, progressive disease, the effects change with age, whether from biological, developmental or emotional perspectives. Recent studies conducted by the

Australian Respiratory Early Surveillance Team for Cystic Fibrosis (AREST CF) has found that, by as early as 3 years of age, almost a third of children with CF have evidence of inflammation and irreversible lung damage (Sly et al., 2013). This research has implications for new CF therapies, which would need to be commenced as early as possible after diagnosis.

Researchers are working to cure CF. Although there is debate as to what exactly would constitute a cure for CF, it is generally accepted that a cure would lead to patients with CF having normal lung structure and function (Shanthikumar & Massie, 2017). The hope is that all people living with CF will eventually have access to CFTR restorative treatments and that these therapies are initiated as early as possible.

The psychological effects of living with cystic fibrosis

As a result of the multitude of improvements in health care, including the more effective medications described in the previous section, there has been a remarkable improvement in the health, quality of life, and median survival of patients with CF. Children born with CF in the 21st century are expected to live into their mid-50s (Burgel et al., 2015; Dodge, Lewis, Stanton, & Wilsher, 2007). Consequently, for an increasing majority of children and adolescents with CF and their parents, research into their psychosocial wellbeing that was undertaken before the millennium is likely to be somewhat redundant (Duff & Oxley, 2016). Improvements in CF management and health outcomes, including NBS and a much more complex suite of clinical interventions, have also meant that the landscape of parents' experiences of the condition have changed (Branch-Smith, 2016). Now that CF is no longer considered a fatal disease of childhood (as it was so often described in the past), the experiences of parents have shifted from a necessary focus on treatments to reduce mortality to the recognition that the increase in treatments places a consequent burden on caregivers (Bregnballe, Schiøtz, Boisen, Pressler, & Thastum, 2011; Ziaian et al., 2006).

After the diagnosis of CF, parents must learn about an unfamiliar and often frightening condition at a time when they are still getting to know their new baby. This requires

adjustment in parenting roles and plans for the future (Götz & Götz, 2000; Quittner et al., 1998). When the baby with CF is the first born child, couples are adjusting not only to the diagnosis but also to an unfamiliar new role as parents. Parents are generally able to establish a family routine that includes CF care and management. Medical activities such as performing airway clearance or administering medication to a child may initially feel unnatural and awkward to parents, and the parenting role may become confused and more medicalised during these early years (Madge, Francis, & Bilton, 2016).

As CF does not impact intellectual functioning, parents are encouraged to have normal social, emotional, and cognitive expectations for their affected children (Quittner & Opipari, 1994). This is often easier said than practised, as the tendency for many parents is to "wrap the child in cotton wool" to protect them, especially given the focus of CF clinics on the prevention of respiratory infections. In my own clinical experience, I often worry that parents alienate their family and friends due to the stringent house rules that a number of them impose on visitors, for example.

The complex challenges required of parents to manage their children's CF treatment increases parental stress and has been associated with depression, poor sleep quality and relationship strain in parents of young children with CF (Glasscoe, Lancaster, Smyth, & Hill, 2007; Yilmaz et al., 2008). The International Depression Epidemiological Study (TIDES) evaluated the prevalence of anxiety and depression in parent caregivers of children and adolescents with CF who were recruited from 154 CF Centres across nine European countries and the United States (Quittner, Goldbeck, et al., 2014). Parents of children and adolescents up to 18 years of age completed standardised measures of depression and anxiety. Elevated symptoms of depression were found in 37% of mothers and 31% of fathers, while elevated levels of anxiety were found in 48% of mothers and 36% of fathers. The same study used standardised measures of depression and anxiety in adolescents aged 12 years and over and adults with CF. Elevated symptoms of depression were found in 10% of adolescents and 19% of adults, while elevated symptoms of anxiety were found in 22% of adolescents and 32% of adults with CF. Symptoms of common psychological distress are therefore highly prevalent in

parents, as well as adolescents and adults with CF. In adults with CF, depression and anxiety are associated with worse self-management, pulmonary function, quality of life, increased hospitalisations and greater healthcare costs (Smith, Modi, Quittner, & Wood, 2010; Snell, Fernandes, Bujoreanu, & Garcia, 2014). In general, the relationships of depressed mothers with their offspring are marked by a greater likelihood of maternal over-intrusiveness, emotional withdrawal and a general failure to sensitively engage (Murray, Hipwell, Hooper, Stein, & Cooper, 1996). It is similarly anticipated that elevated symptoms of emotional distress and common mental disorder in parents of children with CF would also effect parent-child interactions. For example, a study of mothers of children newly diagnosed with CF showed that in addition to increased parenting stress, greater difficulty was reported around their caregiver roles (Quittner, DiGirolamo, Michel, & Eigen, 1992).

Fathers and chronic illness

Parenting a child with a chronic health condition includes a unique set of challenges for both mothers and fathers throughout all phases of the illness and across the lifespan. However, fathers of children with chronic health conditions have been under-represented in research (Goldstein, Akre, Belanger, & Suris, 2013). A small number of studies have attempted to describe fathers' adjustment to a diagnosis of paediatric chronic illness, but even fewer studies have examined how paternal adjustment relates to child adjustment (Bennett Murphy, Flowers, McNamara, & Young-Saleme, 2008). The majority of existing studies have focused on the fathers of children with a cancer diagnosis.

Sloper (2000) studied 58 fathers and 68 mothers of children with cancer. They found that while both mothers and fathers had high levels of distress, different coping and appraisal factors were related to adjustment for each group. Perceived strain of the illness and ability to manage it, together with family cohesion, were the predictors of maternal distress. Fathers' adjustment was predicted by employment problems, hospital admissions, and family cohesion. In other words, there were different results for mothers and fathers, which reinforces the importance of understanding paternal as well

as maternal experiences. While it was pleasing that this study explored both fathers and mothers, it did not examine the relationship between these variables on child adjustment.

Research suggests that there are gender differences in the ways parents respond to the diagnosis of paediatric chronic illness. Notwithstanding changes in gender roles within families that have taken place over the past few decades in Australia as more women work full time, my experience with families looking after children with CF is that mothers still tend to immerse themselves in the illness and take primary responsibility for treatment-related demands, such as clinic visits, administration of daily treatments and hospitalisations, as described over 25 years ago (Madan-Swain & Brown, 1991). Most fathers continue working to support the family financially (Quittner et al., 1998; Reay, Bignold, Ball, & Cribb, 1998).

While researchers such as Quittner et al. (1998) conceptualise fathers' work as an economic imperative, others suggest that paternal employment is also related to stress avoidance and the need to maintain control (Cayse, 1994). If fathers need to appear strong, calm and in control, this could have detrimental consequences on their well-being (Reay et al., 1998). On the other hand, data reported in the paediatric cancer literature also suggests that fathers maintain some distance as a protective factor, reducing their levels of stress and emotional distress (Noll et al., 1995). Fathers, like mothers, can experience stress (Calzada, Eyberg, Rich, & Querido, 2004) and can find it difficult to discuss their family situation (Goble, 2004). This can compound their own expectations that they should be "strong and silent" to portray strength within the family. In order to appear strong, fathers may limit their involvement in an attempt to maintain control. These issues could also expect to operate in families of children with CF, given declining lung function with increasing age, greater likelihood of hospitalisation with age and the onset of new comorbidities such as CFRD, which may be sources of anxiety for fathers. However, we also know that contemporary fathers are commonly highly engaged in family life. Thus, the extent to which these data extend to contemporary families with CF is unknown.

Bennett Murphy et al. (2008) examined the role of fathers in caring for children with cancer. Psychological adjustment, coping, and work patterns of twenty mothers and fathers were investigated. The study included a comparison group of twenty fathers of healthy children. They found that fathers did not differ from mothers in the cancer group, or fathers in the comparison group, in terms of psychological adjustment or coping. However, fathers of children with cancer spent more hours at work and more hours caring for other children than fathers in the comparison group. The fathers of children with cancer made a significant contribution to child care (4.5 hours a day, on average). Although there was considerable variability between study fathers, most fathers participated in getting the child to a clinic or hospital, helped siblings get to scheduled activities, and assisted with housework. Expensive medical bills associated with the United States healthcare system were a likely contributor to the long hours worked by the fathers in this study. Mothers in this study spent fewer hours in paid employment and spent most of their time with their children. One of the strengths of this study is the inclusion of fathers, but it is limited by a small sample size. The extent of bias in recruitment is also unknown. Fathers who spent more time with their families may have been more likely to participate in this study, raising questions about the generalisability of these data.

In one of the few Australian studies in this area, a qualitative analysis by Peck and Lillibridge (2005) of four fathers of children with various chronic illnesses found that fathers used optimism about expectations of their child's achievements as a way to help them to manage the emotional turmoil they initially experienced following the diagnosis. This approach helped them to gain a sense of normality in their daily lives rather than thinking about future possibilities of their child's deteriorating health. This study is also limited by the small sample size which would have precluded thematic saturation.

Ware and Raval (2007) also utilised qualitative methods to investigate eight fathers who had a child with a life-limiting illness. All of the fathers relayed a heightened feeling of love for their sick child, wanting to make the most of their time with them, and enjoying their relationship. Participants acknowledged that balancing the conflicting needs of all

their children was difficult, especially as the needs of the sick child often took precedence. Similarly McGrath and Huff (2003), in an Australian study of six fathers of young children with leukaemia, found that fathers acknowledged the tension between wanting to spend time with all of their children whilst recognising the increased needs of their unwell child. The fathers in this study talked about caring for the well children at home as an alienating experience, as they longed to be with the ill child in hospital. They also acknowledged the disruption to the well siblings' normal routines, the need for siblings to shoulder extra domestic responsibilities and the siblings' grief at separation from parents. The fathers in this study also mentioned the possibility that siblings may have to deal with inappropriately directed anger from the father. Both of these studies were again limited by very small sample sizes, with clear opportunities to replicate these types of studies with larger cohorts.

More recently Goldstein et al. (2013) reviewed the scarce literature concerning the fathers of adolescents with chronic disease. The authors postulated two reasons for the difficulty in recruiting fathers for research. First, they suggested that as mothers are commonly the primary health carer, fathers are less likely to attend clinic with the child, which is often the site of recruitment into research studies. Second, Goldstein et al. (2013) contended that communication with fathers remains relatively infrequent and a low priority for healthcare teams. In their review, they found that the father's key support was his partner, and that fathers were less likely than mothers to seek broad and varied forms of support.

In our local setting in Melbourne, Australia, the CF healthcare team works hard to engage fathers from the time of diagnosis and reinforces the importance of fathers being actively engaged in their child's CF care. However, the extent to which CF care and wider parenting roles are balanced within different families, and the implications of this for children with CF and their healthy siblings, is unknown.

The impact of chronic illness on sibling relationships and adjustment

The empirical and theoretical literature about the impact of chronic illness on siblings is limited. It has also been characterised by conflicting results. As Bluebond-Langner (2000, p. 191) states "just about the only point on which there is agreement is that growing up with an ill sibling is not easy".

The majority of studies on the impact of having a sibling with a chronic illness report adverse effects on well siblings. In a 20 year old review of siblings of children with cancer, asthma, CF and other illnesses, Williams (1997) found elevated rates of externalising problems, such as aggression with peers and delinquency. A more recent meta-analysis of over 50 studies conducted by Sharpe and Rossiter (2002) found modest negative psychological effects on well siblings of children with chronic conditions, with an increased likelihood of internalising versus externalising behaviour problems. Five years ago, Vermaes et al. (2012) updated the review by Sharpe and Rossiter (2002) by adding 13 new research reports. They found a significant but small negative effect of chronic health conditions on siblings. Siblings of children with chronic health conditions had more internalising problems, more externalising problems and less positive self-attributes than siblings of healthy children. Older siblings and siblings of children with life-threatening conditions were found to be at higher risk for psychological problems.

Some studies have reported positive effects of having a sibling with a chronic illness. Silver and Frohlinger-Graham (2000) found that the female siblings of children with a chronic illness reported higher levels of interpersonal sensitivity than female siblings of children without a chronic illness. Other positive outcomes include a greater feeling of maturity and responsibility (Snethen & Broome, 2001). This finding may be related to the caretaking role that older, healthy siblings often take on such as providing assistance with medical treatments or household tasks.

Taylor, Fuggle, and Charman (2001) investigated the psychological adjustment of healthy siblings in relation to their attitudes and perceptions about their brother's or sister's chronic health condition. They also studied the siblings' mothers' awareness of

these attitudes and perceptions. Sixty-two well siblings and mothers of children with a range of chronic physical disorders completed the Strengths and Difficulties Questionnaire, a brief behavioural screening measure, among other measures. Study participants were also interviewed and illustrative comments were documented that matched the quantitative results. Well siblings' adjustment was also assessed using the Strengths and Difficulties Questionnaire (Goodman, 1997), as rated by mothers. The accuracy of mothers' reports on well siblings' attitudes and perceptions of their sibling's chronic physical disorder was defined as the statistical agreement between their independent responses on parent and child versions of the Sibling Perception Questionnaire (Carpenter & Sahler, 1991). The majority of the siblings in this study did not have adjustment problems, although the sample had slightly increased rates of emotional symptoms compared with the general population. Mothers rated well siblings as having more negative attitudes and perceptions about the physical disorder than those reported by siblings themselves. Better sibling adjustment was associated with higher maternal awareness of their attitudes and perceptions. One notable limitation of this study was that the investigators relied on maternal report to measure adjustment in the well sibling, rather than on self-report data from siblings themselves. Taylor et al. (2001) acknowledged that any future research should directly assess well siblings, rather than use indirect assessment such as that obtained when relying on a parent proxy report.

Sibling relationships are considered important reciprocal influences that foster social and cognitive development. Research has focused on healthy sibling pairs or siblings that include a child with a cognitive or physical impairment. There is limited information on siblings' perceptions of the relationship in the context in which one sibling has a chronic illness. In one of the few studies of the sibling relationship in families of children with chronic illness, Vogt (2000) compared 53 children with diabetes and their well siblings (aged 8 to 14 years). No comparison group was recruited because this study focused on the sibling dyad in terms of the sibling relationship, coping, and adaptation to diabetes. Data were collected using the Sibling Relationship Questionnaire (Furman & Buhrmester, 1985) and the School-agers Coping Strategies Inventory (Ryan-Wenger, 1990). A parent completed the Child Behavior Checklist for

each child (Achenbach, 1991). Vogt (2000) found no statistically significant differences in the perceptions of sibling relationship quality between the children with diabetes and their siblings. The warmth factor of the Sibling Relationship Questionnaire (SRQ) was positively correlated with the number of coping strategies used, the coping frequency score, and the coping effectiveness score, and negatively correlated with the conflict factor on the SRQ for both groups (children with diabetes and their well siblings). The conflict subscale of the SRQ was negatively correlated with social competency scores for the children with diabetes and the warmth factor of the SRQ for the siblings. These data provide some evidence to support the notion of the importance of siblings in terms of the reciprocal influences of siblings on their relationships, but do not provide any insights about the processes that might be involved.

Bluebond-Langner (2000) conceptualised two different approaches to understanding the impact of chronic life-threatening illnesses on well siblings. The most common understanding that was reflected in research at that time conceptualised disease as an entity with primarily negative effects on family functioning and communication. Most research at that time employed a quantitative approach to identifying the factors that produced this effect, with researchers using standardised instruments and questionnaires to measure the adjustment of siblings.

The second view conceptualised chronic illness as a complex process that sets the family and its members apart from others and creates challenges for siblings and their relationship with others. This approach emphasised the value of qualitative and ethnographic research, including participant and naturalistic observations, and open-ended interviews as methods of data collection. Plans, roles, duties, obligations, and priorities change as family life is interrupted by the burdens of treatment, with the disease a constant companion. Bluebond-Langner (2000) contended that any approach to studying the impact of chronic illness must include an understanding of peoples' everyday lives, their lived experience with the illness, how they view the illness and the meanings it has for them over the course of the illness. She proceeded to study 175 families of children with CF and included 40 in-depth family observations (Bluebond-Langner, 2000). She concluded that the well siblings' views and responses are part of a

complex process involving the patient's health status and experience of illness, the sibling's interpretation of the ill sibling's condition, the parental responses to the patient, and the sibling's assessment of these responses.

Each research methodology has its relative merits, according to the nature of the specific research question, with their combination in various forms of mixed-methods research appreciated to have particular value (Johnson & Onwuegbuzie, 2004). Rather than dichotomous perspectives, quantitative methods can be seen as a very valuable approach to measure both patient and family impacts. Incorporating qualitative methods in research studies would similarly assist our understanding of children's and adolescents' thoughts and feelings about the impact of their siblings' chronic illness.

The positive and negative impact of cystic fibrosis on sibling adaptation

A very early study by Harder and Bowditch (1982) on the impact of CF on sibling adaptation found no evidence of a negative impact and reported that children were more likely to mention the positive aspects of having a sibling with CF. However, Eiser (1993) reported that siblings of ill children were less likely to have opportunities for joint activities.

Deeley (1996) investigated the impact of having a sibling with CF from the perspective of the well child. Her study aimed to identify both positive and negative aspects for the well siblings. Nineteen children with a mean age of 11 years from 12 families who had a child with CF participated in the study, at a time when children with CF were relatively unwell. Deeley (1996) asked a series of questions including "Are there difficult things about having a brother/sister with CF? What are they?" and "Are there any good things about having a brother/sister with CF? What are they?" Children in this study described both positive and negative issues, but the negative issues had more of an impact on their daily lives. Two of the nineteen children reported that there were no difficult issues and six children reported that there were no positive issues to having a sibling with CF. Parental differential treatment was reported as a difficult issue by half of the participants. One quarter of the children reported reduced opportunities for

participating in joint activities with siblings as a difficult issue. Even when children found an activity that their sibling could join in, it often had to be terminated prematurely due to illness. Five children mentioned the negative impact on the sibling relationship because they could not do the same things as children with healthy siblings. Making sacrifices was another negative issue identified by participants in this study. This related to sacrificing their own activities for the sake of their ill sibling, which had an impact on friendships, and prevented the children from pursuing their favourite activities.

The positive aspects of having a sibling with CF, as found by Deeley (1996), included differential treatment and attention from others when siblings were in hospital and treats, such as special holidays or gifts from the local CF group. Two older (14 year old) siblings also mentioned personal development as a positive aspect of having a sick sibling. This included understanding others when they are upset and a better understanding of the illness. A limitation of this study for contemporary practice is that it was conducted at a time when people with CF largely died during childhood and adolescence, which is usually no longer anticipated.

Russo and Hogg (2004) conducted a small pilot study to explore both positive and negative issues that arise for the healthy siblings of children with CF across different stages of development. They conducted brief interviews with nine children from three different age groups: under six years, 7-12 years and 13-18 years. As expected, Russo and Hogg (2004) found that the younger age group were limited in expressing their feelings about the positive and negative aspects of having a sibling with CF. Feelings of jealousy due to lack of attention were reported by the three children in the 7-12 year old age group. Younger children simply wished for CF to go away, whereas a greater degree of acceptance appeared to be present from age 13. The older group also spoke about their hopes and desires for a cure for CF, which was not articulated by the younger groups. Their study provided valuable insights about the lived experience of siblings of children with CF. While the study raised questions about the potential for parental differential treatment to be experienced as a negative aspect of CF, this was not

articulated as a feature of this pilot work, and there was no assessment of sibling adjustment.

In addition to age, Wennstrom, Berg, Kornfalt, and Ryden (2005) identified some interactions with gender in the self-evaluation of healthy siblings of those with CF aged 6-14 years old. They studied 55 sibling pairs and compared self-evaluation scores using the "I think I am" self-evaluation questionnaire. No differences were identified between siblings with and without CF and in comparison to standardised scores for a Swedish population. The healthy siblings scored higher than the Swedish reference group for skills, talents and abilities. Wennstrom et al. (2005) suggested that well siblings feel superior to their siblings with CF in some aspects of their lives, and hypothesised that this might reflect differences in the amount of time required in CF care by their ill sibling. They also identified a small number of differences by gender in a limited number of subscales. For example, they found that girls in families with CF (both healthy siblings and those with CF) scored lower than the reference group for mental well-being and the relation to parent and family sub-scores.

Parental differential treatment

Parental differential treatment refers to inequities in the type or frequency of parental behaviour directed toward siblings in the same household (Opipari, 1996). The term parental differential treatment was first used by developmental psychologists in the context of studying normal sibling relationships. This research thread was explored by McHale in her studies of siblings in families with a physically disabled child in which she examined parent self-report of time spent with younger and older children (McHale & Pawletko, 1992).

Although siblings have approximately half of their genetic makeup in common, they less commonly share personality traits (Daniels & Plomin, 1985). There is evidence to suggest that the family environment is an important influence on children's social and emotional development (Brody, Stoneman, & MacKinnon, 1986; Grych & Fincham, 1990). To date, however, the emphasis has been on between-family environmental factors (such as socio-economic status) that influence child development, rather than on

processes within families. In a seminal review from the early 80s, Maccoby and Martin (1983) contended that researchers must assess processes that differ within families given that variables which differentiate families have not accounted for substantial proportions of the variance in child outcomes. The focus on intra-familial variations is evident in the literature on relationships between variations in siblings' family environment and children's social and emotional adjustment. Several non-shared influences have been identified, including differences in peer relationships, variations in temperament and parental differential treatment (Brody, Stoneman, & McCoy, 1992; Stocker, Dunn, & Plomin, 1989; Volling & Belsky, 1992). Of these non-shared experiences, parental differential treatment has been most commonly linked with child outcomes. In particular, it has been shown to be strongly predictive of the psychological wellbeing of individual siblings (McHale & Pawletko, 1992). Given the extent of parent care that is required in families with at least one child with CF, this non-shared influence potentially operates as a source of parent differential treatment and is important to study further in these families.

Parental differential treatment involves interactions between three family members (i.e., a parent and two siblings) and is a reflection of ongoing relationships and processes within a family. In families with CF, where the extent of non-shared influences is substantial, understanding the processes around how parent differential treatment occurs and is modified within the family is highly relevant.

There are various ways in which parents are able to differentially attend to their children. This can be expressed in relation to the provision of tangible rewards (for example, food, gifts, clothing and money), subjective differences in relation to parent expressions of affection and inconsistencies around punishment. Other differences relate to opportunities, whether in relation to sports and hobbies or choice of school. Parent time is limited, and the amount of time that parents spend with each child is another way in which parental differential treatment can be expressed.

Parental differential treatment occurs to some degree in all families. However, it may occur to a greater degree in families with a child with a chronic illness (Quittner &

Opipari, 1994). A critical question is whether greater differential treatment occurs in families in which parents are caring for a disabled or chronically ill child, due to the increased time demands associated with the child's care needs.

Reports in the literature describe how parents can feel guilty about the inherited nature of their child's condition, such as CF (Priddis, Dunwoodie, Balding, Barrett, & Douglas, 2010), as though they intentionally gave their child CF (Havermans, Tack, Vertommen, Proesmans, & de Boeck, 2015). Parental guilt is a factor that can drive differential treatment through attempts to compensate the child for missed opportunities, whether opportunities are actually missed or it is perceived they will be missed in the future (Quittner, Opipari, Regoli, Jacobsen, & Eigen, 1992). For parents of children with CF, the guilt of it being an autosomally inherited genetic condition may be compounded by the seriousness and severity of the condition, making parental differential treatment a potentially potent issues for families.

McHale and Pawletko (1992) published the first study to explore maternal differential treatment in a high-risk context that compared families in which the younger school-age member of a sibling pair was intellectually disabled versus families in which both children were able bodied. The subjects were 62 children aged 8 to 14 years old, half of whom had a disabled younger sibling and half of whom had an able bodied younger sibling. A telephone checklist, undertaken with mothers, was used to assess differential involvement in mother-child activities, as well as a home interview of mothers to assess differences in the discipline techniques used with each child. The authors found greater maternal involvement with the younger disabled siblings in activities such as helping and play. While these authors predicted differential treatment that favoured less discipline of disabled children, they found no evidence that parents' discipline differed between healthy and disabled siblings. It is not known to what extent the results from this study, while providing important information relevant to childhood intellectual disability, are generalisable to children with chronic health conditions such as CF. Beyond the obvious fact that most children with chronic health conditions do not have an accompanying intellectual deficit, a key difference is that most children with chronic

health conditions require daily medication or other forms of treatment that are less common in children with intellectual disability.

McHale and Pawletko (1992) used a predetermined checklist of activities to measure mothers' activity patterns. This approach limited the number of possible activities they could measure. It also required mothers to first report on their behaviour with one sibling and then the other. This may have increased their awareness of the differences in the behaviours they directed toward each child.

In terms of measuring parental differential treatment, the majority of previous investigations have used videotaped observations of mothers interacting with their children during an activity (for example, playing a small hand-held computer game) (Chamberlain & Reid, 1987). This is an obtrusive methodology that may lead to bias because of social desirability responding by mothers. An additional issue relates to the artificial nature of the observed tasks (for example, non-spontaneous play observed by a researcher), rather than assessing the same behaviours as they occur in the natural environment (Quittner & Opipari, 1994).

Quittner and Opipari (1994) improved on these approaches by employing an extensive diary tracking procedure that enabled them to obtain a continuous account of mothers' time and activities as they unfolded throughout the day. This diary methodology has been well validated in studies of role strain in couples raising a child with CF (Quittner et al., 1998) and also used in studies of adherence behaviours (Grossoehme et al., 2013; Grossoehme et al., 2015). In addition to measuring time, they assessed who was involved in each activity and how positive or negative the quality of that time was. Measurement of parental differential treatment through the diary tracking procedure is advantageous for many reasons. In addition to being less obtrusive, it can be administered without invoking social desirability bias, and provides a means of measuring the relevant components of differential treatment as they change across developmental stages.

As described earlier, another limitation is that few studies have included fathers in their assessment of differential treatment. The only studies in the literature that involve assessment of fathers are by Brody et al. (1992) and Volling and Belsky (1992). Both indicated that paternal differential treatment has an important role in determining the quality of children's sibling relationships, which underscores the importance of studies of parental differential treatment investigating the practices of both mothers and fathers.

Parental differential treatment and cystic fibrosis

Differential treatment of children is perhaps to be expected by parents given the physical and emotional demands placed on them by the daily management and care of children with CF (Foster et al., 2001). In a qualitative study investigating the impact of CF and treatment on eight patients, eight mothers, one father, and eight siblings, Foster et al. (2001) conducted semi-structured interviews that included questions about the management of the illness and impact of the illness on the family. The patients and siblings were aged between 9 and 21 years. Qualitative analyses revealed high levels of parental involvement in treatment, minimal involvement of siblings, and preferential treatment towards patients. Patients were reported to receive greater attention than siblings because of their illness and the daily treatment demands, whether they were symptomatic or not. Parents were found to be less tolerant of siblings' misbehaviour similar to the findings of Walker, Garber, and Van Slyke (1995). Parents described much of their differential treatment as unintentional. Both patients and parents attributed sibling resentment to the differential treatment (Foster et al., 2001). This study was limited by the wide age range of the participants with CF and siblings and the reliance on subjective reports of parental differential treatment. An objective measure would provide a more accurate account of the amount of time parents spend with their children with CF and the types of activities they engage in.

Derouin and Jessee (1996) conducted a qualitative study investigating siblings' perceptions of family disruption when a brother or sister had CF or asthma. Data were gathered by phone interviews with siblings of chronically ill children employing open-ended questions that focused on the impact of the illness on the sibling and the family

unit. The sample size was small; fifteen families from the two illness groups participated. Six male and nine female respondents, with a mean age of 10.1 years, participated in the study. The average age of the well sibling was slightly older than the average age of the chronically ill child. For siblings of both disease groups, positive outcomes included strengthening relationships, achieving independence, and experiencing satisfaction from improvement in the health of the unwell sibling. Negative outcomes included worrying about the brother's or sister's illness, being jealous of the attention paid to the ill child, and the restriction of family events. Nine of the 15 (60%) healthy siblings of a child with CF reported experiencing parental differential treatment in favour of the ill child.

Quittner and Opipari (1994) assessed parental differential treatment using the daily phone diary (DPD). This method enables analysis of the amount of time parents spend with each child individually, the types of activities and parents' ratings of their mood. Theoretically, cued recall of all events over a 24 hour period limits participants' understanding of the behaviours of interest to the researchers (in this case, the differences in time that parents spend with their different children) and therefore decreases the likelihood of parents giving socially acceptable responses. Quittner and Opipari (1994) studied 40 mothers of toddlers and pre-schoolers (20 with CF and 20 without CF) and measured differential treatment using home interviews, nightly phone ratings, and DPDs. Little evidence of parental differential treatment was found in the home or phone interview data. Using the DPD, mothers were found to spend more time with their younger children with CF than their older healthy children, particularly in activities related to play and mealtimes, even after excluding the time spent in medical care. Mothers in the CF group also rated the time spent with older children as significantly more negative than time spent with younger children. However, differences in parental differential treatment were found in the CF versus comparison families on the DPD variables, which provide a sensitive assessment of activities.

The study conducted by Quittner and Opipari (1994) was limited by several factors. By focusing on families with very young children they could not get ratings of the quality of the sibling relationship or measures of child functioning except from maternal report.

Second, they did not collect DPD data from fathers to measure how fathers spent their time. It would be important to know if fathers in CF families spend more time with older siblings to compensate for the greater caregiving demands on the mother.

In a follow up study of 48 older children, Opipari (1996) examined parental differential treatment of school age and young adolescent siblings in families with and without a child with CF. Opipari (1996) also found greater parental differential treatment in families of children with CF, with poorer emotional and psychological adjustment reported by older healthy siblings. This study provided the first evidence of an association between parental differential treatment and measures of child functioning. The assessment of parental differential treatment was limited to maternal interactions with siblings. Although Opipari (1996) obtained an assessment of older siblings' involvement with fathers, levels of paternal differential behaviour were not assessed.

Marciel (2004) conducted the first study to examine the magnitude of parental differential treatment at different points in development. The primary objective of this study was to assess parental differential treatment in children with CF and their healthy siblings across three developmental stages. The second aim was to determine the short-term stability of parental differential treatment over a six-month period, using DPDs to assess parental differential treatment. The participants in this study were 81 parents of children with CF (aged 1 to 18 years) and their healthy siblings (aged 0 to 25 years). Families were placed into one of three developmental age groups according to the age of their child with CF: pre-school, school-age, or adolescent. Parents were called three times consecutively on one weekend day and two weekdays. These DPD data were collapsed by activity and averaged across the three days.

Marciel (2004) found parental differential treatment in favour of the child with CF across all three developmental cohorts. Parents particularly directed more time towards children with CF in the preschool age cohort compared with those in the school-age and adolescent cohorts. Three diary assessments (at baseline, three, and six months) were available for a subset of 15 families and the extent of differential treatment was found to be stable across all three time points. In other words, caregivers who treated their

children more differentially continued to do so, placing the healthy siblings at risk for negative outcomes.

A limitation of this study was the lack of a control group, which would have enabled normative developmental changes in parental differential treatment to be addressed. Marciel (2004) also noted the lack of measures of the quality of the sibling relationship and the psychological and social adjustment of the healthy siblings. Including these outcome measures would provide inferences about the consequences of differential treatment on the well siblings. Marciel (2004) concluded that "given the consistency of differential treatment found in this study, future research should carefully examine these variables" (Marciel, 2004, p. 34). The 81 parents in this study were predominantly mothers (89%). Although mothers are commonly the primary caregivers in families with CF, there is not yet any study that has collected DPDs from both mothers and fathers, which is arguably needed in order to better understand the complex family dynamics and interactions in families in which a child has CF.

Methodological problems with previous sibling research

As the critique of many of these studies of the impact of chronic illness on sibling relationships would suggest, there are many limitations within the existing literature. In addition, a recent review of research concerning siblings' perspectives within the familial experiences of chronic childhood illness found that many of studies were based on information provided by adult participants rather than children (Knecht, Hellmers, & Metzing, 2015). The authors correctly concluded that due to the extent of reliance on proxy perspectives (such as parents) it is difficult to gain an accurate impression of a sibling's world when growing up with a brother or sister affected by chronic illness. Disappointingly, this conclusion is consistent with that made 15 years earlier by Bluebond-Langner (2000) when she commented that conclusions based on interviews, questionnaires, behaviour checklists or other instruments that are only administered to parents or teachers are problematic. What makes this especially pertinent is that Menke (1987) found that parents underestimated the extent to which healthy children were worried and suggested that parents' preoccupation with the ill child meant that they had

few resources left for their other children. Until relatively recently, researchers have rarely sought to obtain responses directly from well siblings about what it is like to have a brother or sister with a chronic illness, a limitation of much of the early research in this field. Direct measures from well siblings are therefore vital to accurately understand their lived experience. Beyond involving well siblings directly, it is also important to appreciate the bi-directional nature of the sibling relationship (Dunn & McGuire, 1992).

A further limitation of the literature is the lack of use of comparison groups; it is difficult to draw conclusions from the various studies about the impact of chronic illness on sibling relationships in the absence of this (Lobato, Faust, & Spirito, 1988). Without a comparison group of families not affected by chronic illness, it is impossible to know if it is the illness that is responsible for parental differential treatment and whether negative sibling relationships or poor adjustment are a function of the chronic illness.

Researchers have noted that sibling relationships do not exist in isolation from broader family relationships (Hetherington, 1994). As identified throughout this literature review, there has been very little research conducted on the views and experiences of fathers concerning their involvement in caring for their children with CF (Hayes & Savage, 2008). Existing knowledge on how parents manage the care of their children with CF is predominantly based on mothers' perspectives. As a result of the increased demands of raising a child with CF, families may choose to divide child care responsibilities in many different ways. For example, the mother may provide more care for the ill child but the father might take greater responsibility for the other children. The recruitment of fathers is therefore needed to enhance our understating of whether differential parental differential treatment actually exists and if so, to what extent it differs for mothers and fathers of children with CF.

Most of the sibling research has been conducted in the United States and Europe, with very little Australian research. Cuskelly (1999) noted that culture is likely to be an important mediator for sibling experience. Beyond culture, there are also differences in clinical practices. For example, Australia introduced NBS well before it was introduced across the United States. How families relate to their children with CF diagnosed in the

era of NBS will differ from when CF was clinically diagnosed, if only in the age of diagnosis and experience of illness.

As with the uncertainty about the generalisability of research from intellectually disabled children to the families of children with chronic health conditions, so too is there uncertainty about the extent to which evidence from a single disease applies more widely to other disorders. The focus of non-categorical research, that is research including different conditions, is to explore and understand putative similarities across a variety of chronic health conditions, such as the financial and emotional impact on a family of having a child with a chronic health condition (Stein & Jessop, 1982). Such impacts are viewed as meaningful, regardless of whether, for example, a child has CF, diabetes, or cancer (Havermans, Croock, Vercruysse, Goethals, & Diest, 2015). However, Vogt (2000) highlights that few studies describe the similarities or differences of the illness trajectories or provide a rationale for inclusion of the various disorders, which could be expected to create differential impacts.

Havermans, Croock, et al. (2015) emphasise the value of a categorical approach in clinical settings, as this is where specific services are typically delivered to children and their families. Most research is also categorical, that is, focused on an individual health condition and its specific characteristics which are perceived to be more meaningful or influential on a particular outcome (Gallo & Knafl, 1993; Williams, 1997). Few researchers have investigated the impact of specific disease trajectories and their associated daily treatment regimens on sibling adjustment (Drotar & Crawford, 1985; Gallo & Knafl, 1993; Lobato et al., 1988). As a result, it is unclear to what extent research on parental differential treatment within one condition that, for example, might have a static course (such as diabetes) is relevant to another that is of a chronic progressive course (such as CF). When it comes to the state of research about parental differential treatment in families with chronic health conditions, the relative paucity of knowledge about any disease suggests that the research priority should be a disease-specific approach, at least initially, but with interest in how results might generalise more widely.

Aims of this Study

As outlined previously, family-centred care for children with CF has typically overlooked the needs of healthy siblings. A considerable literature exists on the impact of chronic illness on the affected child and their parents. In Australia, the importance of the healthy siblings' perspective gained attention with the publication of Kate Strohm's first book "Siblings: brothers and sisters of children with disability" (Strohm, 2002) with the inaugural Siblings Australia conference held soon after in 2004. There is still, however, a paucity of research on the adjustment of well siblings and remarkably little research on the siblings of children with chronic health conditions including CF.

Due to improved treatments, the life expectancy for people with CF in Australia and internationally has increased dramatically. The change in CF from a disease leading to death in infancy to a disease of adulthood with a long and more complicated course imposes a huge potential for psychosocial ramifications for the affected individual and the extended family. In particular, the relationship between affected and unaffected siblings is more enduring and therefore is likely to be more significant than ever before across the life-course of both the individual with CF and their siblings.

The overall objective of this study was to assess the extent of parental differential treatment in families with a child with CF and a healthy sibling and to measure the impact of parental differential treatment on the social and emotional adjustment of the well siblings in families caring for a child with CF. To do this, I set out to recruit two types of families - those with a child with CF and an older healthy sibling and those with two healthy children to use as the comparison group. I wished to recruit both fathers and mothers, as I was particularly interested in the function of the family as a whole.

The current study is similar in design to the research by Opipari (1996) and used DPDs to obtain an objective and sensitive measure of parental differential treatment and was designed to address some of the limitations and unanswered questions of previous research. In particular, the measurement of parental differential treatment was expanded to include fathers. Specifically, the DPD enabled calculation of the amount of time

parents spent in a variety of activities with their younger and older children to be collected from both mothers and fathers. In addition, information was collected directly from the older siblings on a range of social and emotional outcomes, while both younger and older siblings also reported on the quality of the sibling relationship.

The first aim was to examine the magnitude and type of differential treatment occurring among families with and without a child with CF. Differential treatment by mothers and fathers in favour of the younger child was expected in both types of families, as assessed by the DPD (Quittner & Opipari, 1994). However, a greater magnitude of differential treatment by mothers and fathers was expected in the CF versus comparison group.

The types of activities engaged in by mothers and fathers in both groups of families were also examined. Given that mothers are typically the primary caregivers for their child with CF, they were expected to spend more time in medical activities than fathers in the CF group. After excluding time spent in medical care, mothers in the CF group were still predicted to spend more individual time with the younger child and more time in activities, such as meal times, due to the importance of nutrition in the management of CF.

The second aim of the study was to assess the relationship between the extent of parental differential treatment and child outcomes for the older children in both groups. Across groups, a greater magnitude of differential treatment in favour of the younger sibling was expected to be associated with less adequate social and emotional functioning in older siblings (emotional distress, social skills, behaviour problems). In addition, an interaction effect was expected with greater differential treatment in those families caring for a child with CF. Thus, a negative association between parental differential treatment and social and emotional adjustment was expected in the older, healthy children in the CF versus comparison group.

The third aim of the study was to assess the relationship between parental differential treatment and the quality of the sibling relationship in both groups. For both groups, a

greater magnitude of maternal and paternal differential treatment in favour of the younger sibling was expected to be associated with worse sibling relationship quality (i.e., decreased positive and increased negative sibling behaviours). In addition, siblings in the CF group were expected to report worse sibling relationship quality than siblings in the healthy comparison group.

The fourth aim was to collect descriptive information on both the positive as well as the negative or challenging aspects of being a sibling of a child with CF. It was anticipated that sibling experiences would differ according to the age of the healthy sibling. Healthy siblings aged 7 – 10 years were expected to cite concrete rewards (for example, gifts from organisations, trips awarded through charities) as the main advantage of having a sibling with CF, but healthy siblings aged 14 – 16 years were expected to report that having a sibling with CF led to greater maturity and a heightened sensitivity to and understanding of the needs of people with chronic illness. The negative aspects of having a sibling with CF were expected to include being unable to participate in after-school activities or plan family excursions for the healthy siblings aged 7 – 10 years, whereas emotional concerns about the health of their sibling with CF was expected to be the main disadvantage cited by healthy siblings aged 14 – 16 years.

METHODS

Setting

The Royal Children's Hospital (RCH) in Melbourne, Victoria, is a specialist paediatric hospital that provides a range of clinical services, and health promotion and prevention programmes for children and adolescents. The hospital is the major specialist paediatric hospital in Victoria, and also cares for some children from Tasmania and southern New South Wales, other states around Australia, and overseas. The specialist CF service at the RCH cares for approximately 260 children from birth to 19 years, making it one of the largest paediatric CF centres in the world.

I have worked as the CF Clinical Nurse Consultant on the RCH's CF team for over 25 years. This position involves the provision of counselling and support for children with CF and their families who attend the hospital's CF service.

Ethics approval

Ethics approval was obtained from The Royal Children's Hospital Ethics in Human Research Committee on 2 May 2003 (Project No 22158), as shown in Appendix A. In addition to the original approval, two minor modifications were obtained across the project in relation to the final versions of the information statements (copies not shown in Appendix).

Subjects

Development of a sibling database

Information about the number of siblings in families attending the CF service at the RCH is not systematically recorded. In order to undertake this research, an initial task required the development of a sibling database. This information was compiled by systematically asking all parents at outpatient visits for details about their other children (living and deceased). This information included: name, date of birth and sex. Within

the database, this information was linked to the sibling with CF to derive a list of healthy siblings who met the inclusion criteria for the study.

Eligibility criteria and recruitment of CF families

Families in the CF group were recruited from the Department of Respiratory and Sleep Medicine at the RCH in Melbourne in 2003. Families were eligible for participation if they had one child with CF between 6 and 14 years, an older sibling without CF between 7 and 15 years and no child who had died of the disease. This age group was chosen for two reasons: to enable direct measures to be obtained from the siblings rather than parent report, and to facilitate comparisons between the results of this study and previously published work by Quittner and Opipari (1994). The disadvantage of restricting the age of eligible siblings was the potential for a smaller sample size.

Families with children with other chronic illnesses or disabilities in addition to CF were excluded because of the difficulty in separating the impact of CF from other conditions. This occurred in two cases in which the sons with CF had also been diagnosed with autism. Thirty-nine families from the CF clinic met the eligibility criteria. All eligible families agreed to participate (see Figure 4).

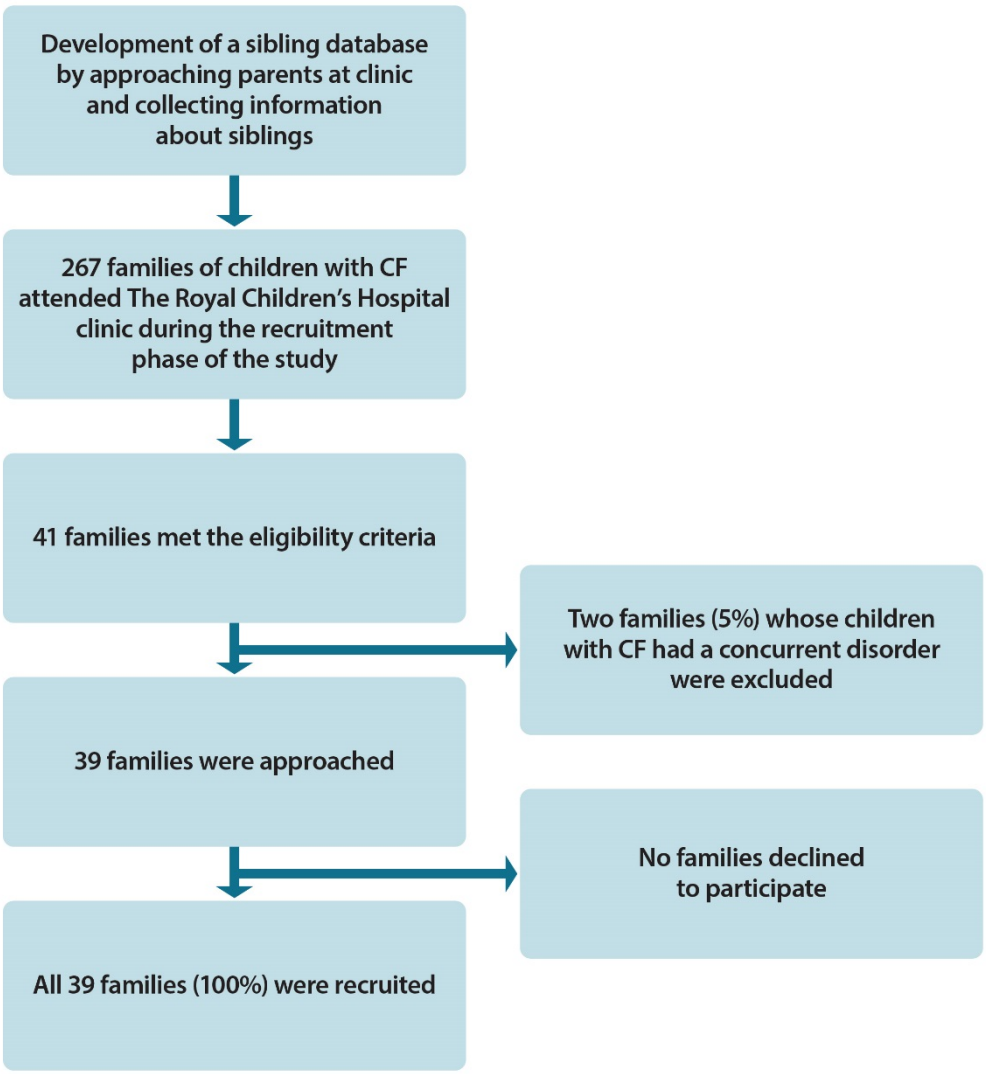

Figure 4. Flow chart summarising the recruitment of CF families.

Recruitment of comparison families

Different methods of recruiting families for a comparison group were considered. These included advertising for participants, and approaching the Department of Education and Training Victoria for access to suitably aged students. The disadvantages of these methods included a low response rate, time required to recruit an adequate sample, the need to obtain ethics approval from the Department of Education and Training Victoria and the challenge of matching the CF sample for socio-economic status. After weighing

up the pros and cons of each method, it was decided to recruit the comparison group by asking parents in the CF group to nominate a family with similarly aged children who were not related to them and did not have a child with a chronic illness or disability. This method was employed with the expectation that the CF families would nominate comparison families that were similar in terms of age, income and socio-economic status. A better response rate was also anticipated using this approach because potential comparison families were likely to be friends of the CF families, and would show their support by participating in the study. Twenty-nine comparison families were recruited for the study (see Figure 5).

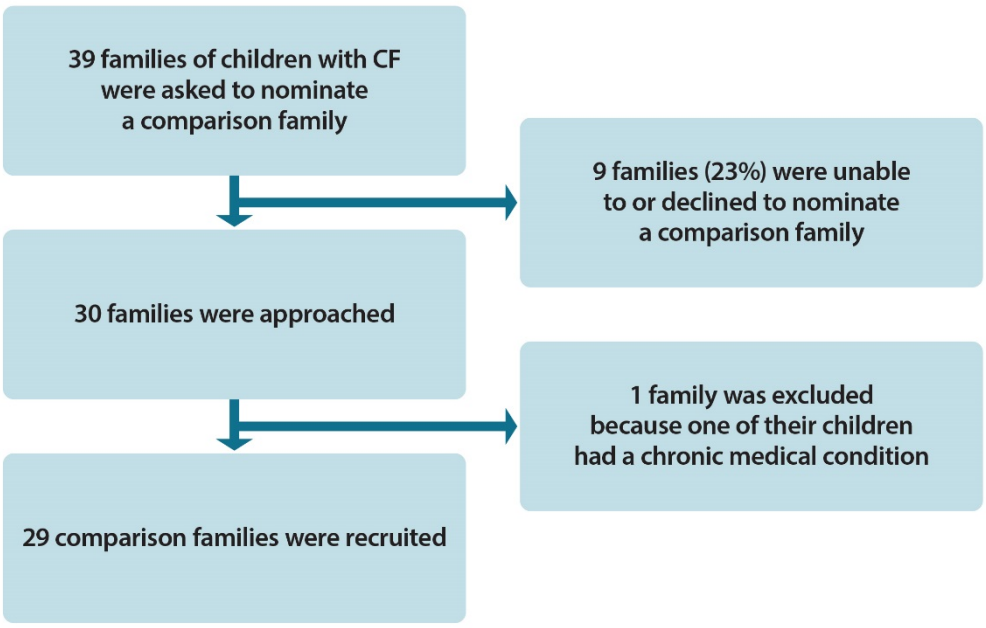

Figure 5. Flow chart summarising the recruitment of comparison families.

Nine families in the CF group did not nominate a comparison family. Of these, two families declined because they had not disclosed to others that their child had CF, one family was experiencing marital distress and six families said they were unable to think of anyone. One nominated comparison family was excluded because one of their children had a chronic medical condition.

A series of independent sample *t*-tests and χ^2 analyses were used to examine differences between families with CF who nominated a comparison family and CF families who did not, on a range of demographic variables: mother's age, father's age, mother's and father's educational level. The family experiencing marital distress and the one who nominated an unsuitable comparison family were not included in these analyses. Overall, no significant differences in demographic characteristics were observed. There was a slight difference between the two groups in terms of attendance at counselling (for any reason). A higher proportion of families who did not provide a comparison family reporting attending counselling (75% versus 35% of the families who did provide a comparison family); however, it was not statistically significant.

Those who did and did not nominate a comparison family were compared on the basis of CF child characteristics. Although there was no significant difference in the age or lung function of the children with CF in these two groups, a significant difference was found in the number of days spent in hospital in the previous year $t(29.6) = 2.36$, $p<.05$. Children with CF whose families nominated a comparison family spent an average of 15 days in hospital (*SD* = 31.5 days) in the previous year, compared to an average of one day (*SD* = 2.83 days) for families who declined to nominate a comparison family. It is possible that some families are still reluctant to disclose the diagnosis of CF within their social network. The finding that children with CF in the families who declined to nominate a comparison family spent significantly less time in hospital suggests that this allows the diagnosis to be hidden more easily.

Final sample

Sixty-eight children between seven and 15 years, their younger siblings and both parents (except in single parent families) participated in this study. Younger siblings were between the ages of 6 and 14 years. In 39 of the families, the younger siblings had CF (CF group), while in the other 29 families, the younger siblings were not affected by any chronic illness or disability (Comparison group).

Parent characteristics

Mothers and fathers in both groups were generally in their early forties. A *t*-test revealed no significant difference in the ages of the parents in both groups (see Table 1). Slightly more than half of the mothers had a tertiary education and 26.5% of fathers in the CF group and 32.1% of fathers in the Comparison group had completed tertiary education.

In terms of marital status, 15.4% of mothers in the CF group were single compared to 3.4% in the Comparison group. One of the CF families separated soon after the study visit; this father did not continue to participate. One father in the Comparison group chose not to participate because he was not interested in being involved in research. All of the fathers in the CF group were employed and 66.7% of mothers in the CF group worked either full or part-time. In the Comparison group, 92.6% of fathers and 86.2% of mothers were either working full or part-time.

In terms of family size, 46.2% of the CF families had 2 children, 41.0% had 3 children and 12.8% had 4 or more children. In the Comparison group, 41.4% of the families had 2 children, 41.4% had 3 children and 17.2% had 4 or more children. A χ^2 analysis found no difference between the CF and Comparison families on these variables.

Tobacco use is known to be more prevalent in lower socio-economic groups (Haustein, 2006). Thirty-five percent of families in the CF group had one or more smokers in the home compared with 24% for the Comparison group.

χ^2 tests indicated that there were no significant differences between the groups on the following demographic variables: mothers' education level, fathers' education level, marital status, family size, and presence of smokers in the household.

Table 1

Means and Standard Deviations for Parent and Family Characteristics

		CF GROUP			COMPARISON GROUP	
Age (years)	*n*	M	*SD*	*n*	M	*SD*
Mothers	39	40.08	5.01	29	40.45	4.50
Fathers	34	43.82	5.82	28	44.32	5.64
Mothers' Education Level	39			29		
Secondary		6 (15.4%)			4 (13.8%)	
Trade/TAFE		13 (33.3%)			10 (34.5%)	
Tertiary		20 (51.3%)			15 (51.7%)	
Fathers' Education Level	34			28		
Secondary		34 (47.1%)			11 (39.3%)	
Trade/TAFE		9 (26.5%)			8 (28.6%)	
Tertiary		9 (26.5%)			9 (32.1%)	
Marital Status	39			29		
Partnered		33 (84.6%)			28 (96.6%)	
Single		6 (15.4%)			1 (3.4%)	
Percentage Employed						
Mothers	39	26 (66.7%)		29	25 (86.2%)	
Fathers	34	34 (100%)		28	26 (92.6%)	
Family Size	39			29		
2 children		18 (46.2%)			12 (41.4%)	
3 children		16 (41.0%)			12 (41.4%)	
4 children		5 (12.8%)			5 (17.2%)	

Significantly fewer parents in the CF group had attended a parenting course than the Comparison group, χ^2 (1, N=68) = 5.28, p=.022. In terms of participation in counselling sessions, more families in the CF group (56%) had attended family counselling than the Comparison group (34%). This relationship, despite being descriptively large, was not statistically significant, primarily because of insufficient power. It is important to note that counselling for the CF group was more likely to be focused on sibling issues (37%) than in the Comparison group (8%).

Child characteristics

FEV_1 is the maximal volume of air exhaled in the first second of a forced expiration from a position of full expiration (Miller et al., 2005) and is used as one measure of the severity of respiratory disease in CF. The 39 siblings with CF varied in disease severity based on pulmonary functioning, with a mean FEV_1 of 92.8%, ranging from 37% to 136%. The group was divided according to lung function: mild lung disease (70% predicted FEV_1 and above), moderate lung disease (40-69% FEV_1) and severe disease (below 40% predicted FEV_1). Thirty-five siblings with CF (89.7%) had mild lung disease, 3 (7.7%) had moderate lung disease and one sibling with CF (2.6%) had severe disease. The majority of the group (64%) had not required hospitalisation in the past year. Of the 14 children who required one or more admissions to hospital in the previous year, the mean length of stay was 11.5 days (ranging from 2 to 120 days). There was a significant difference between the younger siblings in the two groups in the number of days of school missed in the previous year, t(54) = 3.16, p = .003. The children with CF missed significantly more days of school (M_{days} = 29.48 versus M_{days}= 10.80 for Comparison group).

No group differences were found on any of the other child characteristics. A t-test revealed no significant difference in the ages of the younger or older siblings across groups (see Table 2).

Table 2

Means and Standard Deviations for Child Characteristics

	CF GROUP (*n* = 39)		COMPARISON GROUP (*n* = 29)	
Age (years)	M	SD	M	SD
Younger sibling	10.26	2.16	10.08	2.11
Older sibling	12.72	2.29	12.78	2.18

χ^2 tests indicated no significant group differences in the gender of either the target or younger siblings or the gender composition of the sibling dyads. Across both groups, 38 of the target siblings were girls and 30 were boys, and 32 of the younger siblings were male and 36 were female. Table 3 shows the gender composition of the sibling dyads for both groups.

Table 3

Gender Composition and Sibling Dyads Across Both Groups

	CF GROUP (*n* = 39)	COMPARISON GROUP (*n* = 29)
Gender		
Younger sibling		
Female	19 (48.7%)	17 (58.6%)
Male	20 (51.3%)	12 (41.4%)
Older sibling		
Female	24 (61.5%)	14 (48.3%)
Male	15 (38.5%)	15 (51.7%)
Dyads		
Girl – girl pairs	11 (28.2%)	9 (31.0%)
Boy – boy pairs	7 (18.0%)	7 (24.1%)
Older girl – younger boy pairs	13 (33.3%)	5 (17.2%)
Older boy – younger girl pairs	8 (20.5%)	8 (27.6%)

χ^2 tests indicated that there were no significant group differences in birth order (see Table 4). In conclusion, the parents and siblings across the two groups had similar demographic profiles.

Table 4

Birth Order for the CF and Comparison groups

	CF GROUP (*n* = 39)	COMPARISON GROUP (*n* = 29)
Birth Order		
Younger sibling		
second-born	29 (74.4%)	19 (65.5%)
subsequent	10 (25.6%)	10 (34.5%)
Older sibling		
first-born	29 (74.4%)	19 (65.5%)
subsequent	10 (25.6%)	10 (34.5%)

Procedure

Families of children with CF were contacted first by telephone. This call, from the principal investigator, was followed with written information about the study. In accordance with ethics committee procedures, the packages sent to families included information about the study requirements, consent forms for the parents and assent forms for children (see Appendix A). Parents and children were asked to read the information statements and contact the principal investigator if they had any questions. This information explained that counselling was available to all siblings and parents should any distress be caused by the questions asked in the study.

A package containing an introductory letter about the study, copies of information statements, consent forms for the parents and assent forms for the children was given to each mother in the CF group to give to potential comparison families (see Appendix A). The letter explained that, for privacy reasons, the CF family had not revealed any details about the potential comparison family to the principal investigator or research associates, and that participation in the study was voluntary and confidential. The letter requested that the family contact the principal investigator if they were willing to participate in the study.

Prior to commencement of data collection the study questionnaires were administered to a sibling pair with no chronic illness, and a family from the CF clinic that was not eligible for participation because they had more than one child with CF. The aim was to pilot the questionnaires to determine their appropriateness and the length of time for administration. No amendments were indicated from this preliminary work.

Methods of data collection

Two methods of data collection were used in the study. First, the target siblings (older siblings from both groups), their next youngest brother or sister and their mothers participated in a meeting with the principal investigator. During this meeting, the target siblings completed questionnaire measures regarding their social and emotional adjustment and the quality of their relationship with their younger sibling. The target siblings from the CF group also answered four open-ended questions about the advantages and challenges of having a brother or sister with CF. Younger siblings also completed the sibling relationship questionnaire about the quality of the sibling relationship. The principal investigator administered the questionnaires for children 11 years and under while the older participants completed them via self-report. Each child completed the questionnaires in a quiet area separate from and without input from their mother or sibling. For half of the sample the order of administration of the questionnaires was reversed. Mothers reported on family demographic information.

Parental differential treatment of younger and older siblings was assessed using the DPD. Following the face-to-face meeting, mothers and fathers were interviewed

separately by telephone to assess their activity patterns over the same 24-hour period. This was done on three occasions (two weekdays and one weekend day) consistent with previously published work (Grossoehme et al., 2013; Grossoehme et al., 2015; Quittner & Opipari, 1994). For all activities lasting at least five minutes, parents were asked to report the type of activity, its duration and who was present. Phone diaries were only conducted on typical days (not during holidays, hospitalisation or times of significant disruption to the family's routines).

A research assistant was employed to collect the phone diary data using funding from an Australian Cystic Fibrosis Research Trust grant. Utilising a research assistant for the collection of this information stemmed from the principal investigator's concern that the clinical relationship between the parents in the CF group and the principal investigator could lead to socially desirable responses. Given that poor adherence to CF treatments is quite common (Quittner, Zhang, et al., 2014) with studies indicating 50% adherence or less, parents of children with CF may have been reluctant to accurately disclose the amount of time spent on medical treatment if asked by the principal investigator. In accordance with Quittner and Opipari (1994), all phone diaries were collected by one interviewer to maintain a consistent tracking style.

Data collection took two years (from 2003 to 2005) and involved more than 7,000 km of travel to enable the recruitment of CF and comparison families from Victoria and southern New South Wales.

Measures

Demographic measures

Demographic data were collected from mothers during the study visit. The demographic variables were chosen as standard descriptive variables, together with variables of interest in relation to proxy measures of parents' relative socioeconomic status (for example educatonal level and smoking status) and interest in parenting. These included:

- maternal age and highest level of education completed

- marital status
- paternal age and highest level of education completed
- number of smokers living in the household
- attendance at a parenting course
- participation in family counselling and, if so, whether sibling issues were a feature of the counselling.

Clinical measures

The following clinical data were obtained from the RCH's records for the children with CF who participated in the study: best measure of lung function in the six months prior to the study visit, FEV_1, and total number of days spent in hospital in the 12 months prior to the study visit.

School absence

During the study visit, written parental permission was obtained to contact the younger children's schools in both groups to assess the number of absences from school during the previous school year. These data were unavailable for several students because their schools had changed their system of data storage and the data could not be retrieved.

Open-ended questions

Older siblings in the CF group were asked four questions from a pilot study conducted by Russo and Hogg (2004). These were:

- What happens to you when your brother (or sister) goes into hospital?
- What are three good things about having a brother (or sister) with CF?
- What are the three things that make it difficult having a brother (or sister) with CF?
- What would make things better or easier for you?

The verbal responses to these questions were recorded directly onto paper by the researcher at the time that each participant was interviewed. Data analysis was done manually using thematic content analysis (Burnard, 1991); transcripts were each analysed independently by myself and one of my supervisors (Professor Susan Sawyer). A familiarisation process was first conducted by reading and re-reading the responses to each of the four questions and deciding on the common themes. Both raters then independently coded each response using the agreed upon themes. Where anomalies were noted or disagreements occurred, transcripts of the responses were re-examined until consensus was reached. The use of these questions (rather than exploratory interviews), together with the age of the respondents, resulted in a decision to only report major themes.

Quality of the sibling relationship

The SRQ (Furman & Buhrmester, 1985) was used to assess the perceived quality of the relationship between the target children and their younger siblings. Permission was obtained from the author to use this measure, as shown in Appendix B. The SRQ is a 48-item, self-report questionnaire designed to measure four factors pertaining to qualities of the sibling relationship: warmth/closeness, relative status/power, conflict and rivalry. These four factors were extracted from 16 scales: intimacy, prosocial, companionship, similarity, nurturance by sibling, nurturance of sibling, admiration by sibling, admiration of sibling, affection, dominance by sibling, dominance of sibling, quarrelling, antagonism, competition, maternal partiality and paternal partiality. Scores are computed for each of the 16 scales by averaging the three items designed to assess that scale. Young people are asked to respond according to a five-point Likert format from “hardly at all” to “extremely much.” The authors reported an average internal consistency coefficient (Cronbach’s α) across scales of .80. Test-retest reliability was moderate to high, with a mean of $r = .71$, ranging from .58 to .86 (Furman & Buhrmester, 1985).

Target siblings' adjustment

Several aspects of the target siblings' adjustment were measured: depression, social skills and behaviour problems.

Symptoms of depression were assessed using the Childhood Depression Inventory (CDI) (Kovacs, 1981). This is a 27-item self-report measure designed to assess the occurrence of cognitive, affective and behavioural symptoms of depression. For each question, children had 3 choices of sentences that described their thoughts, feelings, and actions during the preceding two weeks. Questions were scored on a 0-2 scale, and higher scores represented the presence of a greater number of depressive symptoms. The CDI yields both a total score and scores on five subscales: negative mood, interpersonal problems, ineffectiveness, anhedonia and negative self-esteem. This scale is appropriate for children ranging in age from 7 to 17 years and has been extensively normed. Adequate test-retest reliability (*r*'s .41 to .69 over a one year period) and internal consistency (Cronbach α coefficients of .70 to .89) have been reported (Kovacs, 1981). The CDI distinguishes between depressed and non-depressed children and is sensitive to treatment effects.

The Social Skills Rating System (SSRS) (Gresham & Elliott, 1990) was used to assess the target siblings' social competencies and problem behaviours. The SSRS-Student is a self-report questionnaire designed to assess social behaviours and skills in the domains of cooperation, assertion, empathy and self-control. A 34-item elementary version of this questionnaire was used with primary school age children and a 39-item version was used for children in secondary school. On this measure, children were asked to rate the frequency with which they engaged in various social behaviours (for example, "I make friends easily", "I disagree with adults without fighting or arguing") using a 3-point scale that ranged from "never" to "very often".

In addition to frequency ratings, importance ratings were completed by older students (in Years 7 to 12). Students rated each behaviour according to its perceived importance for their relationship with others. The problem behaviours domain assesses externalising problems, internalising problems and hyperactivity.

Items were scored on a 0-2 scale, with higher scores on the social skills scale indicating greater social skills and higher scores on the behaviour problem scale indicating more behavioural problems. Total scores on these measures were converted to standard scores, provided in the scoring manual, to allow for comparability between different versions of this measure.

The student version of this measure has demonstrated adequate internal consistencies (α =.83) and four-week test-retest reliability (*r*=.68). There is strong evidence supporting both the content and criterion-oriented validity of this instrument (Gresham & Elliott, 1990). Parent and teacher versions of the SSRS are also available but were not used in this study.

Parental differential treatment

Parental differential treatment of younger and older siblings was evaluated by examining parents' daily activity patterns using the DPD software (Quittner & Opipari, 1994). The DPD employs a cued-recall procedure that systemically tracks mothers and fathers through all their activities and interactions over the 24-hour period preceding the phone call. For all activities lasting five minutes or longer, parents were asked to report the type of activity, its duration in minutes and who was present. The interviewer facilitated the reconstruction of the mothers' and fathers' activities by providing prompts, such as information about a previous behaviour (for example, "After you finished breakfast, what did you do next?"). Using this procedure, moderate to high correlations have been found across days for time spent in various activities (*r* = .40 to .82) and with various companions (*r* = .53 to .71) over a period of three weeks (Quittner & Opipari, 1994). The DPD has also been found to demonstrate appropriate validity including test retest reliability (Quittner & Opipari, 1994), inter-rater reliability greater than 90%, (Quittner et al., 1998) and convergent criterion validity when compared with electronic monitoring (Quittner, Modi, Lemanek, Ievers-Landis, & Rapoff, 2008).

Mothers and fathers were surveyed separately for the same 24-hour period on three occasions (two weekdays, one weekend day), as outlined in the DPD manual (see

Appendix C). Reported activities were placed into categories that represented common daily routines. A sample of the DPD data entry screen is shown in Figure 6. These activities included basic child care, medical care, household tasks, meals (preparation and eating), recreation (at home and externally), work, school (parent learning), self-care, rest, sleep, and other.

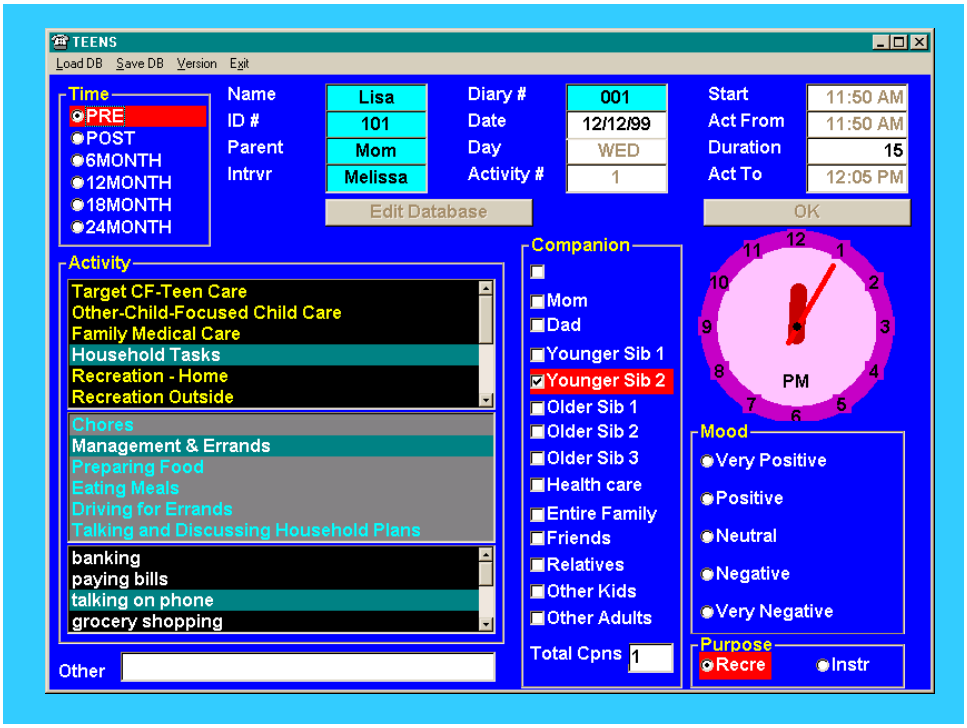

Figure 6. Daily Phone Diary software data entry screen.

Table 5 summarises the measures completed by the study participants in both the CF and Comparison groups. A copy of each measure used in this study is contained in Appendix D.

Table 5

Measures Completed by the CF and Comparison groups

	CF group				Comparison group			
Measure	Younger child	Older child	Mother	Father	Younger child	Older child	Mother	Father
Demographics			✓				✓	
Daily Phone Diary			✓	✓			✓	✓
Child Depression Inventory		✓				✓		
Social Skills Rating System		✓				✓		
Sibling Relationship Questionnaire	✓	✓			✓	✓		
Four open-ended questions		✓						
School absence in the previous year	✓				✓			
Number of days in hospital in previous year	✓							
Best lung function (FEV_1) in the previous 6 months	✓							

RESULTS

Statistical Approach

First, the protocol for entering and cleaning the data is described. Second, the data were checked and evaluated for normality and skew. Next, the analytic plan for testing each of the four hypotheses are presented, with tabulated descriptive statistics followed by the relevant inferential tests. Finally, the process for coding and analysing the open-ended questions is described.

Quantitative data were analysed using SPSS Version 22. For the first hypothesis, which assessed parental differential treatment, the primary analysis was a three-way mixed analysis of variance (ANOVA). The second hypothesis involved a series of correlational analyses that examined the associations between parental differential treatment and measures of emotional distress, social skills and behaviour problems in the older siblings, using the CDI and SSRS. The same analytic framework was applied to the third hypothesis, which evaluated the relationship between parental differential treatment and sibling relationship quality, using the SRQ. Finally, the fourth hypothesis, utilising the open-ended, qualitative data is reported. The responses to four open-ended questions relating to the sibling experience were categorised into common themes using consensus coding.

Data Entry

All data were entered into a single SPSS spreadsheet. The variables were coded as: (1) demographics (for example, age, gender), (2) illness severity measures (for example, lung function), (3) SRQ sub-scale and total scores, (4) CDI sub-scale and total scores, (5) SSRS sub-scale and total scores, and (6) DPD data.

Initial Data Cleaning and Distribution of Variables

All variables were initially analysed using exploratory data analysis to: (1) identify data entry errors, (2) identify any notable outliers, (3) indicate potentially important patterns of results at the descriptive level, and (4) test assumptions underlying parametric

procedures. The process included: visual inspection of stem-and-leaf plots, histograms and normality plots; consideration of significance tests (for example, K-S procedure and Levene test) associated with the assumptions of normality and homogeneity of variance; scatterplots and measures of both skewness and kurtosis.

No outliers of concern were identified for any variable. Although some variables exhibited some skewness, the level of skewness was not considered significant enough to warrant either data transformation or use of non-parametric procedures. In no case was the assumption of homogeneity of variance violated, and the assumption of independence of error was met for all variables.

Results of the study

Hypothesis 1: Evidence of parental differential treatment

The first aim of the study was to examine the magnitude and type of differential treatment occurring among families with and without a child with CF. Differential treatment by mothers and fathers in favour of the younger child was expected in both types of families, as assessed by the DPD. However, a greater magnitude of differential treatment by mothers and fathers was expected in the CF versus Comparison group.

The types of activities engaged in by mothers and fathers in both groups of families were also examined. Given that mothers are typically the primary caregivers for their child with CF, they were expected to spend more time in medical activities than fathers in the CF group. After excluding time spent in medical care activities, mothers in the CF group were still predicted to spend more time with the younger child in activities, such as meal times, due to the importance of nutrition in the management of CF.

DPD data were excluded for four families. In two cases, the data were collected during an atypical period; in one case after a double lung transplant and in the other when a child was in plaster. The other two families met the eligibility criteria for the study but had living arrangements affected by schooling; in one family the older child attended boarding school and in the other family, the mother and children lived in town during

the week (away from the family farm). Thus, the target siblings were not living with the entire family. In all four cases, the families had been enrolled in the study and study visits had been completed. The need to exclude the DPDs for the latter two families could have been avoided by more careful initial screening.

Collection of the DPDs was mostly straightforward. However, the research assistant administering the DPDs reported difficulty collecting phone diaries from six families. No DPD data were collected from four CF families and one Comparison family, despite numerous attempts to arrange suitable times. Eventually, these families declined to participate in this aspect of the study. In one other Comparison family, DPD data were collected from the mother, but the father declined to participate. Complete data were collected from 30 of the 39 CF families and 24 of the 29 Comparison families.

Descriptive statistics for all of the DPD measures are shown in Table 6 and are presented graphically in Figures 7 - 18. It is worth noting that fathers in the CF group spent twice as much time with the younger child compared to the older child during the three-day period when the DPD data were collected. This was also the case when time spent in medical care was excluded from the record of Total Time and for all separate activities (for example, meal times). In contrast, fathers in the Comparison group spent a similar amount of time with both their younger and older child.

Mothers from both groups also spent more time with the younger than the older child. This was the case for all activities except time spent in medical care. Mothers in the Comparison group spent no time in medical care with either child. In contrast, mothers in the CF group spent an average of 18 minutes in medical care with the younger child (CF) compared to 2 minutes with the older child.

Mothers in the CF group spent 5 more minutes on medical care with the younger child (CF) than fathers in the CF group. In contrast, mothers in the CF group spent less time in recreational activities with the younger child with CF than the father.

Table 6

Mean Parental Time (Minutes per Day) Spent with Younger and Older Siblings by Group (SD in parentheses)

	Mother				Father			
	CF		Comparison		CF		Comparison	
	Younger	Older	Younger	Older	Younger	Older	Younger	Older
Individual Time	53.33 (66.55)	21.11 (26.95)	43.19 (48.57)	12.92 (17.50)	40.56 (51.62)	20.33 (27.39)	22.64 (40.25)	25.97 (30.72)
Total Time	217.02 (107.74)	198.69 (116.12)	212.69 (88.77)	201.47 (83.85)	151.44 (76.54)	134.94 (82.72)	163.19 (75.82)	155.84 (74.23)
Individual Time Excluding Medical Care	36.72 (47.87)	20.40 (26.92)	43.19 (48.57)	12.92 (17.50)	38.28 (52.42)	19.71 (28.06)	22.64 (40.25)	24.31 (30.16)
Total Time Excluding Medical Care	215.20 (107.94)	197.63 (116.99)	211.15 (87.75)	199.93 (82.50)	150.78 (76.61)	134.94 (82.72)	162.50 (75.85)	155.14 (74.19)
Individual Time Spent in Meal Times	4.61 (9.43)	2.44 (6.30)	2.50 (6.08)	0.56 (2.72)	3.89 (8.00)	0.89 (4.87)	2.71 (6.08)	2.78 (7.80)
Individual Time Spent in Household Tasks	6.72 (13.73)	1.06 (2.85)	14.93 (38.95)	0.00 (0.00)	6.28 (31.10)	2.61 (12.03)	0.00 (0.00)	5.42 (17.93)
Individual Time Spent in Recreation	9.78 (24.85)	6.22 (16.53)	10.21 (24.00)	0.56 (2.72)	14.44 (27.67)	11.00 (24.79)	8.61 (20.29)	5.42 (11.70)
Individual Time Spent in Child Care	14.88 (21.73)	9.66 (14.89)	15.56 (16.49)	11.81 (17.55)	13.68 (27.69)	5.63 (8.78)	11.32 (25.51)	10.70 (17.17)
Individual Time Spent in Medical Care	18.45 (28.45)	2.13 (6.47)	0.00 (0.00)	0.00 (0.00)	3.68 (9.52)	0.40 (2.17)	0.00 (0.00)	1.67 (5.65)

It is also worth noting that several parents in each group reported spending no individual time with the younger or older child during the three-day period when the DPD data were collected. These figures are reported in Table 7. A similar number of mothers and fathers across both groups spent no individual time with the younger or older child.

Table 7

Number of Parents in Each Group Reporting Spending 0 Minutes of Individual Time with Either Child

MOTHER			
CF		COMPARISON	
Younger	Older	Younger	Older
7	12	6	11
FATHER			
CF		COMPARISON	
Younger	Older	Younger	Older
12	9	12	10

Diary Results

DPD data were analysed using a 2 x 2 x 2 mixed ANOVA. The between-subjects factor was group (CF versus Comparison). The two within-subjects factors were parent (mother, father) and sibling (younger, older). The hypotheses being tested were: (1) differential treatment by both mothers and fathers would be found in favour of the younger child in both types of families, (2) a greater magnitude of differential treatment by both mothers and fathers would be found in the CF group, and (3) these findings would be consistent across conditions involving children spending individual time with parents and when time was spent with both the target child and others (i.e. total time). Mixed support was found for these hypotheses.

Initially, the nine DPD variables reported in Table 6 were entered into a single multivariate analysis, using the design described in the previous paragraph. Two significant multivariate main effects were evident; no significant interactions were found. There was a significant multivariate main effect for Parent, $\Lambda = .60$, $F(1, 43) = 3.19$, $p = .005$, $\eta^2 = .40$, and for Sibling, $\Lambda = .59$, $F(1, 43) = 3.39$, $p = .003$, $\eta^2 = .42$. The analyses reported below present the follow-up univariate results.

For individual time, a main effect was found for siblings, $\Lambda = .78$, $F(1, 52) = 14.71$, $p < .001$, $\eta^2 = .22$, with estimated marginal means indicating that younger children spent nearly twice as much time alone (individual time) than their older siblings, across both groups and both types of parents (see Figure 7) except for fathers in the Comparison group who spent a similar amount of time with younger and older children.

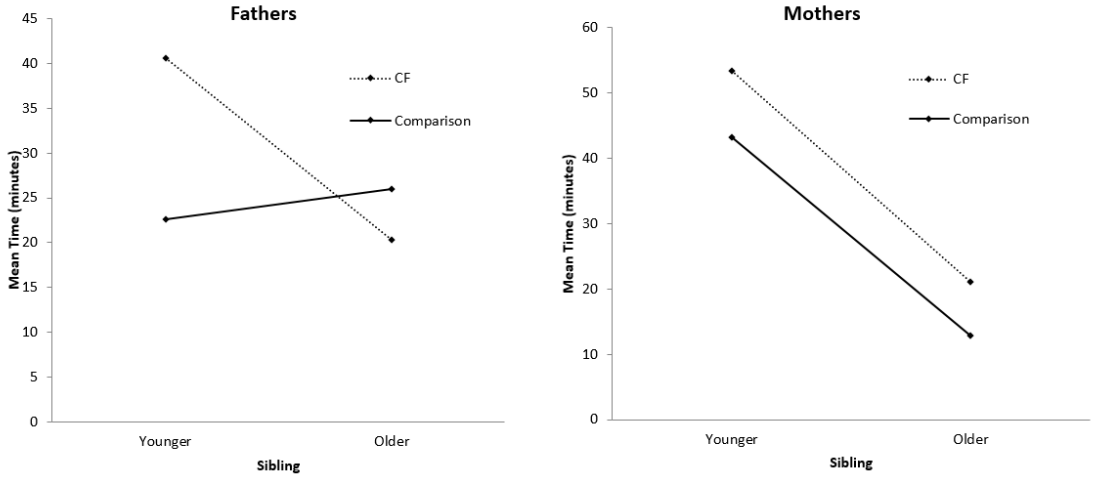

Figure 7. Sibling by Group interactions for mothers and fathers separately for individual time.

This pattern of results did not change when the analysis was repeated excluding Medical Time, $\Lambda = .82$, $F(1, 51) = 11.52$, $p = .001$, $\eta^2 = .18$. Younger children, regardless of whether they had CF or not, spent more individual time with both of their parents than older children (see Figure 8) except for fathers in the Comparison group who spent similar times with younger and older children.

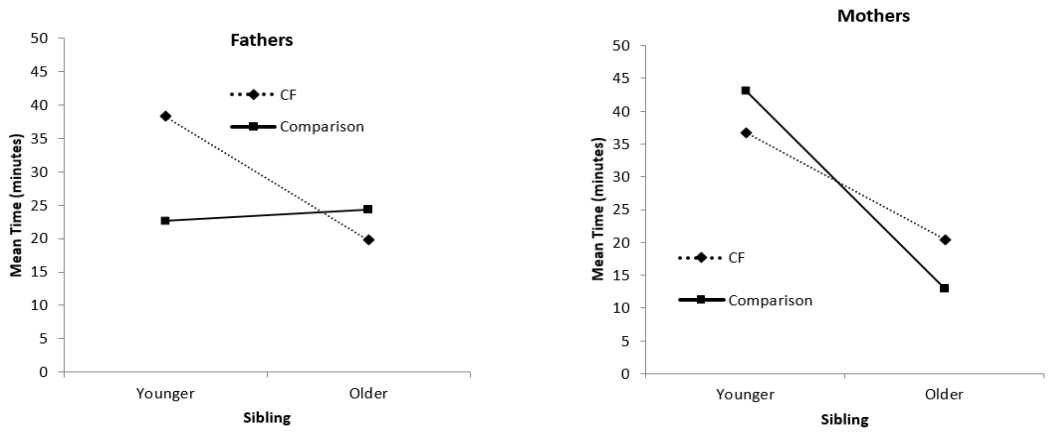

Figure 8. Sibling by Group interactions for mothers and fathers separately for individual time excluding medical care.

Despite the fact that the Parent x Group x Sibling three-way interaction was not statistically significant, visual inspection of the Group x Sibling interaction for mothers in comparison to fathers revealed an interesting pattern (see Figure 7); specifically, there was evidence of parental differential treatment by fathers but not mothers. For mothers in a family with a child with CF, there was a small but consistently greater amount of individual time spent with both younger and older children in comparison to mothers in a family without a child with CF. For fathers, the presence of a child with CF in the family had a more pronounced effect. There was a larger and more obvious difference in the amount of individual time fathers spent with younger siblings with CF (17.92 minutes). There was no notable difference in the amount of time that fathers spent with older siblings (5.64 minutes), regardless of the presence of a child with CF. For mothers in the CF group, the difference in individual time spent with younger and older siblings was very similar (10.14 minutes for younger siblings, 8.19 minutes for older siblings). It should be noted that these trends are descriptive only and not statistically significant.

The same analysis was used to compare the time spent with the target child and other people (total time). A significant main effect for siblings was found, $\Lambda = .81$, $F(1, 52) = 11.85$, $p = .001$, $\eta^2 = .19$, with younger children receiving more time than their older

siblings across both groups and both parents. In addition, a significant main effect for parents was found, $\Lambda = .78$, $F(1, 52) = 14.80$, $p < .001$, $\eta^2 = .22$, indicating that mothers, across both groups and both siblings, spent more time than fathers with children when others were present (see Figure 9).

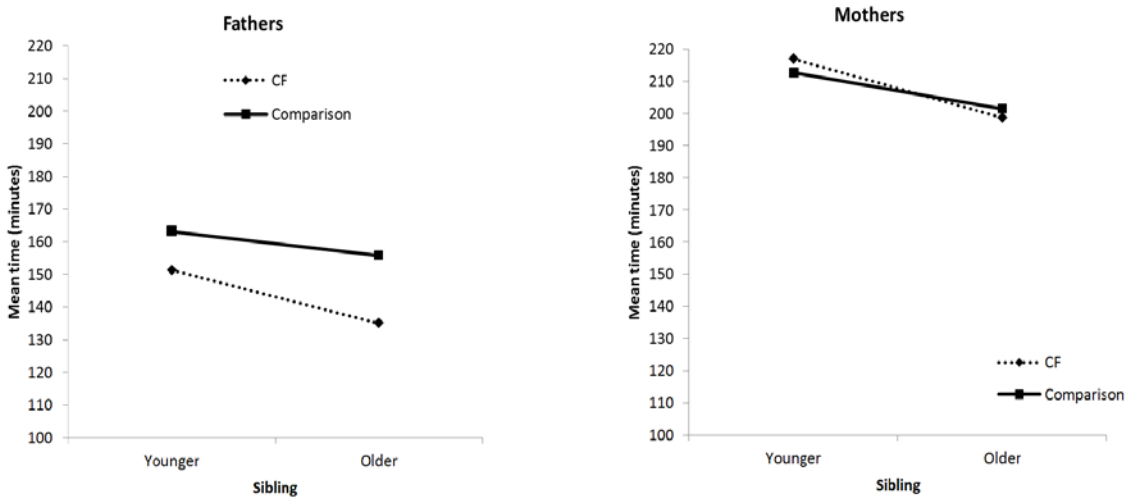

Figure 9. Sibling by Group interactions for mothers and fathers separately for total time.

When this analysis was repeated excluding medical care, an almost identical pattern of main effects was evident: a significant sibling main effect, $\Lambda = .81$, $F(1, 52) = 11.88$, $p = .001$, $\eta^2 = .19$, and a parent main effect, $\Lambda = .79$, $F(1, 52) = 14.25$, $p < .001$, $\eta^2 = .22$ (see Figure 10).

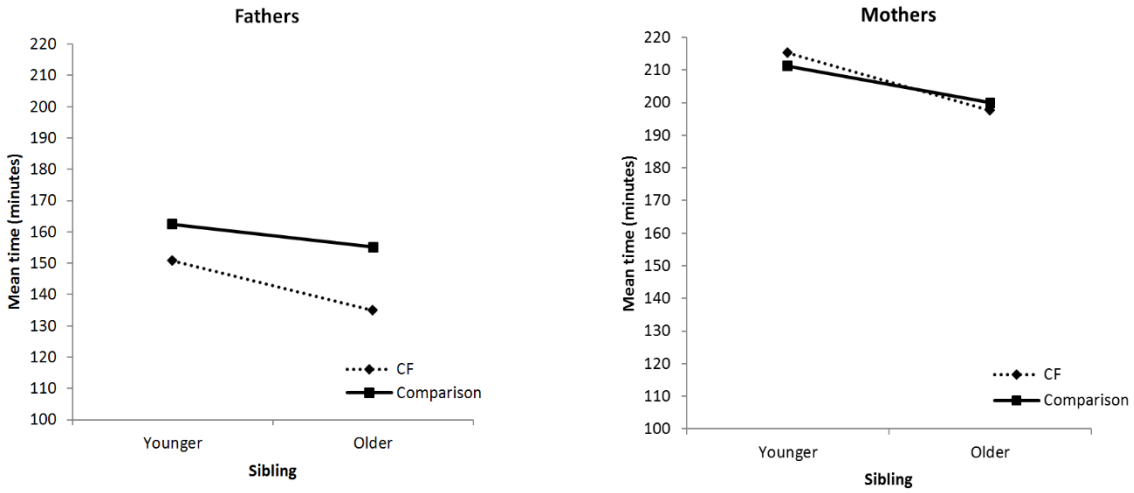

Figure 10. Sibling by Group interactions for mothers and fathers separately for total time excluding medical care.

Next, the time spent in meals was analysed (see Figure 11). A significant main effect for siblings was found, $\Lambda = .89$, $F(1, 52) = 6.27$, $p = .015$, $\eta^2 = .11$, indicating that parents (both mothers and fathers) spent over twice the amount of time with younger siblings (Younger: $M_{minutes} = 3.43$; Older: $M_{minutes} = 1.67$) in mealtime activities except for fathers in the Comparison group who again spent a similar amount of time with younger and older children.

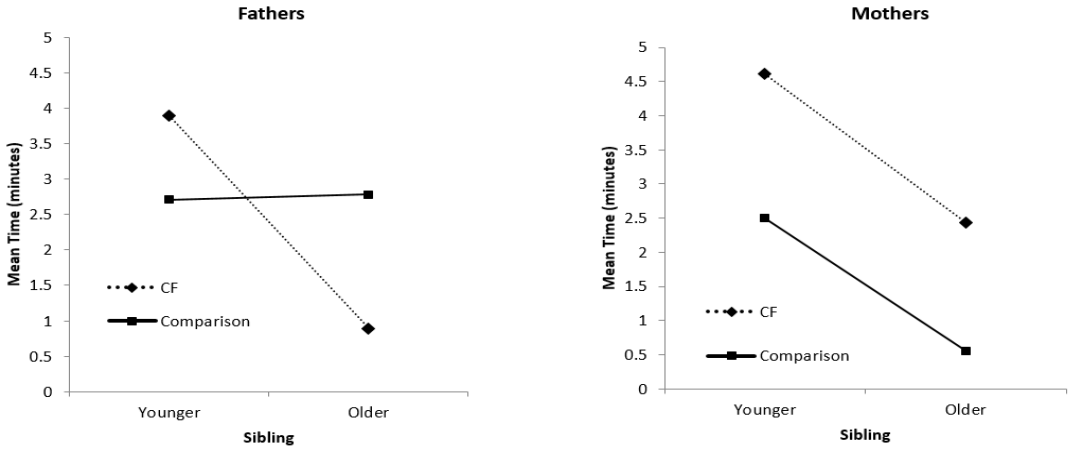

Figure 11. Sibling by Group interactions for mothers and fathers separately for individual time spent in meal times.

Analyses of time spent in recreation and child care activities (see Figures 12 and 13) show similar trends. Again, fathers and mothers spent more time with younger children for both activities. Fathers in the Comparison group spent similar amounts of time in child care activities with younger and older children (see Figure 13).

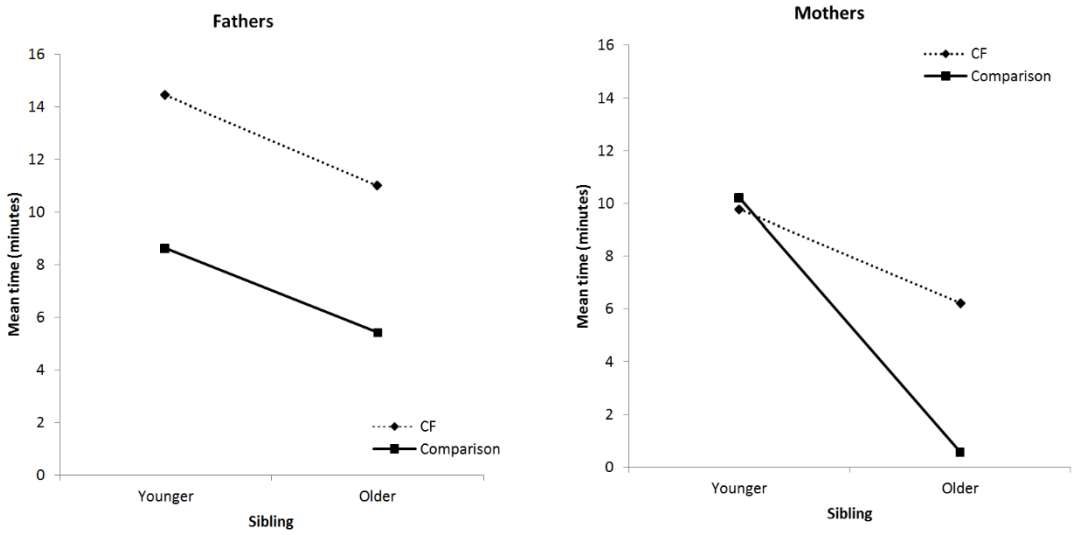

Figure 12. Sibling by Group interactions for mothers and fathers separately for individual time spent in recreation.

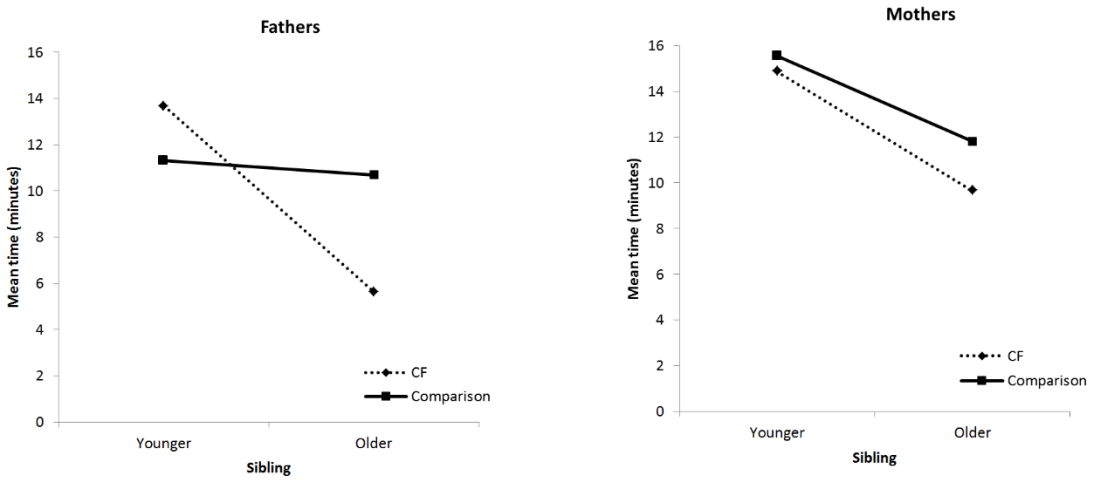

Figure 13. Sibling by Group interactions for mothers and fathers separately for individual time spent in child care.

The analysis of time spent in household tasks revealed a significant parent x sibling interaction, $\Lambda = .91$, $F(1, 52) = 5.01$, $p = .03$, $\eta^2 = .09$, which is illustrated in Figure 14, and reveals that across both the CF and Comparison groups, there was little difference in the amount of time fathers spent with younger and older siblings, whereas for mothers, considerably more time was spent with younger siblings. In fact, mothers spent less time in household tasks with older siblings than fathers, which is somewhat surprising given the results from other specific activities.

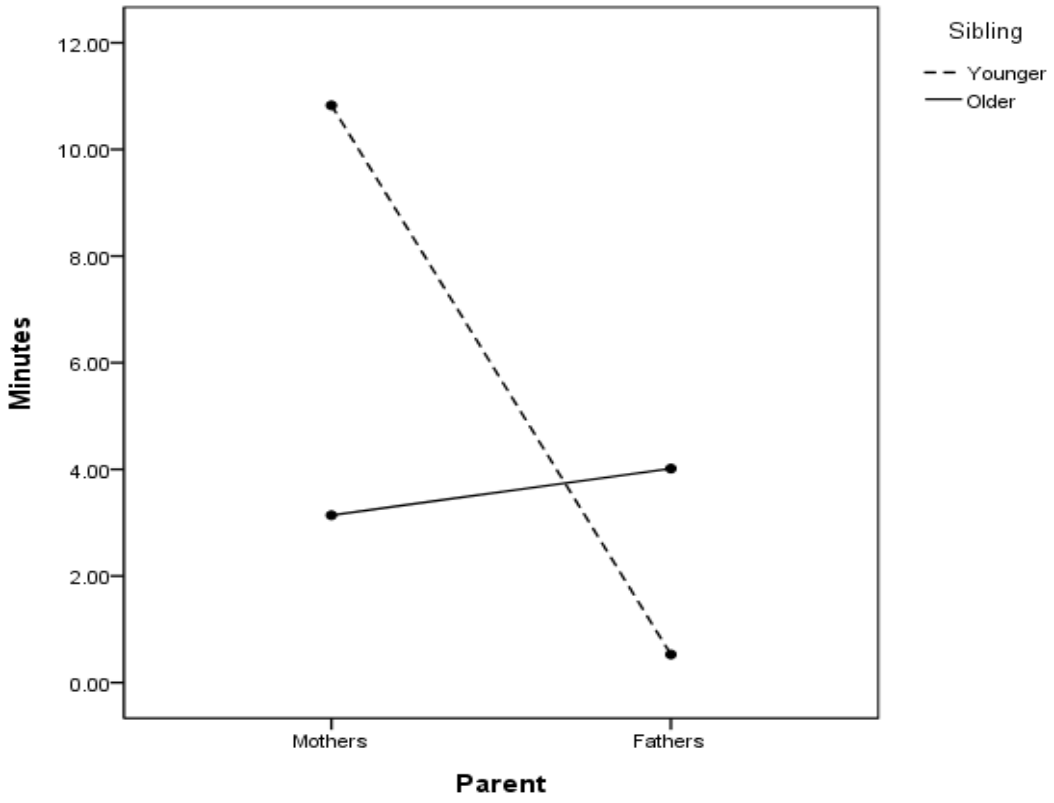

Figure 14. Parent by Sibling interaction for time spent in household tasks.

The analysis of time spent in medical care revealed several important findings. The specific hypothesis being tested was that mothers were expected to spend more time in medical care than fathers in the CF group (see Figure 15).

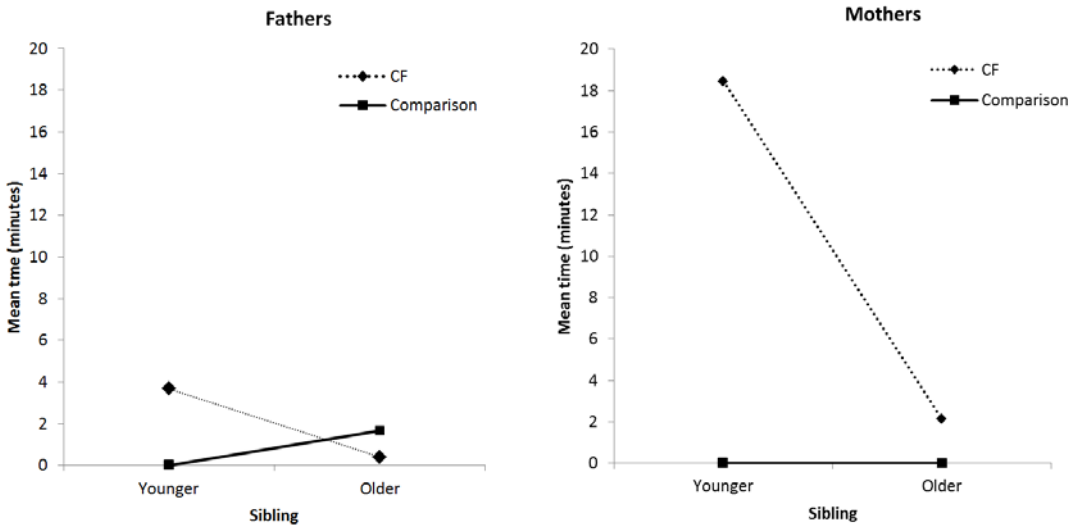

Figure 15. Sibling by Group interactions for mothers and fathers separately for individual time spent in medical care.

This analysis revealed the highest number of significant results, with only the three-way interaction failing to reach significance. There were significant main effects for Parent, $\Lambda = .92$, $F(1, 51) = 4.44$, $p = .040$, $\eta^2 = .08$, Sibling, $\Lambda = .84$, $F(1, 51) = 9.66$, $p = .003$, $\eta^2 = .16$, and Group, $F(1, 51) = 12.02$, $p = .001$, $\eta^2 = .19$. Inspection of the marginal means revealed that mothers ($M_{minutes} = 5.14$) spent more time on medical care than fathers ($M_{minutes} = 1.44$), younger children ($M_{minutes} = 5.53$) received more medical care than older children ($M_{minutes} = 1.05$), and, not surprisingly, children in the CF group spent more time in medical care ($M_{minutes} = 6.16$) than children in the Comparison group ($M_{minutes} = 0.42$).

Of greater interest, however, were the three significant two-way interactions: Group by Parent, $\Lambda = .88$, $F(1, 51) = 6.66$, $p = .013$, $\eta^2 = .12$ (Figure 16); Sibling by Group, $\Lambda = .79$, $F(1, 51) = 13.59$, $p = .001$, $\eta^2 = .21$ (Figure 17); and Sibling by Parent, $\Lambda = .89$, $F(1, 51) = 6.03$, $p = .018$, $\eta^2 = .11$ (Figure 18). Examination of the Group by Parent interaction indicated that this was due mainly to the large amount of time mothers spent with younger children with CF in medical care; for fathers, there was very little difference in time spent in medical care between the two groups. This effect was found across both sibling groups, and provided the most direct support for the hypothesis.

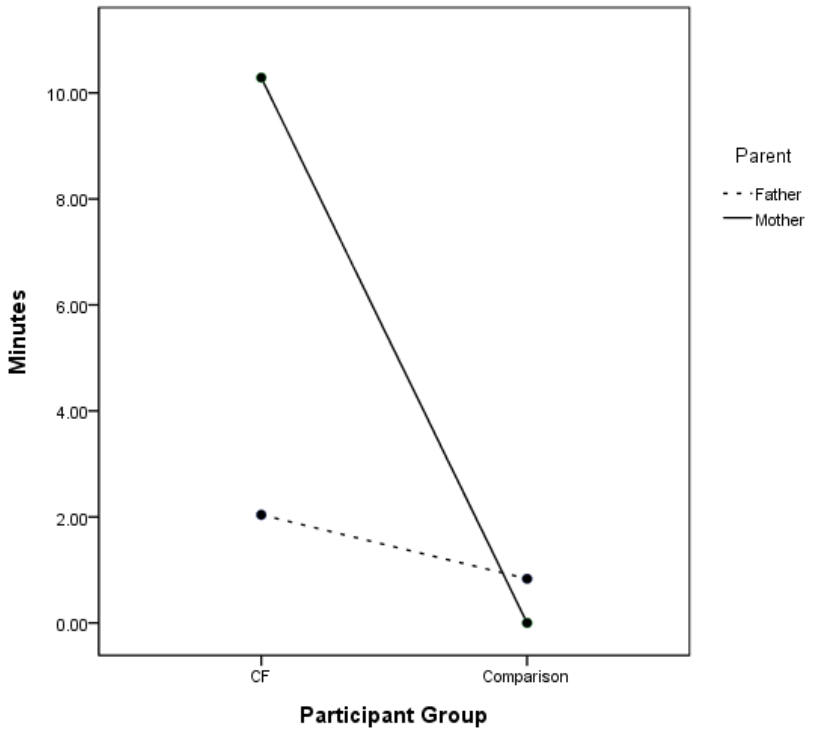

Figure 16. Group by Parent interaction for time spent in medical care.

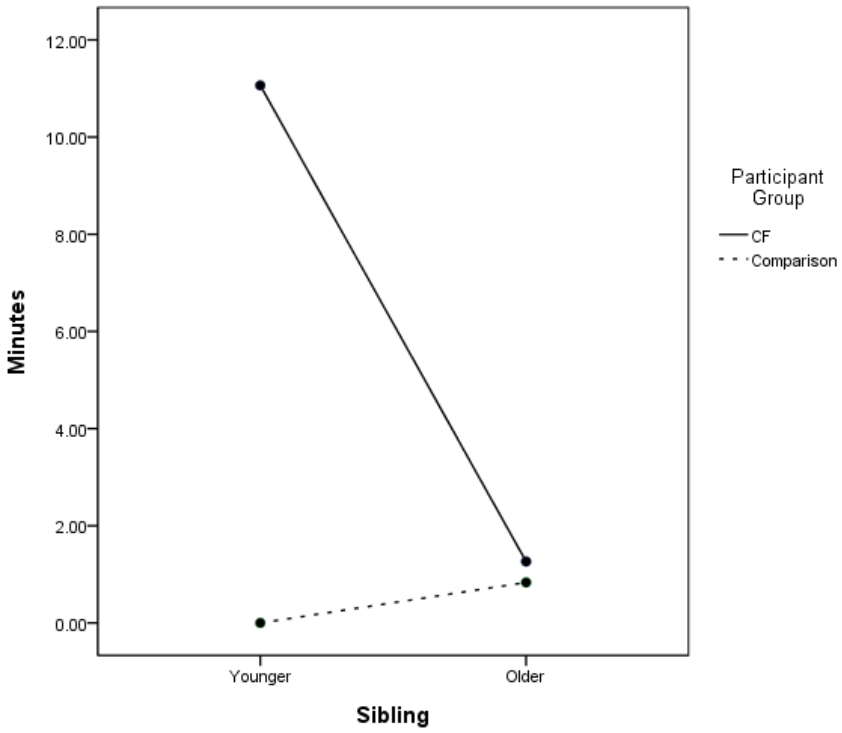

Figure 17. Sibling by Group interaction for time spent in medical care.

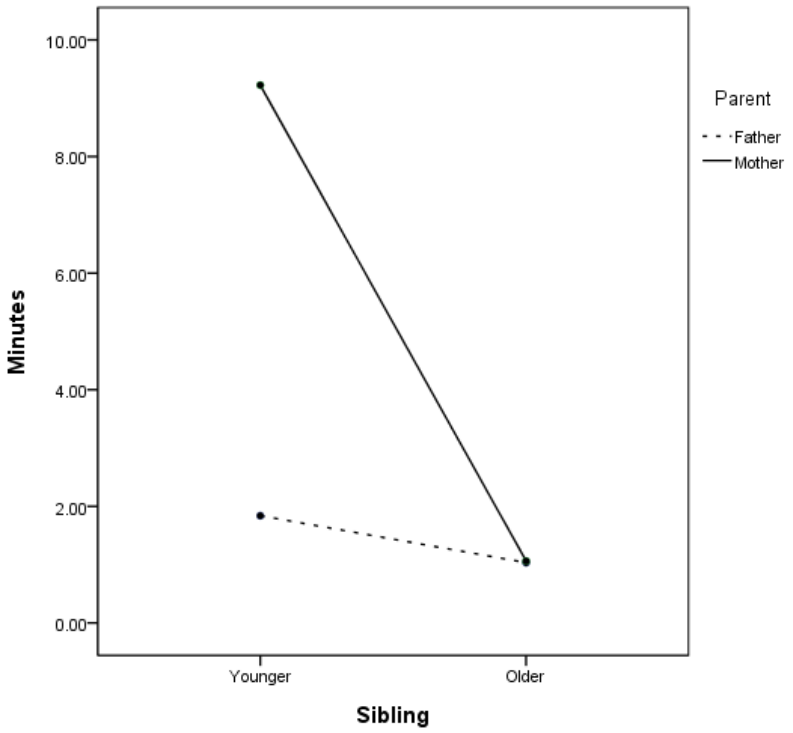

Figure 18. Sibling by Parent by interaction for time spent in medical care.

A similar pattern was seen for the Sibling by Parent interaction - very little difference across the sibling groups for fathers, but a considerable difference for mothers, with much more time being spent with younger siblings. This result was across both CF and Comparison groups. For the Sibling by Group interaction, not surprisingly, there was very little difference amongst three of the cells in the interaction. Only the large amount of time spent on medical care with younger siblings in the CF group (i.e., the children CF) diverged from that pattern. This finding was across both parent groups.

Hypothesis 2: Relationship between parental differential treatment and child outcome measures

Parental differential treatment was calculated by subtracting individual time spent alone with the older sibling from individual time spent alone with the younger sibling, for mothers and fathers in both groups. These parental differential treatment scores were then correlated with the child outcome variables separately for the CF and Comparison

groups to test the hypothesis that parental differential treatment affects the social and emotional adjustment for the older sibling in a family caring for a child with CF.

Children's outcomes included depression on the CDI, behavioural and social adjustment on the SSRS factor and subscale scores. Minimal support was found for this hypothesis. Descriptive statistics for parental differential treatment and the CDI and SSRS sub-scales are shown in Table 8. All CDI sub-scale means and the CDI total score mean were slightly, but not significantly, below the corresponding normative values for both the CF and Comparison groups (Kovacs, 1992); however, the sample means for the SSRS sub-scales and total score were consistently higher than the normative means (Gresham & Elliott, 1990) for both groups, although again these difference were not statistically significant. There were no statistically significant differences between the older siblings in the CF and Comparison groups on any of the CDI and SSRS total and sub-subscale scores.

Table 8

Descriptive Results for Parental Differential Treatment with the Older Sibling and the SSRS and CDI Sub-Scales

	Participant Group					
	CF			Comparison		
	n	*M*	*SD*	*n*	*M*	*SD*
Fathers' Differential Treatment[a]	30	20.22	61.80	24	-3.33	53.98
Mothers' Differential Treatment[a]	33	34.19	65.91	26	29.10	52.05
SSRS Cooperation	39	14.54	3.63	29	14.72	2.91
SSRS Assertion	39	14.38	2.85	29	15.10	2.50
SSRS Empathy	39	16.54	2.58	29	16.72	3.01
SSRS Self-Control	39	12.10	3.23	29	12.52	2.28
SSRS Total Score	39	102.46	22.63	29	112.07	13.91
CDI Negative Mood	39	2.08	2.50	29	1.66	1.74
CDI Interpersonal Problems	39	0.64	1.11	29	0.38	0.82
CDI Ineffectiveness	39	1.28	1.49	29	1.07	1.36
CDI Anhedonia	39	2.69	2.44	29	2.10	2.23
CDI Negative Self Esteem	39	1.33	1.74	29	0.93	1.19
CDI Total Score	39	8.03	7.37	29	6.14	6.20

Note: [a] minutes.

Only one significant correlation was observed, which involved fathers' differential treatment and the SSRS Self-Control scale for the CF group, r (N = 30) = -.38, p = .04, indicating that high levels of paternal differential treatment for children with CF were associated with lower levels of self-control in the older sibling. Thus, only minimal support was provided for hypothesis 2.

Hypothesis 3: Relationship between parental differential treatment and sibling relationship quality

The same approach to the analysis of the variables for Hypothesis 2 was applied to the analysis of the relationship between parental differential treatment and sibling relationship quality on the SRQ. Greater level of parental differential treatment in favour of the younger sibling was expected to be associated with worse sibling relationship quality. This hypothesis was partially supported for fathers. Further, siblings in the CF group were expected to report worse sibling relationship quality than siblings in the Comparison group. As reported below, limited support was found for this hypothesis; specifically, a small number of age-related main effects were identified, but no significant differences were found for the CF group.

Descriptive statistics for the SRQ sub-scales are shown in Table 9.

Table 9

Descriptive Statistics for the SRQ Sub-Scales

	Participant Group					
	CF			Comparison		
	n	*M*	*SD*	*n*	*M*	*SD*
Younger						
Rivalry	38	3.03	0.32	29	2.97	0.19
Relative Power	38	-0.21	0.31	29	-0.36	0.36
Warmth	38	2.90	1.08	29	3.16	0.83
Conflict	38	2.79	0.86	29	2.92	0.77
Prosocial Behaviour	38	2.85	1.07	29	3.09	0.82
Maternal Partiality	38	3.05	0.51	29	3.03	0.40
Nurturance Of Sibling	38	2.48	0.99	29	2.41	0.72
Nurturance By Sibling	38	2.97	1.15	29	3.09	1.00
Dominance Of Sibling	38	1.92	0.80	29	2.00	0.58
Dominance By Sibling	38	2.25	0.94	29	2.77	0.95
Paternal Partiality	37	3.01	0.43	29	2.90	0.25
Affection	38	3.40	1.27	29	3.83	0.88
Companionship	38	3.04	1.23	29	3.17	1.08
Antagonism	38	2.84	1.08	29	2.93	0.88
Similarity	38	2.72	1.03	29	2.92	0.86
Intimacy	38	2.34	1.14	29	2.41	1.17
Competition	38	2.32	1.09	29	2.70	1.06
Admiration Of Sibling	38	3.16	1.26	29	3.54	0.92
Admiration By Sibling	38	2.77	1.38	29	3.17	1.10
Quarrelling	38	3.19	1.08	29	3.11	0.93
Older						
Rivalry	39	2.75	0.50	29	2.89	0.36
Relative	39	0.34	0.36	29	0.32	0.33

Warmth	39	3.13	0.98	29	3.15	0.77
Conflict	39	2.95	0.89	29	2.99	0.67
Prosocial Behaviour	39	3.00	0.97	29	3.13	0.62
Maternal Partiality	39	2.71	0.58	29	2.77	0.57
Nurturance Of Sibling	39	3.44	0.92	29	3.48	0.66
Nurturance By Sibling	39	2.39	0.99	29	2.70	0.95
Dominance Of Sibling	39	2.80	1.00	29	2.91	0.79
Dominance By Sibling	39	2.47	0.93	29	2.43	0.71
Paternal Partiality	38	2.77	0.59	29	3.01	0.58
Affection	39	3.74	1.25	29	3.95	0.98
Companionship	39	3.19	1.15	29	3.22	1.07
Antagonism	39	2.99	1.08	29	3.04	0.65
Similarity	39	2.94	1.09	29	2.85	1.07
Intimacy	39	2.34	1.07	29	2.29	1.17
Competition	39	2.67	1.12	29	2.61	0.98
Admiration of Sibling	39	3.44	1.25	29	3.34	0.89
Admiration By Sibling	39	3.24	1.15	29	3.29	0.95
Quarrelling	39	3.19	0.94	29	3.31	0.88

No significant correlations between parental differential treatment and SRQ scores were found, for either mothers or fathers, in the Comparison group; however, several significant correlations were found for the CF group for fathers' differential treatment. Six significant associations were found: four for the older siblings and two for the younger siblings (i.e., those with CF). The significant correlations with fathers' parental differential treatment were, in order of magnitude: Maternal Partiality (Older), r (N = 30) = -.53, p = .002; Rivalry FS (Older), r (N = 30) = -.48, p = .008; Nurturance by Sibling (Older), r (N = 30) = -.40, p = .028; Quarrelling (Younger), r (N = 29) = .39, p = .039; Competition (Older), r (N = 30) = .37, p = .043; Paternal Partiality (Younger), r (N = 29) = .37, p = .05. High levels of paternal differential treatment for children with CF were associated with the following SRQ subscales for the older sibling: lower levels of maternal partiality, higher levels of rivalry, lower levels of nurturance by the younger sibling and higher levels of competition. For the younger sibling with CF, high levels of

paternal differential treatment were associated with higher levels of quarrelling and paternal partiality.

Analysis of SRQ Factors

A series of 2 x 2 mixed ANOVAs, with group (CF and Comparison) and age (younger, older sibling) as the between subject factors, were conducted. Only two significant results emerged from these analyses - significant age main effects for rivalry, $\Lambda = .91$, $F(1, 65) = 6.65$, $p = .012$, $\eta^2 = .09$, and relative power, $\Lambda = .42$, $F(1, 65) = 90.28$, $p < .001$, $\eta^2 = .58$. Inspection of the marginal means revealed a higher mean value on rivalry for younger siblings. The rivalry score consists of maternal and paternal partiality scales. This result indicates that younger siblings across both groups (CF and Comparison groups) rated themselves as favoured by mothers and fathers as compared to older siblings. The relative power factor is the sum of nurturance of sibling and dominance of sibling minus the sum of nurturance by sibling and dominance by sibling. A higher mean value on relative power for older siblings indicates their rating of their relative status over their younger sibling, again across both groups. No other results were significant.

Analysis of SRQ Sub-Scales

A series of 2 x 2 mixed ANOVAs, with group (CF and Comparison) and age (younger, older sibling) as the single between subject factors, were conducted. Four significant results emerged from these analyses—significant age main effects for maternal partiality, $\Lambda = .91$, $F(1, 65) = 6.65$, $p = .012$, $\eta^2 = .09$; nurturance of sibling, $\Lambda = .51$, $F(1, 65) = 63.62$, $p < .001$, $\eta^2 = .49$; nurturance by sibling, $\Lambda = .85$, $F(1, 65) = 11.56$, $p = .001$, $\eta^2 = .15$; and dominance of sibling, $\Lambda = .60$, $F(1, 65) = 43.22$, $p < .001$, $\eta^2 = .49$. Inspection of the marginal means revealed a higher mean value for maternal partiality for younger siblings across both groups. While this suggests differential treatment of younger children by mothers, it was independent of the presence of a child with CF. The higher mean value for nurturance of sibling by older siblings, consistent with their more mature development, was independent of disease status. The reverse of this was experienced by younger siblings across both groups who rated a higher mean value for

nurturance by their older sibling. A higher mean value of dominance of sibling for older siblings was also found, but again, was independent of disease status. No other results were significant.

Significant correlations between Parental Differential Treatment and SRQ, SSRS and CDI subscales are shown in Table 10. The significance levels for the correlations presented in Table 10 were automatically adjusted in SPSS to account for over-inflation of the familywise error rate due to multiple testing. This adjustment permits individual correlations to be assessed against an α of .05.

Table 10

Significant Correlations Between Parental Differential Treatment and SRQ, SSRS and CDI Sub-Scales

CF				
Variable 1	Variable 2	*N*	*r*	*p*
Father Differential Treatment Total Time Excluding Medical Care	CDI Negative Mood	30	.43	.02
Father Differential Treatment Total Time Excluding Medical Care	CDI Interpersonal Problems	30	.43	.02
Father Differential Treatment Total Time Excluding Medical Care	CDI Anhedonia	30	.44	.02
Father Differential Treatment Total Time Excluding Medical Care	CDI Total	30	.44	.01
Father Differential Treatment Total Time	CDI Negative Mood	30	.40	.03
Father Differential Treatment Total Time	CDI Interpersonal Problems	30	.41	.02
Father Differential Treatment Total Time	CDI Anhedonia	30	.42	.02
Father Differential Treatment Total Time	CDI Total	30	.43	.02
Father Differential Treatment Recreation	CDI Negative Mood	30	-.41	.02
Father Differential Treatment Child Care	CDI Interpersonal Problems	29	.47	.01
Father Differential Treatment Child Care	CDI Negative Self Esteem	29	.39	.04
Father Differential Treatment Child Care	CDI Total	29	.38	.04
Mother Differential Treatment Meal Times	SSRS Total	33	.38	.03

Mother Differential Treatment Meal Times	SSRS Self-Control	33	.39	.02
SRQ Rivalry Younger	Father Differential Treatment Household Tasks	29	.52	<.01
SRQ Conflict Younger	Mother Differential Treatment Recreation	32	.40	.02
SRQ Maternal Partiality Younger	Father Differential Treatment Household Tasks	29	.46	.01
SRQ Warmth Younger	Father Differential Treatment Child Care	28	-.39	.04
SRQ Prosocial Younger	Father Differential Treatment Child Care	28	-.42	.03
SRQ Maternal Partiality Younger	Father Differential Treatment Child Care	28	-.40	.03
Nurturance Of Sibling Younger	Father Differential Treatment Child Care	28	-.42	.03
Nurturance By Sibling Younger	Father Differential Treatment Child Care	28	-.42	.03
Competition Younger	Mother Differential Treatment Household Tasks	32	.35	.05
Admiration Of Sibling Younger	Mother Differential Treatment Recreation	32	-.40	.02
Companionship Younger	Father Differential Treatment Child Care	28	-.41	.03
Similarity Younger	Father Differential Treatment Child Care	28	-.47	.01
Intimacy Younger	Father Differential Treatment Child Care	28	-.44	.02
Admiration By Sibling Younger	Father Differential Treatment Child Care	28	-.41	.03
Quarrelling Younger	Father Differential Treatment Time Alone Medical Care	28	.39	.04

SRQ Rivalry Older	Father Differential Treatment Time Alone Excluding Medical Care	29	-.48	.01
SRQ Warmth Older	Father Differential Treatment Total Time Excluding Medical Care	30	-.42	.02
SRQ Warmth Older	Father Differential Treatment Total Time	30	-.41	.02
SRQ Prosocial Older	Father Differential Treatment Total Time Excluding Medical Care	30	-.42	.02
SRQ Prosocial Older	Father Differential Treatment Total Time	30	-.41	.03
SRQ Maternal Partiality Older	Father Differential Treatment Time Alone Excluding Medical Care	29	-.54	<.01
Nurturance Of Sibling Older	Father Differential Treatment Total Time Excluding Medical Care	30	-.41	.03
Nurturance Of Sibling Older	Father Differential Treatment Total Time	30	-.39	.04
Nurturance By Sibling Older	Father Differential Treatment Time Alone Excluding Medical Care	29	-.41	.03
SRQ Maternal Partiality Older	Father Differential Treatment Household Tasks	30	-.54	<.01
Quarrelling Younger	Father Differential Treatment Child Care	28	.40	.03

Quarrelling Younger	Mother Differential Treatment Child Care	32	-.47	.01
SRQ Conflict Older	Father Differential Treatment Child Care	29	.43	.02
Nurturance Of Sibling Older	Father Differential Treatment Child Care	29	-.54	<.01
Affection Older	Father Differential Treatment Total Time Excluding Medical Care	30	-.46	.01
Affection Older	Father Differential Treatment Total Time	30	-.44	.02
Companionship Older	Father Differential Treatment Total Time Excluding Medical Care	30	-.52	<.01
Companionship Older	Father Differential Treatment Total Time	30	-.51	<.01
Admiration Of Sibling Older	Father Differential Treatment Total Time Excluding Medical Care	30	-.37	.04
Admiration Of Sibling Older	Father Differential Treatment Total Time	30	-.37	.04
Paternal Partiality Older	Father Differential Treatment Child Care	29	-.37	.05
Competition Older	Father Differential Treatment Child Care	29	.41	.03
COMPARISON				
Father Differential Treatment Child Care	SSRS Cooperation	29	-.40	.03
Father Differential Treatment Child Care	SSRS Self-Control	29	-.53	.00
Father Differential Treatment Total Time Excluding Medical Care	SSRS Empathy	24	-.57	.00
Father Differential Treatment Total Time	SSRS Empathy	24	-.57	.00

Father Differential Treatment Meal Times	SSRS Empathy	24	.42	.04
Father Differential Treatment Recreation	SSRS Cooperation	24	-.43	.03
Mother Differential Treatment Time Alone Excluding Medical Care	CDI Interpersonal Problems	26	-.45	.02
Mother Differential Treatment Total Time Excluding Medical Care	CDI Interpersonal Problems	26	-.45	.02
Mother Differential Treatment Total Time	CDI Interpersonal Problems	26	-.45	.02
Mother Differential Treatment Household Tasks	SSRS Empathy	26	-.40	.04
Mother Differential Treatment Meal Times	SSRS Empathy	26	-.50	.01
SRQ Rivalry Younger	Mother Differential Treatment Time Alone Excluding Medical Care	26	-.52	.01
SRQ Rivalry Younger	Mother Differential Treatment Total Time Excluding Medical Care	26	-.52	.01
SRQ Rivalry Younger	Mother Differential Treatment Total Time	26	-.52	.01
SRQ Relative Power Younger	Mother Differential Treatment Total Time	26	.40	.04
SRQ Maternal Partiality Younger	Mother Differential Treatment Time Alone Excluding Medical Care	26	-.50	.01
SRQ Maternal Partiality Younger	Mother Differential Treatment Total Time Excluding Medical Care	26	-.50	.01

SRQ Maternal Partiality Younger	Mother Differential Treatment Total Time	26	-.50	.01
Nurturance Of Sibling Younger	Mother Differential Treatment Time Alone Excluding Medical Care	26	.53	.01
Nurturance Of Sibling Younger	Mother Differential Treatment Total Time Excluding Medical Care	26	.53	.01
Nurturance Of Sibling Younger	Mother Differential Treatment Total Time	26	.53	.01
Dominance Of Sibling Younger	Mother Differential Treatment Time Alone Excluding Medical Care	26	.47	.02
Dominance Of Sibling Younger	Mother Differential Treatment Total Time Excluding Medical Care	26	.47	.02
Dominance Of Sibling Younger	Mother Differential Treatment Total Time	26	.47	.02
Quarrelling Older	Father Differential Treatment Meal Times	30	.37	.04
SRQ Conflict Younger	Mother Differential Treatment Recreation	26	-.45	.02
Paternal Partiality Younger	Father Differential Treatment Time Alone Excluding Medical Care	24	.41	.05
Dominance By Sibling Younger	Father Differential Treatment Recreation	24	.41	.05

Paternal Partiality Younger	Father Differential Treatment Child Care	24	.53	.01
Intimacy Younger	Mother Differential Treatment Child Care	26	-.42	.03
Admiration Of Sibling Younger	Mother Differential Treatment Child Care	26	-.41	.04
Quarrelling younger	Mother Differential Treatment Recreation	26	-.39	.05
Admiration Of Sibling Older	Mother Differential Treatment Child Care	26	-.43	.03

Hypothesis 4: Results for open-ended questions

The results for hypothesis 4 are based on the common themes derived from both the positive and negative aspects of being a sibling of a child with CF. It was anticipated that the sibling experience would differ according to the age of the healthy sibling. Healthy siblings aged 7 – 10 years were expected to cite concrete rewards (for example, gifts from organisations, trips awarded through charities) as the main advantage of having a sibling with CF, but healthy siblings aged 14 – 16 years were expected to report that this experience led to greater maturity and a heightened sensitivity to and understanding of the needs of people with chronic illness. The negative aspects of having a sibling with CF were expected to include being unable to participate in after-school activities or to plan family excursions for the healthy siblings aged 7 – 10 years, whereas emotional concerns about the health of their sibling with CF was expected to be the main disadvantage cited by healthy siblings aged 14 – 16 years.

For each of the open-ended questions, respondents were able to endorse more than one category. Responses were divided into age groups. At the time of recruitment the older siblings in the study were aged between 7 and 15 years. Two of the siblings in the CF group turned 16 during the period of data collection. A summary of each response is shown in Appendix E.

Question 1: “What happens to you when your sibling goes into hospital?”

About two thirds (n = 25; 64%) of siblings reported that their brother/sister with CF had been hospitalised since the initial diagnosis. Responses to question 1 were highly concrete and yielded largely descriptive information. While there were no major differences in responses by age of siblings, negative responses were more commonly described by the 14 – 16 year old age group. Figure 19 is a summary of the responses of the 25 siblings whose brother or sister had been in hospital. Responses were categorised into four themes using consensus coding. These categories were: disruption, positive, negative and neutral.

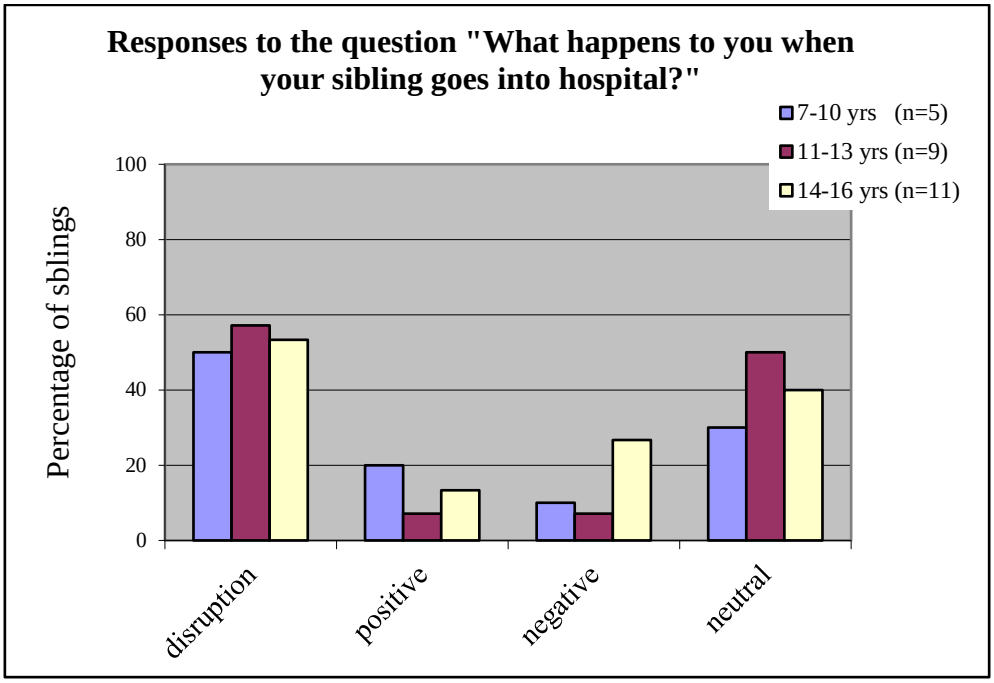

Figure 19. Responses to the question "What happens to you when your sibling goes into hospital?"
Note: siblings could provide more than one response.

Across the age groups, more than half of the siblings (54%) reported a disruption in their normal routines, such as staying with relatives or friends, when their sibling with CF was hospitalised. Forty-one percent of siblings gave a neutral response to this question. For example, one sibling stated:

"Mum stays at the hospital, dad goes to work and I stay with nanna".

Fifteen percent of siblings reported negative experiences. For example, one sibling said:

"I feel a bit left out (not that I'm complaining) as I go off to grandma's house or I'm shipped off to other people's houses".

In contrast, 13% of siblings reported that this time was positive: "I go with my Nan and it's fun."

Question 2: "What are 3 advantages of having a brother/sister with CF?"

The main themes to emerge from this question were categorised as: no advantages, special activities, caring for sibling, knowledge/empathy, closer relationship and other. Across all age groups, "special activities" was the response most frequently cited as the advantage of having a brother or sister with CF. "Special activities" included attending outpatient appointments and playing Nintendo in clinic followed by lunch at McDonalds, family holidays through wish granting organisations such as Starlight and Make A Wish and trampolining or bike riding with the affected sibling. Although the percentage of respondents was highest for the 7-10 year old group (80%), the number of siblings nominating special activities was consistently high across all three age groups. "Caring for sibling" was an advantage identified only by 7-10 year olds. This included helping with chest physiotherapy by doing percussion and completing the siblings' chores when the sibling with CF was unwell. Comments included:

"I like helping with his physiotherapy – patting and helping with his blowing".
(10 year old sibling)

"Knowledge/empathy" was reported as an advantage by the two older age groups of siblings. Fourteen percent of 11-13 year olds and 40% of 14-16 year olds cited this as an advantage. "Knowledge/empathy" included a better understanding of the genetics and biology associated with CF and assisting with school projects that taught others about CF. It also included developing a more sensitive attitude towards people with special health needs. One 13 year old said:

"You learn more about life".

Fourteen percent of 11-13 year olds and 13% of 14-16 year olds cited relationship advantages. This included having a closer relationship with and respecting their sibling more.

Figure 20 is a summary of the responses to question 2.

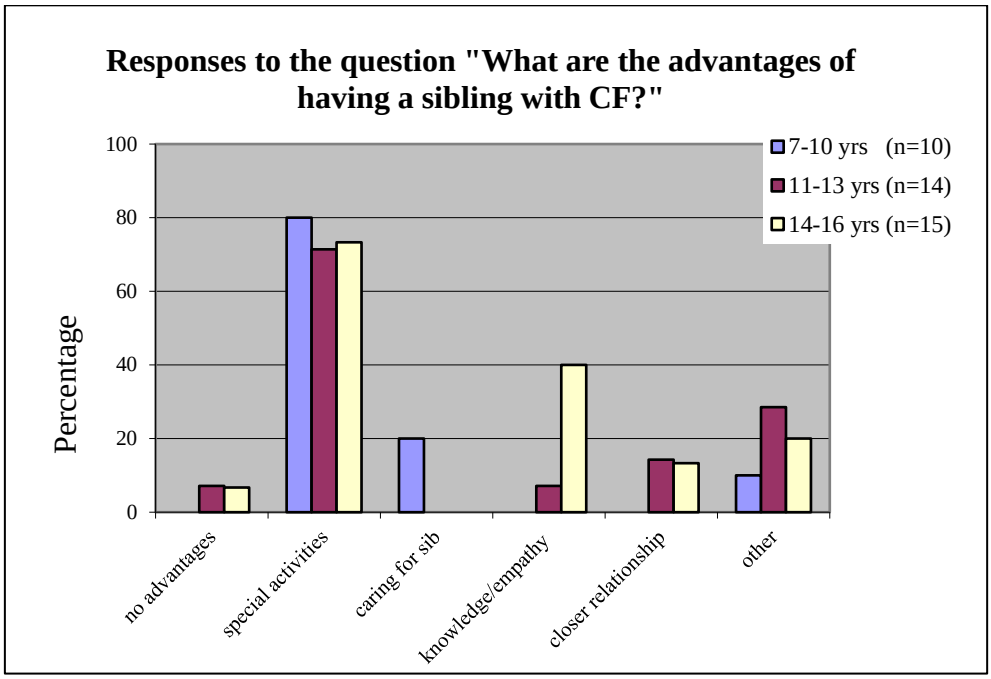

Figure 20. Responses to the question "What are the advantages of having a sibling with CF?"

Note: siblings could provide more than one response.

Question 3: "What are 3 things that are difficult about having a sibling with CF?"

The main themes to emerge from this question were categorised as: differential treatment, emotional, imposition/impact and no disadvantages. There were consistent patterns to the responses for this question. "Imposition/impact" and "emotional disadvantage" were the two dominant themes in siblings' responses across ages. "Imposition/impact" was the most frequent disadvantage cited for having a sibling with CF for 11-13 (71%) and 14-16 year olds (93%). It was equally endorsed along with "emotional disadvantage" by 7-10 year olds (50% for both categories of response). "Imposition/impact" included arguments about CF treatment, being unable to participate in extracurricular activities and being late to school due to the sibling's physiotherapy requirements. "Imposition" increased in frequency with older siblings.

"Emotional disadvantage" was reported consistently across the three age groups. These responses included health concerns about the sibling with CF, such as coughing, poor weight gain, and transmission of infections.

"Differential treatment" was a disadvantage cited by 14% of 11-13 year olds and 27% of 14-16 year olds. Children in the 7-10 year old age group did not report differential treatment. Comments in this category included:

"My brother has a lot of time with mum and dad and is spoilt".
(15 year old sibling)

And:

"When I come to visit my sister in hospital I bear the brunt of her feeling bad and the staff emphasise her – what about me?".
(14 year old sibling)

One 11 year old sibling said:

"My sister with CF is favoured by my grandma. She doesn't get into trouble as much as if I did the same things".

Responses to question 3 are summarised in Figure 21.

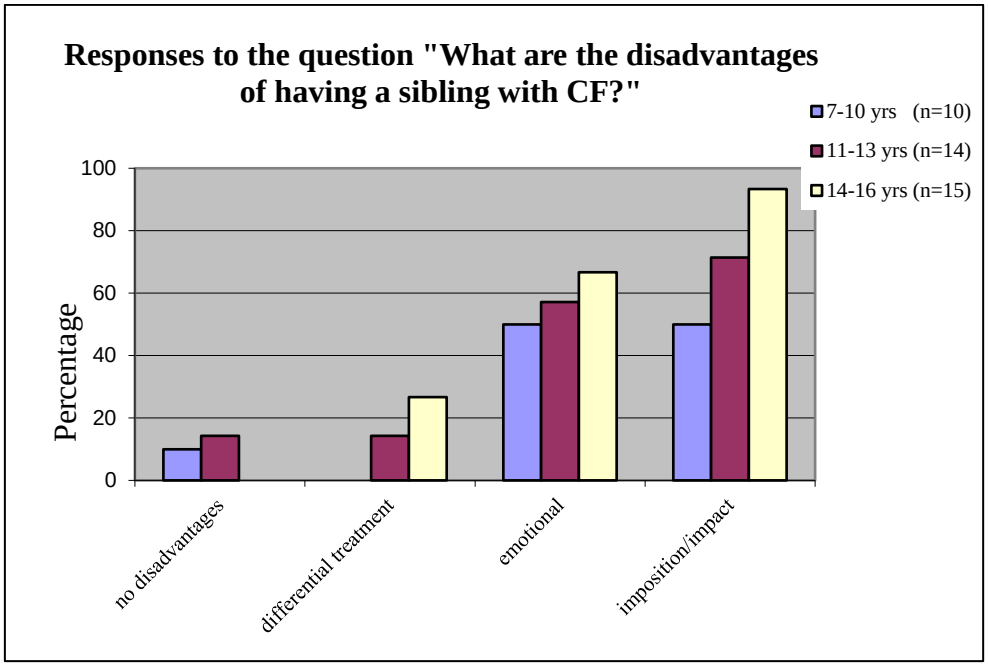

Figure 21. Responses to the question "What are the disadvantages of having a sibling with CF?"

Note: siblings could provide more than one response.

Question 4: "What would make things better/easier for you?"

The main themes to emerge from this question were no suggestions, find cure, impact, health concerns, proximity to RCH, facilities/resources and other. A large number of children in the 7-10 year old age group (50%) were unable to think of anything that what would make things better or easier for them. Responses to this question strongly endorsed the need for sibling facilities and resources, such as camps and groups that provide an opportunity for siblings to share their experiences. This was particularly evident in the 11-13 year old group (29%) and 14-16 year olds (60%). An example of responses included:

"Camps for siblings would provide escape from pumps and coughing and you would get to talk to others who know how you feel".
(14 year old sibling)

And:

"To have a meeting with a group of siblings to hear their perspectives of what it's like".
(15 year old sibling)

"Finding a cure" for CF was mentioned by 20% of children in the 7-10 age group, 29% of the 11-13 year old group and only 7% of 14-16 year olds. Other responses included the "impact" of CF. This category included wishing that the sibling would agree to do physiotherapy in another room so that friends could come over. Another response was that if the sibling did not need to take enzymes, the family could eat out and not worry about forgetting them.

"Proximity to the hospital" related to the provision of CF services in regional areas so that families did not have to travel as much. "Health concerns" included aspects of CF symptoms (such as coughing) the siblings wished could be alleviated, as distinct from hoping for a cure.

Figure 22 summarises the responses to question 4.

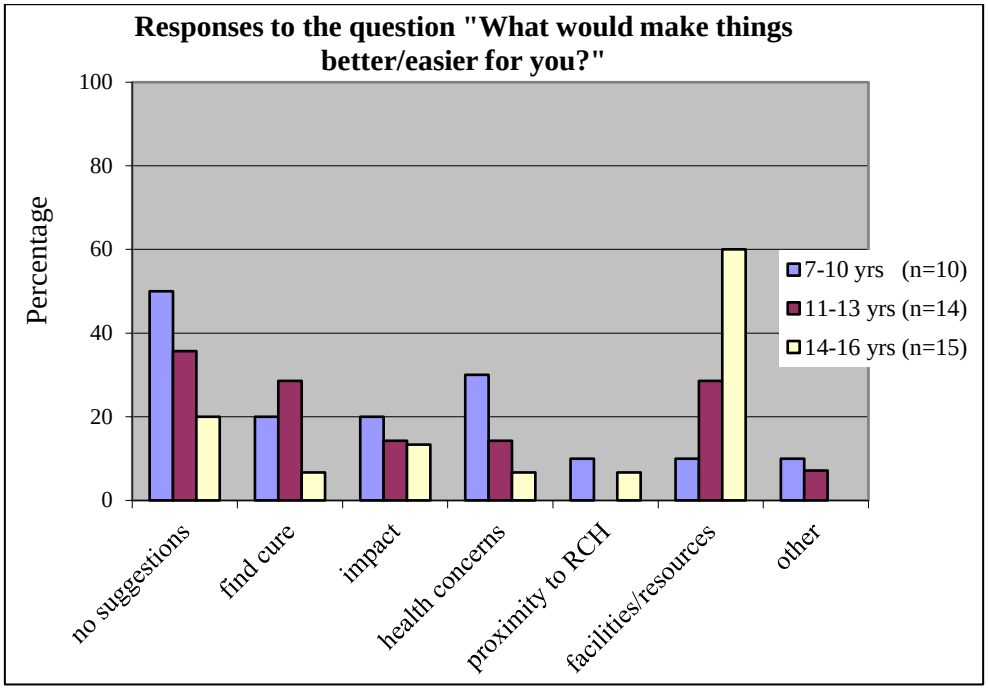

Figure 22. Responses to the question "What would make things better/easier for you?" Note: siblings could provide more than one response.

DISCUSSION

Summary of the main findings

The overall objective of this research was to assess the extent of parental differential treatment in two types of families, those with a well child and a child with CF and those with two healthy children. A second aim was to evaluate the impact of parental differential treatment on the quality of the sibling relationship, and the social and emotional adjustment of well siblings in families caring for a child with CF. The design of this investigation enabled a broader range of differential parental behaviour to be studied through the inclusion of fathers and provided new insights into the complexity of family life with CF in an Australian context.

Overall, this study found evidence of parental differential treatment by fathers but not mothers. For mothers in families caring for a child with CF, a small but consistently larger amount of individual time was spent with both younger and older children. However, for fathers, the presence of a child with CF in the family had a more pronounced effect, with a larger and more obvious difference in the amount of individual time spent with younger siblings, but no notable difference in the amount of time fathers spent with older siblings, regardless of the presence of a child with CF.
Both parents spent over twice the amount of time with younger siblings in mealtime activities, across groups. This is surprising given the emphasis on nutrition in the contemporary management of CF, as was highlighted in the description by the 13 year old patient Harry about the additional issues at mealtimes, which can lead to conflict in response to parental concerns. One would have expected the parents in the CF group to spend significantly more time in mealtime activities with the younger children with CF than parents in the Comparison group. As expected, mothers spent more time with younger children with CF in medical care than fathers and, not surprisingly, children in the CF group spent more time in medical care than children in the Comparison group.

For both groups, a greater level of parental differential treatment in favour of the younger sibling was expected to be associated with poorer sibling relationship quality. This hypothesis was partially supported, but only for fathers. High levels of paternal

differential treatment for children with CF were associated with the following SRQ subscales for the older sibling: lower levels of maternal partiality, higher levels of rivalry, lower levels of nurturance by the younger sibling and higher levels of competition. For the younger sibling with CF, high levels of paternal differential treatment were associated with higher levels of quarrelling and paternal partiality.

Further, siblings in the CF group were expected to report worse sibling relationship quality than siblings in the Comparison group. Very limited support was found for this hypothesis; specifically, a small number of age-related main effects were identified, but no significant differences were found that specifically involved the CF group. Given the importance of sibling relationships across the life-course, and the increasing life expectancy of people with CF, these data are generally encouraging in relation to what they might suggest about the quality of sibling relationships when one child has CF.

The findings of this study extended previous CF studies (Opipari, 1996; Quittner & Opipari, 1994) by recruiting both mothers and fathers. Rather than finding maternal differential treatment, the results of this study revealed evidence of differential treatment by fathers, but not mothers. While not focused on CF, the results of this study are consistent with earlier studies of siblings that included fathers and found evidence for differential treatment playing a role in determining the quality of the children's sibling relationships (Brody et al., 1992; Volling & Belsky, 1992). The results of this study suggest the value of future research exploring how fathers view their role to better understand how they divide their time between their children. Previous cancer research has suggested a range of motivations from guilt to stress avoidance as the explanation for fathers' allocation of time with their unwell child (Sloper, 2000), however it is not known to what extent this also applies to families with a child with CF.

The qualitative aspect of this study yielded interesting information about what it is like to have a brother or sister with CF. As expected, the responses of these siblings differed greatly according to their age and cognitive maturity. Not surprisingly, the daily routines of well siblings were disrupted by the hospitalisation of the sibling with CF. Although some well siblings found the change in arrangements a fun or positive experience (such

as staying with friends or relatives), for many, especially with increasing age, it was a negative one. Siblings commented about feeling left out or being “shipped off” to other people. These findings are likely to be emotionally meaningful in terms of children’s lived experiences of their family life, as children find it very difficult to admit to negative or challenging aspects that may directly or even indirectly be seen to be critical of their siblings (or parents). In this context, despite potentially powerful negative emotional responses, they may have felt unable to acknowledge fully these responses to the researcher or were limited in doing so due to their level of cognitive development.

As predicted, concrete rewards such as family holidays were the most commonly cited advantages of having a sibling with CF in the 7 to 10 year old age group, particularly around special activities during clinic visits and family holidays through wish granting organisations. These positive aspects of having a sibling with CF are consistent with those of Deeley (1996), who in her study of 19 siblings of children with CF reported that treats such as special holidays or gifts from the local CF group were greatly valued.

As siblings grow older, greater cognitive maturity results in the potential for siblings to gain enhanced appreciation of and empathy for the needs of people with CF. This was especially notable for the 14-16 year olds in my study, many more of whom described benefits related to gaining a more sensitive attitude towards people with special health needs than did younger children. Deeley (1996) also found that older (14 year old) siblings mentioned personal development as a positive aspect of having a sick sibling, such as better understanding of others when they are upset. As with these Australian data that showed that the two older age groups enjoyed the benefit of knowledge about genetics and biology of CF that was, for example, helpful with school projects, so too did similarly aged subjects in the study by Deeley (1996) and the small pilot study by (Russo & Hogg, 2004), notwithstanding the different chronologies and countries of these studies.

The most frequently cited challenges about having a sibling with CF reflected the difficulty for siblings to commit to extracurricular activities because of the unpredictable nature of CF. Several siblings talked about not knowing what the day would bring and whether they would come home from school to find out that their sibling required an

admission to hospital. Regularly being late to school was another common theme. Again, while siblings spoke very "matter-of-factly" about the various changes to household arrangements caused by CF, the extent to which healthy siblings responded emotionally to these disruptions to family routine was not captured using this approach. These relatively simple questions were able to generate some interesting insights about the impact of CF. Indeed, it may have been that the concrete aspect of these questions enabled healthy siblings to be relatively frank about some negative aspects, especially for younger children. Wider emotional reactions, anxieties or fears were less well captured overall. One area of concern related to the worry that healthy siblings had about passing on a cold; this was a consistent theme even at a young age. Another child was highly anxious about the risk of death for their sibling which was inconsistent with the health of her brother at the time. These concerns suggested the presence of underlying anxieties and fears children have about their sibling with CF. Rather than brief questions, more in-depth interviews might enhance our understanding of the unique challenges and issues of siblings of children with CF. For example, the ever-present "spectre" of CF was reported by Jessup and Parkinson (2010) in a qualitative study on the impact of living with CF from the perspective of individuals with CF and their parents, but not siblings. As highlighted previously, the degree of difficulty that siblings have in expressing negative emotional impacts that might be perceived as critical of their sibling or family suggests the value of utilising a variety of methods beyond expressed language alone, such as drawing or other forms of play, as used by Jessup and Parkinson (2010). A balance would be required between the presence of parents (which would contribute to children feeling safe with an unknown interviewer) with the need for privacy and confidentiality that might promote greater candour.

Interestingly, parental differential treatment was explicitly articulated as a disadvantage, but only by older siblings. Perhaps this is less able to be expressed by younger children, whose behaviour may better reflect their concerns (such as increasing demands on parents at the time of CF treatments). Some siblings qualified their responses about their brother or sister with CF getting more time and attention from parents with comments such as, "I know it has to be like this." These additive comments were often tinged with remorse for expressing the inequality they experienced or perceived, consistent with the

above interpretation of how difficult it is for children to express opinions that are perceived as critical of their family.

Interestingly, parental differential treatment was reported as a difficult issue by half of the participants in Deeley's (1996) study, who were a similar age to this Australian sample. In that study, the need to make sacrifices was another negative issue identified by participants, including having to forego their own activities for the sake of their ill sibling. This was described as affecting friendships and preventing the children from pursuing their favourite activities (Deeley, 1996). While in the current study, the issue of parental differential treatment was apparent with the main disadvantage being in relation to siblings being unable to participate in extracurricular activities, this finding was not as prominent as in Deeley's (1996) work.

In sum, this study provided preliminary evidence regarding the associations between sibling adjustment and relationship quality and paternal differential treatment. In particular, the qualitative findings reflected both positive and negative impacts of CF on well siblings and suggested the value of paying more attention to siblings.

Measurement issues

DPD data were generally straightforward to both analyse and collect and most families were highly cooperative with this aspect of data collection. A minority of families (four CF families and one comparison family) did not participate in this aspect of the research. The research assistant who collected these data was gently persistent and offered maximum flexibility to parents (offering to call at other times of the day for example) with the aim of collecting phone diaries from all of the parents in the study. Despite the DPD taking only 15 minutes to complete, perhaps the difficulty for some families in completing diaries was due to the busyness of family life, especially family life with CF. However, perhaps the parents who chose not to participate in this aspect of the study were in some way sensitive about what might be identified.

This study assessed mothers' and fathers' differential treatment, however we did not measure children's perceptions of the magnitude and type of differential treatment.

Opipari (1996) explicitly measured children's reports of differential treatment. She found that children's reports about this aspect of their family life converged with both maternal reports of differential treatment and diary-based measures of differential time. Even though this was not directly tested in this study, use of the partiality scales of the SRQ and the open-ended questions assessed children's reports of parental differential treatment. In retrospect, it would have been interesting to have collected explicit reports of these data from siblings, as did (Opipari, 1996). This is recommended for future studies. It may also be important to understand what types of differential treatment are most important to children and whether particular types of differential treatment affect specific aspects of child functioning.

Turning to the question of how family context influenced the links between paternal differential treatment and child functioning, one of the most striking findings was the pattern of associations between paternal differential treatment and child outcomes in the CF group. Paternal differential treatment favouring the younger child with CF was associated with less adequate functioning in the sibling and negative reports of sibling relationship quality. Several important issues arise from these results. First, the results suggested that the magnitude of differential treatment is an important factor in determining its impact on child functioning. This is consistent with the findings of Opipari (1996). In my Comparison group, while there were differences in parental behaviour towards the siblings, there was little evidence that these differences were related to sibling relationship quality or adjustment. In contrast, consistent associations did emerge with the increased magnitude of differential treatment in the CF group. This pattern of results indicated that there may be a threshold of acceptance for differences in parental behaviour above which negative consequences for the well sibling may result.

The results of this study also raised important questions about the meaning of parental differential treatment in the context of daily family routines. Earlier investigators have suggested that the legitimacy of differences in parental behaviour in family contexts in which a child has a chronic illness or disability may weaken its associations with child functioning (McHale & Pawletko, 1992). The results of this study suggested that this is not the case. Although the healthy siblings in this study were not directly asked about their experiences of

parental differential treatment, several of them commented on this as one of the difficulties of having a sibling with CF, often adding that they understood why. Despite this, an association was still found between paternal differential treatment, child outcomes and the quality of the sibling relationship. Perhaps the well siblings expected their mothers to spend more time with the child with CF but not their fathers. Maybe they unconsciously expected fathers to buffer the lack of maternal time by spending additional time with them. More research addressing children's perceptions of parental differential treatment is needed before conclusions can be drawn.

Study strengths

The shift to a family focus of child health has been extended to include the importance of the father's role in their child's day-to-day care. It has only relatively recently been recognised that fathers play an extended role in the family, not only as the bread-winner, but also in terms of nurturance and direct care-giving (Lamb, 2000). Consistent with this, research has only recently broadened its focus to include the effects of paternal behaviour on child health outcomes (Tully, Piotrowska, et al., 2017). One strength of the current study is that it provides some of the first data regarding the role of fathers in families of children with CF. This study provides the first evidence that the paternal differential time variables calculated from the DPD were associated with measures of well sibling functioning. I am unaware of any other studies that have addressed parental differential treatment and CF in Australia, whether by mothers or fathers, or of any other Australian studies that have used the DPD software.

Notwithstanding the large numbers of research studies that CF families at the RCH are invited to participate in, all eligible families were recruited into the study. While obtaining a response rate of 100% of eligible families is a major strength of the study, it is important to consider whether my clinical relationship with the families was an explanatory factor for this in ways that were not solely positive. Given my clinical role, it may have been that families felt unduly coerced to participate. I was mindful of this when recruiting families, and this theme was also regularly discussed in the context of research supervision. An alternative view, and one that I believe is more likely, is that the high

response rate is an endorsement of the perception of the importance of this research by CF families. For example, I actively discouraged one family from enrolling in the study as their daughter with CF was extremely unwell at the time. I was very surprised that they still chose to participate, telling me that this was a neglected and important area of enquiry.

An additional strength of this study was the recruitment of a comparison group. As discussed in the Methods section, many different approaches could have been used to recruit a comparison group. Asking the CF families to nominate another unrelated family with children of similar ages worked extremely well in recruiting a demographically similar group to compare our CF families to, although not all families were able to nominate a comparison family. As previously discussed, a characteristic of the families that were unable to nominate a comparison family was that their children with CF had spent very little time in hospital. It may have been that these families had therefore not been required to share the diagnosis of CF with friends.

While this study was primarily quantitative, the qualitative analysis of the four open-ended questions added to our understanding of the CF sibling experience and highlighted the reality of differential parenting in families of children with CF.

Study limitations

This study had some limitations. First, the results were based on cross-sectional data that cannot provide information about causality or the direction of effects. While these data provided some information about the process (for example, the DPD insights about differential parenting and some of the specific scales of the SRQ), longitudinal mixed methods research which focuses on the richness of interaction patterns between different family members may help to raise hypotheses about more specific processes underlying the observed associations between differential treatment and child functioning.

Further, although differential treatment is usually examined by looking at its impact on child outcomes, the relationship between these variables is more likely to be

bi-directional and more complex than suggested by a simple causal model. Longitudinal research, using mixed methods, that focuses on the complex interactions that occur in daily life would help explicate the relationship between parental differential treatment and child functioning.

Notwithstanding the high response rate, the relatively small sample size in this study reduced its power to detect effects that may have been present. For example, high levels of paternal differential treatment for children with CF were associated with lower levels of self-control in the older siblings but this did not reach significance in this sample. A multi-site study would be required to overcome this limitation.

The children with CF in this study were generally well; one-third had not required hospitalisation since the initial diagnosis. Studying an older sample of adolescents and young adults with CF may increase the likelihood of uncovering the effects of parental differential treatment, given the increased requirements of family care with increasing age (for example; CFRD) and for more unwell people with CF. However, one could also argue that given the impact of new therapies and the fact that children with CF are in better health during childhood and adolescence than before, recruiting an older sample may not uncover greater parental differential treatment. It could be argued that people with CF now live longer due to more complex and time-consuming treatments on a daily basis from diagnosis. Regular surveillance through frequent outpatient assessment and sputum analysis results in an increasingly intensive daily treatment regimen that is carried out at home. Paradoxically, this may result in even greater parental differential treatment despite better health status.

Unlike previous studies in CF (Opipari, 1996; Quittner & Opipari, 1994), this sample of children with CF was diagnosed by NBS, which was introduced in Australia well before being introduced throughout the United States. At the time NBS was introduced, there were concerns that the early diagnosis of CF could reduce parent-infant bonding. While this has not been shown to occur, the extent to which early diagnosis might impact parental differential treatment (such as in fathers as shown in this research) remains

unknown as there are no directly comparable studies of parent-infant bonding prior to the implementation of NBS.

This study did not examine the social networks of children growing up with a brother or sister with a chronic illness. A strong social network may act as a protective factor against the negative effects of parental differential treatment for the siblings of children with CF. Opipari (1996) found evidence to suggest that CF siblings had altered social networks. Specifically, the CF siblings in her study had smaller social networks resulting in fewer relationships with individuals outside the immediate family, less total support and less overall time with their network members on a daily basis when compared to age-matched controls. These results are consistent with the responses from the siblings in this study when asked about the difficulties of having a brother or sister with CF. "Imposition/impact" was the most frequently cited disadvantage across all age groups. This included being unable to participate in extracurricular activities and being late to school due to their sibling's morning treatment regimen. Getting to school before the commencement of classes provides an opportunity for supportive peer interactions as does membership of sporting teams and other interest groups outside of school. The fact that the siblings endorsed the need for contact with other children growing up with a brother or sister with CF also reflected the impact on social support networks. Given the strict cross-infection policies now in place, there are no longer opportunities for siblings to meet other siblings at CF gatherings. These types of alterations in the daily lives of children growing up with a brother or sister with CF may have important implications for their social and emotional development. More research looking at how CF influences the daily lives of healthy siblings and the impact of social networks on child functioning is warranted.

Consistent with limited social networks, especially that link healthy siblings to others with an unwell sibling, the well siblings of children with CF in this study strongly endorsed the value of sibling resources such as camps and support groups. These peer support opportunities would enable them to share their experiences.

The writing up of this thesis was significantly delayed by family health issues. This delay raises some question about the relevance of these data to current families in the context of various changes to clinical practice at the RCH and in relation to CF care, the engagement of fathers and the impact of CF on siblings. Having said this, a large, recent Australian study about the engagement of fathers in clinical interventions suggested that this still remains an issue; the authors suggested that greater attention to including fathers is needed (Tully, Collins, et al., 2017; Tully, Piotrowska, et al., 2017).

Recently, the major advance in CF care has been the development of CFTR-targeted therapies. While these treatments are an additional burden, they are only relevant to less than 10% of the current RCH clinic population. In addition, clinical practice at the RCH has changed with the identification of a group of patients who have established bronchiectasis and are now having regular planned hospital admission for 10 to 14 days at a time, four times a year, which poses an even greater burden and potential impost on siblings. However, this group constitutes less than 20% of the RCH CF clinic. The greater use of ‘Hospital in the Home’, an RCH service where children with CF have intravenous antibiotics at home, is being used more often due to the increased number of admissions and demand for hospital beds in general. This is an attractive option for families of children with CF in terms of avoiding the negative aspects of hospital admission (for example, cross infection, disruption to family routines, travel time) and because children can still attend school while receiving CF treatment. However, as parents are required to monitor treatments at home, the encroachment of CF care on daily family life for the duration of home care is likely to amplify the experience of parental differential treatment for siblings. Given these changes, CF treatment in most families is more “front and centre” than ever before. For this reason, parental differential treatment may be even more relevant and prevalent for contemporary families than when this study was initiated.

Implications of this research for clinical practice

This research raises important implications for clinical practice, both about the role of parents (especially fathers) and healthy siblings. An important question that arises from this work is how to engage fathers specifically, given their important role in the adjustment of healthy siblings. At the very least, given the effect of paternal differential

treatment on sibling adjustment, there are opportunities for CF teams to work harder in setting expectations that both parents be involved in all aspects of CF care, and not just at diagnosis. This is consistent with Tully and colleagues who, beyond CF and chronic illness, recently highlighted the importance of inviting fathers to participate as part of the core parenting team (Tully, Collins, et al., 2017; Tully, Piotrowska, et al., 2017). It also aligns with recommendations from Hayes and Savage (2008) who endorsed the need for supportive interventions with fathers as an integral part of managing the care of children with CF. While most CF teams have involvement with fathers at the time of diagnosis, ongoing contact can be sporadic due to paternal work commitments; it is mostly the mothers of our patients who attend clinic appointments and stay with their child if they are admitted to the hospital. CF teams could work harder to set expectations that fathers attend regular outpatient appointments, notwithstanding the challenge of this for their employment (which also applies to working mothers).

Such efforts to better engage fathers in clinic appointments is consistent with earlier research demonstrating that fathers can feel left out, given they are often not involved in doctor's appointments or the child's treatment regimen (Sterken, 1996). Fathers may also feel less competent because of their perceptions of the mother as the CF expert (Turner-Henson, Holaday, & Swan, 1992) due to more regular attendance at clinic appointments by mothers. These data suggest that for various reasons, fathers may limit their participation in CF care at home, which can then set up unhelpful cycles that reinforce maternal expertise. Ensuring that these issues are addressed by CF services may help to better engage fathers in CF care at home.

Health services have other opportunities to engage both parents, in education sessions that are run by clinics and CF organisations, such as CF Australia. In these forums parents can validate and support each other and share solutions in meaningful ways. Yet, at least in Australia, mothers are still more likely to participate in these activities than fathers. For example, new technologies can be used to host virtual group meetings for parents via webinars facilitated by psychologists or social workers. These are opportunities to discuss the experiences of siblings, parental differential treatment and the

challenges of daily family life with CF. These virtual forums may make it more feasible for both parents to participate, given that neither travel time nor child care is required.

In terms of the extent of care required by parents of children with CF, at least some degree of parental differential treatment is unavoidable. However the qualitative findings from this study suggested that healthy siblings could be supported by family interventions that facilitate parental awareness of well siblings' experiences of and attitudes about CF and its impact on family relationships and communication. Since undertaking this study, my personal practice has greatly changed and I pay far more attention to the well-being of healthy siblings with some relatively simple interventions to buffer the potential impact on them. At diagnosis following NBS, I now consistently talk with both fathers and mothers about the notion of parental differential treatment, highlighting the importance of healthy siblings being included.

Within the education sessions at diagnosis, the RCH CF team also now more consistently provides age-appropriate information to parents to help them reduce the potential impact of parental differential treatment. This includes information that healthy siblings should be told why CF treatments are necessary and why the parent needs to be so involved, as it is often assumed that siblings know this. As siblings mature, their capacity to ask questions, understand more complex information, and experience new concerns and emotional responses evolves, highlighting the need for CF-specific information to be repeatedly shared with healthy siblings.

When I embarked on this research, the proportion of families with healthy siblings was unknown; data were not routinely collected on siblings, whether in terms of number, gender, age or other factors (for example, health status). The only siblings we knew about were those who also had CF, a decreasing incidence in the context of reduced family size and the introduction of NBS. Who are the siblings in our CF clinics? Do they attend clinic at least once a year? How do we engage siblings when they do attend clinic? What is their experience of their sibling's CF in terms of anxieties and fears, resentment or envy? Most clinicians in our CF service would not have known the answers to these questions prior to this research. While there is now a greater emphasis on healthy

siblings, a more systematic approach could be developed. When healthy siblings do attend clinic, I make a point of acknowledging their presence and engaging them in the consultation, as well as the child with CF. There are, however, opportunities to extend this further. At the annual review of all patients, a report is produced that is currently reviewed by the CF consultant with each family and child. This annual review process could provide the opportunity for the CF team to be more inclusive of healthy siblings. Having the consultant check in with families about their other children's wellbeing, even briefly, could help achieve this, as might the development of age-appropriate, written resources for families, including siblings.

As a result of infection control risks, CF organisations have had to discontinue social gatherings of patients. However, this should not limit the opportunities for healthy siblings to gain support from each other as in other disease groups, such as cancer (for example, CanTeen), and contexts (for example, Very Special Kids). Very Special Kids is a Melbourne-based respite service for families of children with severe chronic illness and disability that routinely offers family activities, including those that target siblings. This service provides a Sibling Support Programme for children living with a brother or sister who is unwell. This non-categorical programme offers special or fun activities that are solely for the healthy sibling. This might help counter sibling feelings of envy and resentment. Very Special Kids' Sibling Support Programme provides activities where siblings have the opportunity to meet other siblings in a caring and safe environment. In addition to having fun, a therapeutic framework allows children to not only explore their emotions, but also enhance self-esteem, encourage peer support and reduce their sense of social isolation (Strohm, 2005). Given the value of peer support programmes for adolescents with chronic health conditions (Maslow & Chung, 2013), it is interesting to reflect on the feasibility of programmes that offer support to adolescents with a specific condition or group of conditions (for example, cancer) versus those that are non-categorical, such as Very Special Kids.

The results of my qualitative research that explored what things might make it easier for siblings (question 4) raise the question of whether there is also value in peer support programmes to meet the needs of siblings of patients with chronic health conditions such

as CF. For healthy siblings, non-categorical or generic chronic illness programmes are likely to be a more pragmatic solution than CF-specific programmes, given the limited funding environment and the likelihood that sibling support programmes will not be the highest priority for funding by individual clinical departments.

At the RCH, the Chronic Illness Peer Support programme (ChIPS) provides a successful model that could be adapted to provide non-categorical support for well siblings in addition to individuals with a chronic health condition. The ChIPS programme has been run at the RCH since 1993, established by the Centre for Adolescent Heath. This programme was designed to assist young people in their adjustment to life with a chronic health condition, including CF. The programme recognises that young people with a variety of different medical conditions share similar concerns (Olsson, Boyce, Toumbourou, & Sawyer, 2005). Initial engagement with the programme is through participating in a peer support group that meets for 90 minutes weekly for eight weeks, facilitated by a health professional (nurse, social worker or youth worker) and a peer co-leader. Each group typically includes between six and eight young people with a variety of chronic health conditions. Due to the risk of cross-infection, only one young person with CF is able to participate in each group. The ChIPS programme aims to provide young people with the opportunity to build resilience and well-being and the capacity of the participants to move into other areas of their lives with greater self-confidence and self-acceptance. These aims would have parallels in well sibling programmes.

Olsson et al. (2005) discussed nine psychosocial mechanisms by which peer support groups might improve the resilience and well-being of participants. These mechanisms are: learning new coping techniques; learning how to influence social environments; enlarging perspectives on what is normal; examining altering perspectives; understanding the cause of personal stressors; confirmation of positive changes in attitudes; reduced sense of social isolation; enhanced social identity through group approval; and building empathy through extending help to others. A peer support programme for well siblings may provide similar advantages.

There are also several potential disadvantages associated with peer support programmes for the well siblings of children with CF. For example, a participant may learn new and distressing information about their sibling's condition, such as males with CF being generally infertile. These challenges are managed within the ChIPS programme, where the culture consistently fosters healthy attitudes.

Havermans and colleagues have suggested that it is important to consider which specific interventions might help support which groups of well siblings, such as those with less predictable prognoses (Havermans, Croock, et al., 2015). Arguably, the development of a support programme across the hospital, with input in its development by siblings themselves about what might best meet their needs, could be considered one part of the hospital's commitment to family-focused care, as highlighted by Strohm (2005).

Implications of this research for policy

The Australian and New Zealand CF Psychologists' Network is contributing a chapter to the updated national standards of CF care. Disappointingly, these standards focus remarkably little on working with either families or siblings. Nearly complete, the updated draft of this chapter does not mention siblings. In fact, families are only mentioned at the time of diagnosis, which feels a remarkable omission. During the development of that document, there have been some interesting discussions by members of the network regarding the actual role of the CF psychologist. In particular, the issue of who is the client has been raised. Psychologists working within CF centres are limited by time, but also cited conflict of interest and difficulties with the storage of information if siblings do not have a hospital record. Most psychologists working with children with CF in Australia and New Zealand don't see other family members unless it is part of the direct support for the child with CF. This is very different to psychologists working with adolescents with eating disorders, for example, who are more likely to take a family-focused view and include siblings within the therapeutic environment (Hughes, Burton, Le Grange, & Sawyer, 2017), albeit that their own issues remain unable to be addressed through these mechanisms.

Beyond the development of clinical standards, there are ongoing discussions amongst the Australian and New Zealand CF Psychologists' Network regarding how to support family members, including siblings. There has been less discussion within other professional groups involved in the care of families affected by CF, such as the Australasian CF Nurses' group or by respiratory physicians. This will be increasingly important as the longer life span of people with CF reinforces their reliance on their siblings over time. Ideally, siblings would be considered by a variety of health professionals, including psychologists, nurses and physicians, as an integral part of the family system.

These challenges around engagement of siblings relate to policies that support the implementation of family-centred care. Strohm (2005) argued that preventively-oriented sibling support is not merely an attempt to make siblings feel better, but rather, an important mental health strategy for them. Rather than waiting until clinical problems emerge, a critical step in supporting siblings is through a preventive lens that first supports parents. Agreement by health services about these aspects of family-centred care will be required by CF teams for them to fully support parents.

Implications of this research for future research

Despite the fact that siblings usually maintain their relationships throughout life, there has been little research looking at the longitudinal aspects of sibling relationships when one sibling has CF (Havermans, Croock, et al., 2015). Ideally, one would follow a large group of siblings of children with CF at different developmental stages and points in the illness trajectory to better understand changes in parental differential treatment, sibling relationships and adjustment, and the processes around these, over time. A longitudinal study of parental differential treatment would enable tracking of how these processes unfold over time. Wennstrom, Isberg, Wirtberg, and Ryden (2011) conducted the first prospective, long-term investigation into a population of CF patients in Sweden at 6 - 14 and 18 – 26 years of age. They looked at 37 adult sibling pairs with regard to their self-esteem, life satisfaction and attitudes towards the CF siblingship situation. The self-esteem of women in the sibling pairs had improved since childhood, as measured by the 'As I see myself' questionnaire. Women with and without CF and the men with CF were

found to have lower ratings of life satisfaction on the “Ladder of Life” scale compared to a healthy reference group. The adults with CF viewed themselves on the “Sibling Mirror” (an unstandardised instrument) as independent, thoughtful and mature, but remembered themselves as being spoiled or fussy as youngsters. Healthy siblings considered themselves to be diplomatic, responsible and mature, but remembered themselves as angry, envious and neglected. The authors concluded by reinforcing the importance of longitudinal research into the relationship between siblings, especially given that the well siblings could potentially play a role in treatment planning as people with CF outlive their parents.

With the outlook for people with CF continuing to improve, beyond the focus on children and adolescents as in this study, future research on healthy adult siblings is warranted. Issues for well adult siblings of adults with CF include the genetic implications when they are contemplating having children (whether or not to have carrier testing) and the support needs (emotional, financial and physical) of affected siblings as their health deteriorates.

Despite our increased understanding of differential treatment through the recruitment of fathers, the fact remains that we know very little about how specific types of differential treatment affect child outcomes. Assessing parental differential treatment using different methods and multiple informants will expand this understanding. Studies investigating the type and magnitude of differential treatment and its association with child outcomes in a variety of family contexts may provide a richer understanding of the processes underlying this aspect of the within-family environment. Beyond its intrinsic benefit, replicating this research with families of children with other types of chronic illness may help inform our understanding of parental differential treatment in families with CF.

At the time of this study, there was limited diversity of family composition in our CF clinic. While there were separated single-parent families, there was little family diversity, such as same-sex parents. It is for this reason that in addition to the word “parents”, the terms “mothers” and “fathers” have been used throughout this thesis with the intention of bringing greater visibility to fathers’ roles. Given the increasing diversity of Australian

families, including sexual diversity, future research will benefit from including their perspectives and ensuring that inclusive language is used.

Siblings of patients with CF should be validated by health services as well as by their families. In addition to this research, my clinical experience has affirmed that siblings are brave beyond words, often very mature and display incredible courage. The vision of the RCH is to be a great children's hospital, leading the way in patient and family-centred care. This research reinforces that it is time for us to acknowledge and support the siblings of our patients as part of that vision.

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Appendix A

Ethics approval, information statements, consent forms and introductory letter

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ETHICS IN HUMAN RESEARCH COMMITTEE

APPROVAL

EHRC REF. No:	**22158 A**
PROJECT TITLE:	The impact of Cystic Fibrosis on Sibling Relationships and Adjustment.
Date of Parent/Participant Information Statements and Consent Form:	PIS&Consent Comparison Group dated 12 May 2003. P/GIS & Consent Comparison group v1 dated 12 May 03. PIS & Consent CF Group 12 May 2003 P/GIS & Consent v1 dated 12 May 2003
Date of Study Protocol:	Protocol dated 4 February 2003
INVESTIGATOR(S):	J Glazner, S Sawyer, B Cerritelli

DATE OF ORIGINAL APPROVAL: **13 February 2003**

DURATION: 24 Months

SIGNED: 21/5/03
COMMITTEE REPRESENTATIVE DATE

COMMENTS: Project originally approved 13/02/03 with amended documents received following audit by EHRC on 31/03/03

APPROVED SUBJECT TO THE FOLLOWING CONDITIONS:

ALL PROJECTS

1. Any proposed change in protocol and the reasons for that change, together with an indication of ethical implications (if any), must be submitted to the Ethics in Human Research Committee for approval.
2. The Principal Investigator must notify the Secretary of the Ethics in Human Research Committee, of:
   - Actual starting date of project.
   - Any adverse effects of the study on participants and steps taken to deal with them.
   - Any unforeseen events.
3. A progress report must be submitted annually and at the conclusion of the project, with special emphasis on ethical matters.

DRUG TRIALS

4. The investigators must maintain all records relating to the study for a period of 23 years.
5. The investigator(s) must report any serious adverse event experienced by any subject during the trial to the Sponsor and the Ethics in Human Research Committee within 24 hours.
6. The investigators must ensure that all externally sponsored Clinical Drug Studies have insurance coverage that is current for the entirety of the study.

Royal Children's Hospital, Melbourne

PARENT / GUARDIAN
INFORMATION STATEMENT – CF GROUP

Project No 22158A

Version 2 Date 28/7/03

Title of Project
How does living with a brother or sister with cystic fibrosis affect healthy brothers and sisters?

Thank you for taking the time to read this Information Statement.
This information statement is 5 pages long. Please make sure you have all the pages.

For people who speak languages other than English:
If you would also like information about the research and the Consent Form in your language, please ask the person explaining this project to you.

Your child is invited to participate in a Research Project that is explained below.

What is an Information Statement ?
These pages contain information about a research project we are inviting you and your child to take part in. The purpose of this information is to explain to you clearly and openly all the steps and procedures of this project. The information is to help you to decide whether or not you and/or your child would like to take part in the research.

Please read this information carefully. You can ask us questions about anything in it. You may also wish to talk about the project with others eg. friends or a health care worker. When you understand what the project is about, you can sign the consent form attached if you agree for your child to take part. You will be given a copy of this information and the consent form to keep.

What is the Research Project about?

There has been a lot of research focusing on children with cystic fibrosis (CF) but little is known about the affects of CF on the brothers and sisters. The aim of this research is to improve our understanding of the experiences and needs of brothers and sisters of young people with CF. We want to study the effect, if any, of growing up with a brother or sister with CF. We will do this by comparing a group of children with a brother or sister with CF to a group of children with brothers or sisters who do not have CF. We hope the findings of this study will help families with children newly diagnosed with CF in the future.

Who are the Researchers?

Ms Judith Glazner, Cystic Fibrosis Coordinator and Counsellor, Department of Respiratory Medicine, Royal Children's Hospital – Judith is doing this research as part of her PhD study
Associate Professor Susan Sawyer, Paediatric Respiratory Physician and adolescent Medicine Specialist, Royal Children's Hospital
Ms Belinda Cerritelli, Research Assistant, Department of Respiratory Medicine, Royal Children's Hospital

Why am I and my child being asked to be in this research project?

We are asking you to take part in this study because you are the parent of a child with CF who is between 6 and 14 years of age. We are asking your child with CF and the next oldest brother or sister also to participate.

What are my child's alternatives to participating in this project?

If your child decides not to participate in this study but would like a copy of the findings of this project please contact the principal investigator, Ms Judith Glazner.

What does my child need to do to be in this research project?

To be part of the research project, we are asking:
Child with CF to answer two questionnaires

- One about the physical, social and emotional impact of CF eg taking part in sporting activities and getting together with friends (this is done by interview for 6-11 year olds)
- One about their relationship with their older brother or sister eg how much they do together and how they feel about each other

We will also be collecting information from the hospital about your child's lung function and number of admissions to hospital.
The next oldest brother or sister to answer three questionnaires

- One about their relationship with their brother or sister with CF
- One about their emotions eg feelings of sadness and things that worry them
- One about how they deal with different social situations eg at school with friends

Mothers and Fathers
To enable us to understand how families spend their time we will ask both mothers and fathers to participate in a daily phone diary over 3 consecutive days. The scheduling of these days will be at your convenience and each diary will take about 15 minutes to do over the telephone. We will be asking you questions about the activities you have been involved in and the amount of time you spent on these.
Mothers
In addition to the above, mothers will be asked to:

- Answer one questionnaire about the behaviour of your older child and how they deal with different social situations eg interacting with friends
- To give permission for the investigators to contact the school attended by your child with CF to find out how many days absence they had in the previous school year
- To think of a student in your child with CF's class and who has an older brother or sister. We will give you some information to give to his/her parents in the hope that they will agree to participate in the study.

We will arrange for this information to be collected on the same day as your child's clinic visit to minimise inconvenience for your family. This will take about an hour in addition to your routine clinic visit.

Is there likely to be a benefit to my child?

We hope that a better understanding of the impact of CF on brothers and sisters will enable us to improve the services and supports for the families who attend the clinic.

Is there likely to be a benefit to other children in the future?

It is likely that this study will help families with children diagnosed with CF in the future. The results of this study may also be relevant to the brothers and sisters of young people with conditions other than CF..

What are the possible risks and/or side effects for my child?

As this is a questionnaire based study we do not anticipate any risks or side effects. However, all participants will be informed of the support services available should they be concerned about any aspect of taking part in this study. These services would include consultations with the RCH Mental Health Group.

What are the possible discomforts and/or inconveniences for me or my child?

The questions will take an hour to complete. The nightly diary takes 15 minutes for each parent each night for 3 nights. Where possible we will try to avoid you having to make an extra visit to the hospital

What will be done to make sure the information is confidential?

Participants' names will not be on the questionnaires. The questionnaires will be kept in a locked office at the Royal Children's Hospital for 5 years and then be disposed of by shredding. In order to improve the understanding of and support for families who have children with CF we plan to submit the results for publication in a scientific medical journal. Only summary information will be published

Will I be informed of the results when the research project is finished?

We will send you a summary of the results of the study when the project is completed. This will be in 2004.

You can decide whether or not you give permission for your child to take part in this research project.

You can decide whether or not you would like to withdraw your child from this research project at any time. No explanation is needed.

You may like to discuss your participation in this research project with your family and with your doctor. You can ask for further information before deciding if your child will take part.

If you would like more information about the study or if you need to contact a study representative in an emergency, the person to contact is :

Name: ***Judith Glazner.***

Contact telephone: ***9345 5818***

What are my child's rights as a Participant?

1. I am informed that except where stated above, no information regarding my child's medical history will be released. This is subject to legal requirements.

2. I am informed that the results of any tests involving my child will not be published so as to reveal my child's identity. This is subject to legal requirements.

3. The detail of the procedure proposed has also been explained to me. This includes how long it will take, how often the procedure will be performed and whether any discomfort will result.

4. It has also been explained that my child's involvement in the research may not be of any benefit to him or her. I understand that the purpose of this research project is to improve the quality of medical care in the future.

5. I have been asked if I would like to have a family member or a friend with me while the project is explained to me.

6. I understand that this project follows the guidelines of the National Statement on Ethical Conduct in Research Involving Humans (1999).

7. I understand that this research project has been approved by the Royal Children's Hospital Ethics in Human Research Committee on behalf of Women's and Children's Health Board.

8. I have received a copy of this document.

If you have any concerns about the study, and would like to speak to someone independent of the study, please contact Consumer Liaison, Clinical Support Services Team at the Executive Office, RCH. Telephone 9345 5676 (Monday to Friday 9am-5pm).

Royal Children's Hospital, Melbourne

STANDARD INFORMED CONSENT
FOR PARENT / GUARDIAN TO GIVE CONSENT
FOR THEIR CHILD TO PARTICIPATE IN A RESEARCH PROJECT
(Attach to Parent Information Statement)

Project No 22158A
Version 1 Dated 12/5/03

Title of Project

How does living with a brother or sister with cystic fibrosis affect healthy brothers and sisters?

Principal Investigator(s)	Ms Judith Glazner, Cystic Fibrosis Coordinator and Counsellor, Department of Respiratory Medicine, Royal Children's Hospital Associate Professor Susan Sawyer, Paediatric Respiratory Physician, Ms Belinda Cerritelli, Research Assistant, Department of Respiratory Medicine, Royal Children's Hospital

I (Parent/Guardian name) _____

Parent / Guardian of (child's name) _____
voluntarily consent to him / her taking part in the above titled Research Project, explained to me by

Mr / Ms / Dr / Professor

- I have received a Parent/Guardian Information Statement to keep and I believe I understand the purpose, extent and possible effects of my child's involvement
- I have been asked if I would like to have a family member or friend with me while the project was explained
- I have had an opportunity to ask questions and I am satisfied with the answers I have received
- I understand that the researcher has agreed not to reveal results of any information involving me/my child, subject to legal requirements
- If information about this project is published or presented in any public form, I understand that the researcher will not reveal my/my child's identity
- I understand that if I refuse to consent, or if I withdraw my child from the study at any time without explanation, this will not affect my child's access to the best available treatment options and care from Women's and Children's Health (The Royal Women's Hospital OR The Royal Children's Hospital).
- I understand I will receive a copy of this consent form.

PARENT GUARDIAN SIGNATURE _____ Date _____

I have explained the study to the parent/guardian who has signed above, and believe that they understand the purpose, extent and possible effects of their involvement in this study.

RESEARCHER'S SIGNATURE _____ Date _____

Note: All parties signing the Consent Form must date their own signature.

RCH

Royal Children's Hospital, Melbourne

PARTICIPANT INFORMATION STATEMENT – CF GROUP (CHILD FORM)

Project No 22158A
Version 2 Date 28/7/03

Title of Project
How does living with a brother or sister with cystic fibrosis affects healthy brothers and sisters?

Thank you for taking the time to read this Information Statement.
This information statement is 4 pages long. Please make sure you have all the pages.

For people who speak languages other than English:
If you would also like information about the research and the Consent Form in your language, please ask the person explaining this project to you.

You are invited to participate in a Research Project that is explained below.

What is an Information Statement ?

These pages contain information about a research project we are inviting you to take part in. The purpose of information is to explain to you clearly and openly all the steps and procedures of this project. The information is to help you to decide whether or not you would like to take part in the research.

Please read this information carefully. You can ask us questions about anything in it. You may also wish to talk about the project with your parents or guardians, friends or health care worker. When you understand what the project is about, you can sign the consent form attached if you wish to take part. You will be given a copy of this information and the consent form to keep.

What is the Research Project about?
There has been a lot of research focusing on children with Cystic Fibrosis (CF) but little is known about the affects of CF on the brothers and sisters. The aim of this research is to improve our understanding of the experiences and needs of brothers and sisters of young people with CF. We want to study the effect, if any, of growing up with a brother or sister with CF. We will do this by comparing a group of children with a brother or sister with CF to a group of children with brothers and sisters who do not have CF. We hope the findings of this study will help families with children newly diagnosed with CF in the future.

RCH

Who are the Researchers?

The researchers are:
Ms Judith Glazner, Cystic Fibrosis Coordinator and Counsellor, Department of Respiratory Medicine, Royal Children's Hospital – Judith is doing this research as part of her PhD study
Associate Professor Susan Sawyer, Paediatric Respiratory Physician and Adolescent Medicine Specialist, Royal Children's Hospital
Ms Belinda Cerritelli, Research Assistant, Department of Respiratory Medicine, Royal Children's Hospital

Why am I being asked to be in this research project?

We are asking you to take part in this study because you are either a child with CF aged between 6 and 14 years, or you are the next oldest brother or sister of a child with CF who is aged between 6 and 14 years.

What are the alternatives to participating in this project ?

If you decide not to participate in this study but would like a copy of the findings of this project please contact the principal investigator, Ms Judith Glazner.

What do I need to do to be in this research project?
To be part of the research project, we are asking:
Children with CF

- To complete questions about the physical, social and emotional impact of CF eg taking part in sporting activities and getting together with friends (this is done by interview for 6-11 year olds)
- To answer questions about your relationship with your older brother or sister eg how much they do together and how they feel about each other

We will also be collecting information from the hospital about your lung function and number of admissions to hospital.

The next oldest brother or sister

- To answer some questions about your relationship with your brother or sister with CF
- To complete questions about your emotions eg feelings of sadness and things that worry you and questions about how you deal with different social situations eg at school with friends

We will also be asking your parents to answer some questions..

Is there likely to be a benefit to me?

We hope that a better understanding of the impact of CF on brothers and sisters will enable us to improve the services and supports for the families who attend the clinic.

Is there likely to a benefit to other people in the future?

It is likely that this study will help families with children diagnosed with CF in the future. The results of this study may also be relevant to the brothers and sisters of young people with conditions other than CF.

What are the possible risks and/or side effects?
As this is a questionnaire based study we do not anticipate any risks or side effects. However, all participants will be informed of the support services available should they be concerned about any aspect of taking part in this study. These services would include consultations with the RCH Mental Health Group.

What are the possible discomforts and/or inconveniences?

The questions will take an hour to complete. Where possible we will try to avoid you having to make an extra visit to the hospital.

What will be done to make sure the information is confidential?
Participants' names will not be on the questionnaires. The questionnaires will be kept in a locked office at the Royal Children's Hospital for 5 years and then be disposed of by shredding. In order to improve the understanding of and support for families who have children with CF we plan to submit the results for publication in a scientific medical journal. Only summary information will be published

Will I be informed of the results when the research project is finished?

We will send you a summary of the results of the study when the project is completed. This will be in 2004.

You can decide whether or not to take part in this research project.

You can decide whether or not you would like to withdraw at any time without explanation.

You may like to discuss participation in this research project with your family and with your doctor. You can ask for further information before deciding to take part.

If you would like more information about the study or if you need to contact a study representative in an emergency, the person to contact is :

Name: ***Judith Glazner***

Contact telephone: ***9345 5818***

What are my rights as a Participant?

1. I am informed that except where stated above, no information regarding my medical history will be released. This is subject to legal requirements.

2. I am informed that the results of any tests involving me will not be published so as to reveal my identity. This is subject to legal requirements.

3. The detail of the procedure proposed has also been explained to me. This includes how long it will take, how often the procedure will be performed and whether any discomfort will result.

4. It has also been explained that my involvement in the research may not be of any benefit to me personally. I understand that the purpose of this research project is to improve the quality of medical care in the future.

5. I have been asked if I would like to have a family member or a friend with me while the project is explained to me.

6. I understand that this project follows the guidelines of the National Statement on Ethical Conduct in Research Involving Humans (1999).

7. I understand that this research project has been approved by the Royal Children's Hospital Ethics in Human Research Committee on behalf of Women's and Children's Health Board.

8. I have received a copy of this document.

RCH

If you have any concerns about the study, and would like to speak to someone independent of the study, please contact Consumer Liaison, Clinical Support Services Team at the Executive Office, RCH Unit. Telephone 9345 5676 (Monday to Friday 9am-5pm).

RCH

Royal Children's Hospital, Melbourne

STANDARD INFORMED CONSENT FOR PARTICIPANT TO PARTICIPATE IN A RESEARCH PROJECT

(Attach to Participant Information Statement)

Project No 22158A
Version 1 Dated 12/5/03

Title of Project

How does living with a brother or sister with cystic fibrosis affects healthy brothers and sisters?

Principal Investigator(s)	Ms Judith Glazner, Cystic Fibrosis Coordinator and Counsellor, Department of Respiratory Medicine, Royal Children's Hospital Associate Professor Susan Sawyer, Paediatric Respiratory Physician and Adolescent Medicine Specialist, Royal Children's Hospital Ms Belinda Cerritelli, Research Assistant, Department of Respiratory Medicine, Royal Children's Hospital

I, _____

voluntarily consent to taking part in this research project, which has been explained to me by

Mr / Ms / Dr / Professor _____

- I have received a Participant Information Statement to keep and I believe I understand the purpose, extent and possible effects of my involvement
- I have been asked if I would like to have a family member or friend with me while the project was explained
- I have had an opportunity to ask questions and I am satisfied with the answers I have received
- I understand that the researcher has agreed not to reveal results of any information involving me, subject to legal requirements
- If information about this project is published or presented in any public form, I understand that the researcher will not reveal my identity
- I understand that if I refuse to consent, or if I withdraw from the study at any time without explanation, this will not affect my access to the best available treatment options and care from Women's and Children's Health (The Royal Women's Hospital OR The Royal Children's Hospital).
- **I understand I will receive a copy of this consent form.**

SIGNATURE _____ Date _____

I have explained the study to the participant who has signed above, and believe that they understand the purpose, extent and possible effects of their involvement in this study.

RESEARCHER'S SIGNATURE _____ Date _____

Note: All parties signing the Consent Form must date their own signature.

RCH

Royal Children's Hospital, Melbourne

PARTICIPANT INFORMATION STATEMENT – CF GROUP (PARENT FORM)

Project No 22158A
Version 2 Date 28/7/03

Title of Project
How does living with a brother or sister with cystic fibrosis affects healthy brothers and sisters?

Thank you for taking the time to read this Information Statement.
This information statement is 4 pages long. Please make sure you have all the pages.

For people who speak languages other than English:
If you would also like information about the research and the Consent Form in your language, please ask the person explaining this project to you.

You are invited to participate in a Research Project that is explained below.

What is an Information Statement ?

These pages contain information about a research project we are inviting you to take part in. The purpose of information is to explain to you clearly and openly all the steps and procedures of this project. The information is to help you to decide whether or not you would like to take part in the research.

Please read this information carefully. You can ask us questions about anything in it. You may also wish to talk about the project with your parents or guardians, friends or health care worker. When you understand what the project is about, you can sign the consent form attached if you wish to take part. You will be given a copy of this information and the consent form to keep.

What is the Research Project about?
There has been a lot of research focusing on children with Cystic Fibrosis (CF) but little is known about the affects of CF on the brothers and sisters. The aim of this research is to improve our understanding of the experiences and needs of brothers and sisters of young people with CF. We want to study the effect, if any, of growing up with a brother or sister with CF. We will do this by comparing a group of children with a brother or sister with CF to a group of children with brothers and sisters who do not have CF. We hope the findings of this study will help families with children newly diagnosed with CF in the future.

RCH

Who are the Researchers?

The researchers are:
Ms Judith Glazner, Cystic Fibrosis Coordinator and Counsellor, Department of Respiratory Medicine, Royal Children's Hospital – Judith is doing this research as part of her PhD study
Associate Professor Susan Sawyer, Paediatric Respiratory Physician and Adolescent Medicine Specialist, Royal Children's Hospital
Ms Belinda Cerritelli, Research Assistant, Department of Respiratory Medicine, Royal Children's Hospital

Why am I being asked to be in this research project?

We are asking you to take part in this study because you are the parent of a child with CF who is aged between 6 and 14 years and an older child between 7 and 15 years who does not have CF.

What are the alternatives to participating in this project ?

If you decide not to participate in this study but would like a copy of the findings of this project please contact the principal investigator, Ms Judith Glazner.

What do I need to do to be in this research project?
To be part of the research project, we are asking:
Child with CF to answer two questionnaires

- One about the physical, social and emotional impact of CF eg taking part in sporting activities and getting together with friends (this is done by interview for 6-11 year olds)
- One about their relationship with their older brother or sister eg how much they do together and how they feel about each other

We will also be collecting information from the hospital about your child's lung function and number of admissions to hospital.
The next oldest brother or sister to answer three questionnaires

- One about their relationship with their brother or sister with CF
- One about their emotions eg feelings of sadness and things that worry them
- One about how they deal with different social situations eg at school with friends

Mothers and Fathers
To enable us to understand how families spend their time we will ask both mothers and fathers to participate in a daily phone diary over 3 consecutive days. The scheduling of these days will be at your convenience and each diary will take about 15 minutes to do over the telephone. We will be asking you questions about the activities you have been involved in and the amount of time you spent on these.
Mothers
In addition to the above, mothers will be asked to:

- Answer one questionnaire about the behaviour of your older child and how they deal with different social situations eg interacting with friends
- To give permission for the investigators to contact the school attended by your child with CF to find out how many days absence they had in the previous school year
- To think of a student in your child with CF's class and who has an older brother or sister. We will give you some information to give to his/her parents in the hope that they will agree to participate in the study.

We will arrange for this information to be collected on the same day as your child's clinic visit to minimise inconvenience for your family. This will take about an hour in addition to your routine clinic visit.

Is there likely to be a benefit to me?

We hope that a better understanding of the impact of CF on brothers and sisters will enable us to improve the services and supports for the families who attend the clinic.

Is there likely to a benefit to other people in the future?

It is likely that this study will help families with children diagnosed with CF in the future. The results of this study may also be relevant to the brothers and sisters of young people with conditions other than CF.

What are the possible risks and/or side effects?
As this is a questionnaire based study we do not anticipate any risks or side effects. However, all participants will be informed of the support services available should they be concerned about any aspect of taking part in this study. These services would include consultations with the RCH Mental Health Group.

What are the possible discomforts and/or inconveniences?

The questions will take an hour to complete. Where possible we will try to avoid you having to make an extra visit to the hospital.

What will be done to make sure the information is confidential?
Participants' names will not be on the questionnaires. The questionnaires will be kept in a locked office at the Royal Children's Hospital for 5 years and then be disposed of by shredding. In order to improve the understanding of and support for families who have children with CF we plan to submit the results for publication in a scientific medical journal. Only summary information will be published

Will I be informed of the results when the research project is finished?

We will send you a summary of the results of the study when the project is completed. This will be in 2004.

You can decide whether or not to take part in this research project.

You can decide whether or not you would like to withdraw at any time without explanation.

You may like to discuss participation in this research project with your family and with your doctor. You can ask for further information before deciding to take part.

If you would like more information about the study or if you need to contact a study representative in an emergency, the person to contact is :

Name: ***Judith Glazner***

Contact telephone: ***9345 5818***

What are my rights as a Participant?

1. I am informed that except where stated above, no information regarding my medical history will be released. This is subject to legal requirements.

2. I am informed that the results of any tests involving me will not be published so as to reveal my identity. This is subject to legal requirements.

3. The detail of the procedure proposed has also been explained to me. This includes how long it will take, how often the procedure will be performed and whether any discomfort will result.

4. It has also been explained that my involvement in the research may not be of any benefit to me personally. I understand that the purpose of this research project is to improve the quality of medical care in the future.

5. I have been asked if I would like to have a family member or a friend with me while the project is explained to me.

6. I understand that this project follows the guidelines of the National Statement on Ethical Conduct in Research Involving Humans (1999).

7. I understand that this research project has been approved by the Royal Children's Hospital Ethics in Human Research Committee on behalf of Women's and Children's Health Board.

8. I have received a copy of this document.

If you have any concerns about the study, and would like to speak to someone independent of the study, please contact Consumer Liaison, Clinical Support Services Team at the Executive Office, RCH Unit. Telephone 9345 5676 (Monday to Friday 9am-5pm).

RCH

Royal Children's Hospital, Melbourne

STANDARD INFORMED CONSENT FOR PARTICIPANT TO PARTICIPATE IN A RESEARCH PROJECT

(Attach to Participant Information Statement)

Project No 22158A
Version 1 Dated 12/5/03

Title of Project

How does living with a brother or sister with cystic fibrosis affects healthy brothers and sisters?

Principal Investigator(s)	Ms Judith Glazner, Cystic Fibrosis Coordinator and Counsellor, Department of Respiratory Medicine, Royal Children's Hospital Associate Professor Susan Sawyer, Paediatric Respiratory Physician and Adolescent Medicine Specialist, Royal Children's Hospital Ms Belinda Cerritelli, Research Assistant, Department of Respiratory Medicine, Royal Children's Hospital

I, _____
voluntarily consent to taking part in this research project, which has been explained to me by

Mr / Ms / Dr / Professor _____

- I have received a Participant Information Statement to keep and I believe I understand the purpose, extent and possible effects of my involvement
- I have been asked if I would like to have a family member or friend with me while the project was explained
- I have had an opportunity to ask questions and I am satisfied with the answers I have received
- I understand that the researcher has agreed not to reveal results of any information involving me, subject to legal requirements
- If information about this project is published or presented in any public form, I understand that the researcher will not reveal my identity
- I understand that if I refuse to consent, or if I withdraw from the study at any time without explanation, this will not affect my access to the best available treatment options and care from Women's and Children's Health (The Royal Women's Hospital OR The Royal Children's Hospital).
- **I understand I will receive a copy of this consent form.**

SIGNATURE _____ Date _____

I have explained the study to the participant who has signed above, and believe that they understand the purpose, extent and possible effects of their involvement in this study.

RESEARCHER'S SIGNATURE _____ Date _____

Note: All parties signing the Consent Form must date their own signature.

ROYAL CHILDREN'S HOSPITAL

Victorian Paediatric Respiratory Service

Royal Children's Hospital

Department of Respiratory Medicine
Royal Children's Hospital
Flemington Road, Parkville
Victoria, Australia, 3052
Telephone (+613) 9345 5844
Facsimile (+613) 9349 1289

Department of Respiratory
& Sleep Medicine
Monash Medical Centre
Clayton Road, Clayton
Victoria, Australia, 3168
Telephone (+613) 9594 2900
Facsimile (+613) 9594 6415

Dear Parents,

My name is Judith Glazner and for the past 11 years I have worked at the Royal Children's Hospital with children with cystic fibrosis and their families. Cystic fibrosis is a serious genetic condition with symptoms that usually appear shortly after birth. There has been a lot of research on children with cystic fibrosis, but little is known about the impact on other family members. Therefore, I am conducting a study specifically looking at how cystic fibrosis affects brothers and sisters to improve our understanding of their experiences and needs.

I will do this by comparing brothers and sisters of children with cystic fibrosis to brothers and sisters of children who **do not** have cystic fibrosis. The _____ family has nominated your family to be invited to participate in the comparison group of this study. This would involve you as parents, your child who is between 6 and 14 years of age and the next oldest brother or sister. A detailed description of the project is enclosed.

For privacy reasons the _____ family has not provided any information about you or your family to me. **Participation in the study is entirely voluntary and confidential.** I do, however, need to know whether or not you decide to participate. Please call me on **9375 7682** in the next 7 days with your decision or if you would like further information. If you leave a message please mention the name of the family who nominated you.

Regardless of your decision about participation in the study, I would like to thank you for taking the time to read this letter. There will be no further contact if you indicate that you do not wish to participate.

Yours sincerely,

Judith Glazner
Cystic Fibrosis Coordinator and Counsellor
& Principal Investigator

Royal Children's Hospital, Melbourne

PARENT / GUARDIAN
INFORMATION STATEMENT – COMPARISON GROUP

Project No	22158A

Version 2 Date 28/07/03

Title of Project
How does living with a brother or sister with cystic fibrosis affect healthy brothers and sisters?

Thank you for taking the time to read this Information Statement.
This information statement is 5 pages long. Please make sure you have all the pages.

For people who speak languages other than English:
If you would also like information about the research and the Consent Form in your language, please ask the person explaining this project to you.

Your child is invited to participate in a Research Project that is explained below.

What is an Information Statement ?
These pages contain information about a research project we are inviting you and your child to take part in. The purpose of this information is to explain to you clearly and openly all the steps and procedures of this project. The information is to help you to decide whether or not you and/or your child would like to take part in the research.

Please read this information carefully. You can ask us questions about anything in it. You may also wish to talk about the project with others eg. friends or a health care worker. When you understand what the project is about, you can sign the consent form attached if you agree for your child to take part. You will be given a copy of this information and the consent form to keep.

What is the Research Project about?

This project is about Cystic Fibrosis (CF). CF is a genetic condition with symptoms that usually appear shortly after birth. They include respiratory infections due to accumulation of sticky mucous, problems with digestion and excessive loss of salt in sweat. There has been a lot of research focusing on children with CF but little is known about the effects of CF on the brothers and sisters. The aim of this research is to improve our understanding of the experiences and needs of brothers and sisters of young people with CF. We want to study the effect, if any, of growing up with a brother or sister with CF. We will do this by comparing a group of children with a brother or sister with CF to a group of children with brothers and sisters who do not have CF. We hope the findings of this study will help families with children newly diagnosed with CF in the future

Who are the Researchers?

Ms Judith Glazner, Cystic Fibrosis Coordinator and Counsellor, Department of Respiratory Medicine, Royal Children's Hospital – Judith is doing this research as part of her PhD study
Associate Professor Susan Sawyer, Paediatric Respiratory Physician and adolescent Medicine Specialist, Royal Children's Hospital
Ms Belinda Cerritelli, Research Assistant, Department of Respiratory Medicine, Royal Children's Hospital

Why am I and my child being asked to be in this research project?

We are asking you to take part in this study because you are the parent of a child who is between 6 and 14 years of age. We are asking your child and the next oldest brother or sister also to participate.

What are my child's alternatives to participating in this project?

If your child decides not to participate in this study but would like a copy of the findings of this project please contact the principal investigator, Ms Judith Glazner.

What does my child need to do to be in this research project?

To be part of the research project, we are asking:
Child aged 6 – 14 to answer one questionnaire:

- about their relationship with their older brother or sister eg how much they do together and how they feel about each other

The next oldest brother or sister to answer three questionnaires:

- One about their relationship with their brother or sister
- One about their emotions eg feelings of sadness and things that worry them
- One about how they deal with different social situations eg at school with friends

Mothers and Fathers
To enable us to understand how families spend their time we will ask both mothers and fathers to participate in a daily phone diary over 3 consecutive days. The scheduling of these days will be at your convenience and each diary will take about 15 minutes to do over the telephone. We will be asking you questions about the activities you have been involved in and the amount of time you spent on these.

Mothers
In addition to the above, mothers will be asked to:

- Answer one questionnaire about the behaviour of your older child and how they deal with different social situations eg interacting with friends
- To give permission for the investigators to contact the school attended by your child to find out how many days absence they had in the previous school year

We will arrange to meet with you and your children to collect the above information in a room at the Centre for Adolescent Health, across the road from the Royal Children's Hospital.

Is there likely to be a benefit to my child?

While there is no direct benefit we hope that a better understanding of the impact of CF on brothers and sisters will enable us to improve the services and supports for the families who attend the clinic.

Is there likely to be a benefit to other children in the future?

It is likely that this study will help families with children diagnosed with CF in the future. The results of this study may also be relevant to the brothers and sisters of young people with conditions other than CF.

What are the possible risks and/or side effects for my child?

As this is a questionnaire based study we do not anticipate any risks or side effects. However, all participants will be informed of the support services available should they be concerned about any aspect of taking part in this study. These services would include consultations with the RCH Mental Health Group.

What are the possible discomforts and/or inconveniences for me or my child?

The questions will take about an hour to complete. Appointment times late in the day or during school holidays will be offered so that school absence is minimised. The nightly diary takes 15 minutes for each parent each night for 3 nights.

What will be done to make sure the information is confidential?

Participants' names will not be on the questionnaires. The questionnaires will be kept in a locked office at the Royal Children's Hospital for 5 years and then be disposed of by shredding. In order to improve the understanding of and support for families who have children with CF we plan to submit the results for publication in a scientific medical journal. Only summary information will be published

Will I be informed of the results when the research project is finished?

We will send you a summary of the results of the study when the project is completed. This will be in 2004.

You can decide whether or not you give permission for your child to take part in this research project.

You can decide whether or not you would like to withdraw your child from this research project at any time. No explanation is needed.

You may like to discuss your participation in this research project with your family and with your doctor. You can ask for further information before deciding if your child will take part.

If you would like more information about the study or if you need to contact a study representative in an emergency, the person to contact is :

Name: ***Judith Glazner.***

Contact telephone: ***9345 5818***

What are my child's rights as a Participant?

1. I am informed that except where stated above, no information regarding my child's medical history will be released. This is subject to legal requirements.

2. I am informed that the results of any tests involving my child will not be published so as to reveal my child's identity. This is subject to legal requirements.

3. The detail of the procedure proposed has also been explained to me. This includes how long it will take, how often the procedure will be performed and whether any discomfort will result.

4. It has also been explained that my child's involvement in the research may not be of any benefit to him or her. I understand that the purpose of this research project is to improve the quality of medical care in the future.

5. I have been asked if I would like to have a family member or a friend with me while the project is explained to me.

6. I understand that this project follows the guidelines of the National Statement on Ethical Conduct in Research Involving Humans (1999).

7. I understand that this research project has been approved by the Royal Children's Hospital Ethics in Human Research Committee on behalf of Women's and Children's Health Board.

8. I have received a copy of this document.

If you have any concerns about the study, and would like to speak to someone independent of the study, please contact Consumer Liaison, Clinical Support Services Team at the Executive Office, RCH. Telephone 9345 5676 (Monday to Friday 9am-5pm).

Royal Children's Hospital, Melbourne

STANDARD INFORMED CONSENT
FOR PARENT / GUARDIAN TO GIVE CONSENT
FOR THEIR CHILD TO PARTICIPATE IN A RESEARCH PROJECT
(Attach to Parent Information Statement)

Project No 22158A
Version 1 Dated 2/5/03

Title of Project

How does living with a brother or sister with cystic fibrosis affect healthy brothers and sisters?

Principal Investigator(s)	Ms Judith Glazner, Cystic Fibrosis Coordinator and Counsellor, Department of Respiratory Medicine, Royal Children's Hospital Associate Professor Susan Sawyer, Paediatric Respiratory Physician, Ms Belinda Cerritelli, Research Assistant, Department of Respiratory Medicine, Royal Children's Hospital

I (Parent/Guardian name) _____

Parent / Guardian of (child's name) _____
voluntarily consent to him / her taking part in the above titled Research Project, explained to me by

Mr / Ms / Dr / Professor

- I have received a Parent/Guardian Information Statement to keep and I believe I understand the purpose, extent and possible effects of my child's involvement
- I have been asked if I would like to have a family member or friend with me while the project was explained
- I have had an opportunity to ask questions and I am satisfied with the answers I have received
- I understand that the researcher has agreed not to reveal results of any information involving me/my child, subject to legal requirements
- If information about this project is published or presented in any public form, I understand that the researcher will not reveal my/my child's identity
- I understand that if I refuse to consent, or if I withdraw my child from the study at any time without explanation, this will not affect my child's access to the best available treatment options and care from Women's and Children's Health (The Royal Women's Hospital OR The Royal Children's Hospital).
- I understand I will receive a copy of this consent form.

PARENT GUARDIAN SIGNATURE _____ Date _____

I have explained the study to the parent/guardian who has signed above, and believe that they understand the purpose, extent and possible effects of their involvement in this study.

RESEARCHER'S SIGNATURE _____ Date _____

Note: All parties signing the Consent Form must date their own signature.

RCH – ETHICS IN HUMAN RESEARCH COMMITTEE

Part B **Research Protocol**

PROTOCOL COVER SHEET

Name of Principal Investigator:

Ms Judith Glazner

Names of Associate Investigator (s):

Associate Professor Susan Sawyer
Ms Belinda Cerritelli

Technical Title of Project:

The Impact of Cystic Fibrosis on Sibling Realtionships and Adjustment

CO-OPERATING DIVISION / SUPPORT SERVICES

SIGNATURES MUST BE OBTAINED FROM **ALL** THOSE DIVISIONS / DEPARTMENTS LIKELY TO CONTRIBUTE ANY SERVICES / RESOURCES TO THIS PROJECT.

Please check relevant Divisions / Services

☐ Paediatrics	☐ Obstetrics	☐ Gynaecology
☐ Operating Theatres	☐ Anaesthesia	☐ Pharmacy & Therapeutic Services
☐ Outpatient Services	☐ Inpatient Services	☐ Medical Records
☐ Social Work	☐ Interpreter service	☐ Nursing
☐ BioMedical Engineering	☐ Office/Lab Space	☐ Other (specify)
☐ Other (specify)	☐ Other (specify)	

SIGNATURES OF CO-OPERATING DIVISIONS / DEPARTMENTS

I certify that I have agreed to collaborate in this project within the resources of my department

Name	Signature	Division / Department	Date
See separate sheets			

SIGNATURES OF PRINCIPAL INVESTIGATOR

I undertake that I have the necessary resources to conduct this research and that I have discussed the likely impact of the project with all Divisions likely to be involved, including nursing, and have obtained their signed agreement.

Principal Investigator _____ Date 22 October 2002

SIGNATURES OF ASSOCIATE INVESTIGATOR(S)

Name	Signature	Date
Susan Sawyer		
Belinda Cerritelli		22/10/02

RCH

Royal Children's Hospital, Melbourne

PARTICIPANT INFORMATION STATEMENT – COMPARISON GROUP (CHILD FORM)

Project No 22158A
Version 2 Date 28/7/03

Title of Project
How does living with a brother or sister with cystic fibrosis affects healthy brothers and sisters?

Thank you for taking the time to read this Information Statement.
This information statement is 4 pages long. Please make sure you have all the pages.

For people who speak languages other than English:
If you would also like information about the research and the Consent Form in your language, please ask the person explaining this project to you.

You are invited to participate in a Research Project that is explained below.

What is an Information Statement ?

These pages contain information about a research project we are inviting you to take part in. The purpose of information is to explain to you clearly and openly all the steps and procedures of this project. The information is to help you to decide whether or not you would like to take part in the research.

Please read this information carefully. You can ask us questions about anything in it. You may also wish to talk about the project with your parents or guardians, friends or health care worker. When you understand what the project is about, you can sign the consent form attached if you wish to take part. You will be given a copy of this information and the consent form to keep.

What is the Research Project about?
This project is about Cystic Fibrosis (CF). CF is a genetic condition with symptoms that usually appear shortly after birth. They include respiratory infections due to accumulation of sticky mucous, problems with digestion and excessive loss of salt in sweat. There has been a lot of research focusing on children with CF but little is known about the effects of CF on the brothers and sisters. The aim of this research is to improve our understanding of the experiences and needs of brothers and sisters of young people with CF. We want to study the effect, if any, of growing up with a brother or sister with CF. We will do this by comparing a group of children with a brother or sister with CF to a group of children with brothers and sisters who do not have CF. We hope the findings of this study will help families with children newly diagnosed with CF in the future.

RCH

Who are the Researchers?

The researchers are:
Ms Judith Glazner, Cystic Fibrosis Coordinator and Counsellor, Department of Respiratory Medicine, Royal Children's Hospital – Judith is doing this research as part of her PhD study
Associate Professor Susan Sawyer, Paediatric Respiratory Physician and Adolescent Medicine Specialist, Royal Children's Hospital
Ms Belinda Cerritelli, Research Assistant, Department of Respiratory Medicine, Royal Children's Hospital

Why am I being asked to be in this research project?

We are asking you to take part in this study because you are either a child who is between 6 and 14 years of age, or the next oldest brother or sister of a child aged 6 – 14.

What are the alternatives to participating in this project ?

If you decide not to participate in this study but would like a copy of the findings of this project please contact the principal investigator, Ms Judith Glazner.

What do I need to do to be in this research project?
To be part of the research project, we are asking:
Children aged 6 – 14

- To answer questions about your relationship with your older brother or sister eg how much you do together and you feel about each other

The next oldest brother or sister

- To answer some questions about your relationship with your brother or sister
- To complete questions about your emotions eg feelings of sadness and things that worry you and questions about how you deal with different social situations eg at school with friends

We will also be asking your parents to answer some questions.

Is there likely to be a benefit to me?

While there is no direct benefit we hope that a better understanding of the impact of CF on brothers and sisters will enable us to improve the services and supports for the families who attend the clinic.

Is there likely to a benefit to other people in the future?

It is likely that this study will help families with children diagnosed with CF in the future. The results of this study may also be relevant to the brothers and sisters of young people with conditions other than CF.

What are the possible risks and/or side effects?
As this is a questionnaire based study we do not anticipate any risks or side effects. However, all participants will be informed of the support services available should they be concerned about any aspect of taking part in this study. These services would include consultations with the RCH Mental Health Group.

What are the possible discomforts and/or inconveniences?

The questions will take an hour to complete. We will arrange to meet with you and mother to collect the information in a room at the Centre for Adolescent Health, across the road from the Royal Children's Hospital.

RCH

What will be done to make sure the information is confidential?
Participants' names will not be on the questionnaires. The questionnaires will be kept in a locked office at the Royal Children's Hospital for 5 years and then be disposed of by shredding. In order to improve the understanding of and support for families who have children with CF we plan to submit the results for publication in a scientific medical journal. Only summary information will be published

Will I be informed of the results when the research project is finished?

We will send you a summary of the results of the study when the project is completed. This will be in 2004.

You can decide whether or not to take part in this research project.

You can decide whether or not you would like to withdraw at any time without explanation.

You may like to discuss participation in this research project with your family and with your doctor. You can ask for further information before deciding to take part.

If you would like more information about the study or if you need to contact a study representative in an emergency, the person to contact is :

Name: ***Judith Glazner***

Contact telephone: ***9345 5818***

What are my rights as a Participant?

1. I am informed that except where stated above, no information regarding my medical history will be released. This is subject to legal requirements.
2. I am informed that the results of any tests involving me will not be published so as to reveal my identity. This is subject to legal requirements.
3. The detail of the procedure proposed has also been explained to me. This includes how long it will take, how often the procedure will be performed and whether any discomfort will result.
4. It has also been explained that my involvement in the research may not be of any benefit to me personally. I understand that the purpose of this research project is to improve the quality of medical care in the future.
5. I have been asked if I would like to have a family member or a friend with me while the project is explained to me.
6. I understand that this project follows the guidelines of the National Statement on Ethical Conduct in Research Involving Humans (1999).
7. I understand that this research project has been approved by the Royal Children's Hospital Ethics in Human Research Committee on behalf of Women's and Children's Health Board.
8. I have received a copy of this document.

If you have any concerns about the study, and would like to speak to someone independent of the study, please contact Consumer Liaison, Clinical Support Services Team at the Executive Office, RCH Unit. Telephone 9345 5676 (Monday to Friday 9am-5pm).

RCH

Royal Children's Hospital, Melbourne

STANDARD INFORMED CONSENT FOR PARTICIPANT TO PARTICIPATE IN A RESEARCH PROJECT

(Attach to Participant Information Statement)

Project No 22158A
Version 1 Dated 12/5/03

Title of Project

How does living with a brother or sister with cystic fibrosis affects healthy brothers and sisters?

Principal Investigator(s)	Ms Judith Glazner, Cystic Fibrosis Coordinator and Counsellor, Department of Respiratory Medicine, Royal Children's Hospital Associate Professor Susan Sawyer, Paediatric Respiratory Physician and Adolescent Medicine Specialist, Royal Children's Hospital Ms Belinda Cerritelli, Research Assistant, Department of Respiratory Medicine, Royal Children's Hospital

I, _____

voluntarily consent to taking part in this research project, which has been explained to me by

Mr / Ms / Dr / Professor _____

- I have received a Participant Information Statement to keep and I believe I understand the purpose, extent and possible effects of my involvement
- I have been asked if I would like to have a family member or friend with me while the project was explained
- I have had an opportunity to ask questions and I am satisfied with the answers I have received
- I understand that the researcher has agreed not to reveal results of any information involving me, subject to legal requirements
- If information about this project is published or presented in any public form, I understand that the researcher will not reveal my identity
- I understand that if I refuse to consent, or if I withdraw from the study at any time without explanation, this will not affect my access to the best available treatment options and care from Women's and Children's Health (The Royal Women's Hospital OR The Royal Children's Hospital).
- **I understand I will receive a copy of this consent form.**

SIGNATURE _____ Date _____

I have explained the study to the participant who has signed above, and believe that they understand the purpose, extent and possible effects of their involvement in this study.

RESEARCHER'S SIGNATURE _____ Date _____

Note: All parties signing the Consent Form must date their own signature.

Royal Children's Hospital, Melbourne

PARTICIPANT INFORMATION STATEMENT – COMPARISON GROUP (PARENT FORM)

Project No 22158A
Version 2 Date 28/7/03

Title of Project
How does living with a brother or sister with cystic fibrosis affects healthy brothers and sisters?

Thank you for taking the time to read this Information Statement.
This information statement is 4 pages long. Please make sure you have all the pages.

For people who speak languages other than English:
If you would also like information about the research and the Consent Form in your language, please ask the person explaining this project to you.

You are invited to participate in a Research Project that is explained below.

What is an Information Statement ?

These pages contain information about a research project we are inviting you to take part in. The purpose of information is to explain to you clearly and openly all the steps and procedures of this project. The information is to help you to decide whether or not you would like to take part in the research.

Please read this information carefully. You can ask us questions about anything in it. You may also wish to talk about the project with your parents or guardians, friends or health care worker. When you understand what the project is about, you can sign the consent form attached if you wish to take part. You will be given a copy of this information and the consent form to keep.

What is the Research Project about?
This project is about Cystic Fibrosis (CF). CF is a genetic condition with symptoms that usually appear shortly after birth. They include respiratory infections due to accumulation of sticky mucous, problems with digestion and excessive loss of salt in sweat. There has been a lot of research focusing on children with CF but little is known about the effects of CF on the brothers and sisters. The aim of this research is to improve our understanding of the experiences and needs of brothers and sisters of young people with CF. We want to study the effect, if any, of growing up with a brother or sister with CF. We will do this by comparing a group of children with a brother or sister with CF to a group of children with brothers and sisters who do not have CF. We hope the findings of this study will help families with children newly diagnosed with CF in the future.

RCH

Who are the Researchers?

The researchers are:
Ms Judith Glazner, Cystic Fibrosis Coordinator and Counsellor, Department of Respiratory Medicine, Royal Children's Hospital – Judith is doing this research as part of her PhD study
Associate Professor Susan Sawyer, Paediatric Respiratory Physician and Adolescent Medicine Specialist, Royal Children's Hospital
Ms Belinda Cerritelli, Research Assistant, Department of Respiratory Medicine, Royal Children's Hospital

Why am I being asked to be in this research project?

We are asking you to take part in this study because you are the parent of two children aged between 6 and 15 years.

What are the alternatives to participating in this project ?

If you decide not to participate in this study but would like a copy of the findings of this project please contact the principal investigator, Ms Judith Glazner.

What do I need to do to be in this research project?
To be part of the research project, we are asking:
Child aged 6 – 14 to answer one questionnaire:

- about their relationship with their older brother or sister eg how much they do together and how they feel about each other

The next oldest brother or sister to answer three questionnaires:

- One about their relationship with their brother or sister
- One about their emotions eg feelings of sadness and things that worry them
- One about how they deal with different social situations eg at school with friends

Mothers and Fathers
To enable us to understand how families spend their time we will ask both mothers and fathers to participate in a daily phone diary over 3 consecutive days. The scheduling of these days will be at your convenience and each diary will take about 15 minutes to do over the telephone. We will be asking you questions about the activities you have been involved in and the amount of time you spent on these.

Mothers
In addition to the above, mothers will be asked to:

- Answer one questionnaire about the behaviour of your older child and how they deal with different social situations eg interacting with friends
- To give permission for the investigators to contact the school attended by your child to find out how many days absence they had in the previous school year

We will arrange to meet with you and your children to collect the above information in a room at the Centre for Adolescent Health, across the road from the Royal Children's Hospital.

Is there likely to be a benefit to me?

While there is no direct benefit we hope that a better understanding of the impact of CF on brothers and sisters will enable us to improve the services and supports for the families who attend the clinic.

Is there likely to a benefit to other people in the future?

It is likely that this study will help families with children diagnosed with CF in the future. The results of this study may also be relevant to the brothers and sisters of young people with conditions other than CF.

What are the possible risks and/or side effects?
As this is a questionnaire based study we do not anticipate any risks or side effects. However, all participants will be informed of the support services available should they be concerned about any aspect of taking part in this study. These services would include consultations with the RCH Mental Health Group.

What are the possible discomforts and/or inconveniences?

The questions will take an hour to complete. We will arrange to meet with you and mother to collect the information in a room at the Centre for Adolescent Health, across the road from the Royal Children's Hospital.

What will be done to make sure the information is confidential?
Participants' names will not be on the questionnaires. The questionnaires will be kept in a locked office at the Royal Children's Hospital for 5 years and then be disposed of by shredding. In order to improve the understanding of and support for families who have children with CF we plan to submit the results for publication in a scientific medical journal. Only summary information will be published

Will I be informed of the results when the research project is finished?

We will send you a summary of the results of the study when the project is completed. This will be in 2004.

You can decide whether or not to take part in this research project.

You can decide whether or not you would like to withdraw at any time without explanation.

You may like to discuss participation in this research project with your family and with your doctor. You can ask for further information before deciding to take part.

If you would like more information about the study or if you need to contact a study representative in an emergency, the person to contact is :

Name: ***Judith Glazner***

Contact telephone: ***9345 5818***

What are my rights as a Participant?

1. I am informed that except where stated above, no information regarding my medical history will be released. This is subject to legal requirements.

2. I am informed that the results of any tests involving me will not be published so as to reveal my identity. This is subject to legal requirements.

3. The detail of the procedure proposed has also been explained to me. This includes how long it will take, how often the procedure will be performed and whether any discomfort will result.

4. It has also been explained that my involvement in the research may not be of any benefit to me personally. I understand that the purpose of this research project is to improve the quality of medical care in the future.

5. I have been asked if I would like to have a family member or a friend with me while the project is explained to me.

6. I understand that this project follows the guidelines of the National Statement on Ethical Conduct in Research Involving Humans (1999).

7. I understand that this research project has been approved by the Royal Children's Hospital Ethics in Human Research Committee on behalf of Women's and Children's Health Board.

8. I have received a copy of this document.

If you have any concerns about the study, and would like to speak to someone independent of the study, please contact Consumer Liaison, Clinical Support Services Team at the Executive Office, RCH Unit. Telephone 9345 5676 (Monday to Friday 9am-5pm).

RCH

Royal Children's Hospital, Melbourne

STANDARD INFORMED CONSENT FOR PARTICIPANT TO PARTICIPATE IN A RESEARCH PROJECT

(Attach to Participant Information Statement)

Project No 22158A
Version 1 Dated 12/5/03

Title of Project

How does living with a brother or sister with cystic fibrosis affects healthy brothers and sisters?

Principal Investigator(s)	Ms Judith Glazner, Cystic Fibrosis Coordinator and Counsellor, Department of Respiratory Medicine, Royal Children's Hospital Associate Professor Susan Sawyer, Paediatric Respiratory Physician and Adolescent Medicine Specialist, Royal Children's Hospital Ms Belinda Cerritelli, Research Assistant, Department of Respiratory Medicine, Royal Children's Hospital

I, _____

voluntarily consent to taking part in this research project, which has been explained to me by

Mr / Ms / Dr / Professor _____

- I have received a Participant Information Statement to keep and I believe I understand the purpose, extent and possible effects of my involvement
- I have been asked if I would like to have a family member or friend with me while the project was explained
- I have had an opportunity to ask questions and I am satisfied with the answers I have received
- I understand that the researcher has agreed not to reveal results of any information involving me, subject to legal requirements
- If information about this project is published or presented in any public form, I understand that the researcher will not reveal my identity
- I understand that if I refuse to consent, or if I withdraw from the study at any time without explanation, this will not affect my access to the best available treatment options and care from Women's and Children's Health (The Royal Women's Hospital OR The Royal Children's Hospital).
- **I understand I will receive a copy of this consent form.**

SIGNATURE _____ Date _____

I have explained the study to the participant who has signed above, and believe that they understand the purpose, extent and possible effects of their involvement in this study.

RESEARCHER'S SIGNATURE _____ Date _____

Note: All parties signing the Consent Form must date their own signature.

Appendix B

Letter of permission to use the Sibling Relationship Questionnaire

UNIVERSITY OF
DENVER

Department of Psychology
2155 S. Race St.
Denver, CO 80208
303.871.2478
Fax 303.871.4747

February 19, 2003

Ms. Judith Glazner
Department of Respiratory Medicine
Royal Children's Hospital
Flemington Road
Parkville 3052
Melbourne
AUSTRALIA

Dear Ms. Glazner:

Enclosed you will find a copy of the Sibling Relationship Questionnaire (SRQ). I would be pleased to have you use it, but I do have two requests.

1) You may only want to use certain scales. I do not mind this kind of reduction, but I would appreciate it if the scales that are used are kept intact (i.e., not reducing the number of items to one or two or rewriting specific items). These kinds of changes make it difficult to compare results.

2) I would appreciate receiving information about the results of your work.

I hope you find these scales useful. This letter gives you permission to use the questionnaire. Good luck with your research!

Sincerely,

Wyndol Furman

Wyndol Furman, Ph.D.
Professor

Appendix C

Daily Phone Diary Manual

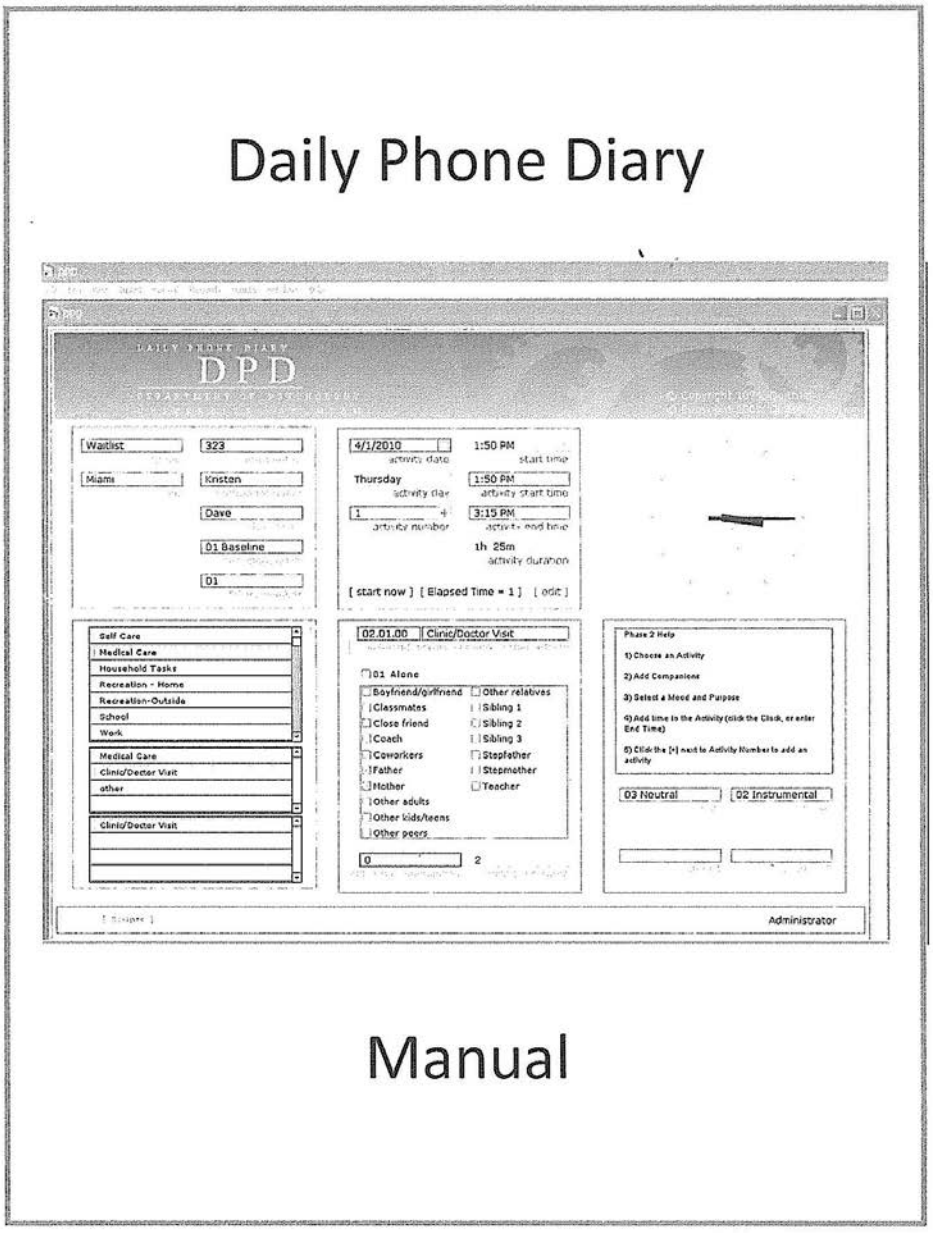


Table of Contents

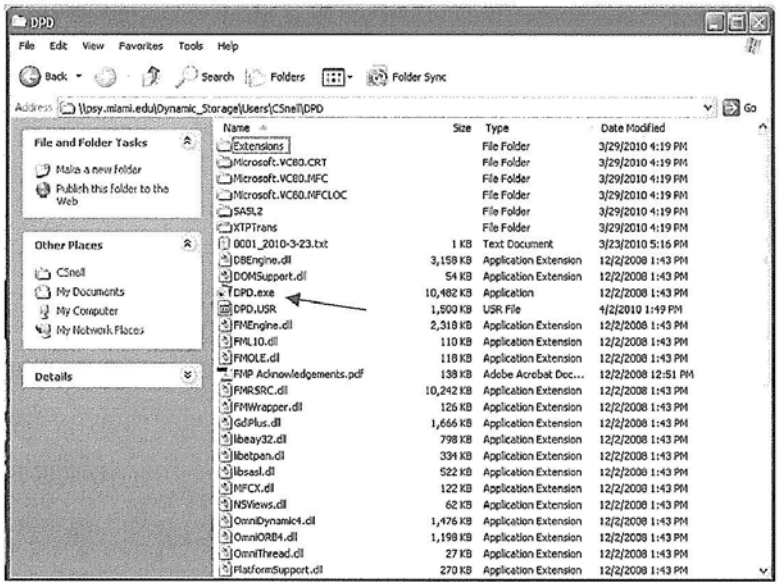
What is the Daily Phone Diary?

The Daily Phone Diary (DPD) is a form of ecological momentary assessment (EMA) that allows interviewers to assess daily activities, mood states, and other variables of interest. This information is collected by asking participants to recall the activities they engaged in over the past 24 hours, who they were with, and what their mood was like during each activity. Mood is rated on a 5-point scale (see page 15), which is provided to participants before the first phone call. The DPD can also be modified to track other variables of interest during each activity.

Phone calls typically last 10-15 minutes each. The frequency of these calls can vary depending on the objectives of the study. In prior research, the DPD has been administered to participants on 3 consecutive days at each assessment. To sample participants' activities both during the week and weekend, we have asked participants to complete calls on two weekdays and one day of the weekend. Since consecutive calls are optimal, diaries should occur on Thursday, Friday, and Saturday evenings or on Sunday, Monday, and Tuesday evenings. For a sample scheduling timeline, see page 22. If you need assistance with the Daily Phone Diary, please contact Dr. Alexandra Quittner at aquittner@miami.edu.

How to Install and Set Up the DPD

1. When you open the main folder on the DPD CD, you will see a number of files and subfolders. Select the icon "DPD.exe" to open the diary program (see screen shot below).



2. Before you begin using the DPD, you will need to import a set of activity codes. To do this, log in as an Administrator (see Administrator section on page 18) and go to the Administrator view by selecting the icon at the bottom left of the main interview screen. Then select Activities Lists and Import New Activities. You can then direct the DPD program to open a text file containing the activity codes for your study. These codes and their labels must be set apart by tabs, as shown below:

01	00	00	Self Care
01	01	00	Basic Self Care

01	01	01	getting ready for bed
01	01	02	bathing/shampooing

Instructions for Conducting the Daily Phone Diary

1. Entering Participant Information

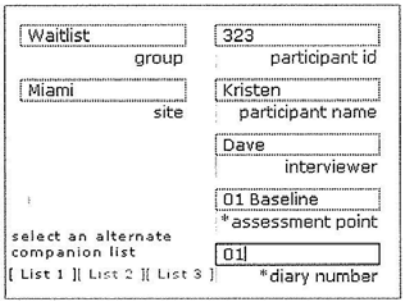
A. The person conducting the DPD should enter: a participant's group assignment (e.g., Immediate Treatment vs. Waitlist), participant ID, participant first name, Interviewer name, assessment point (e.g. Baseline), and diary number (e.g. 1 of 3 in a series). *Only the assessment point and diary number must be completed in order to begin the diary.*

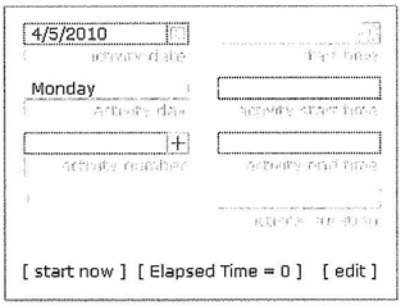
B. The program automatically records and completes the activity date and start time, using 24 hours before the Interviewer opens the diary program (since the DPD asks about activities over the last 24 hours, the clock is set to 24 hours earlier). The Interviewer can then enter activities into the DPD program during the call.

C. If you need to enter data from an call that already took place (e.g. no access to a computer at the time), click on the calendar and/or clock icons to the right of the activity date and start time to enter a different start time using a 12-hour (AM/PM) or a 24-hour (military time) clock. The DPD will give you a reminder message about setting a custom start time. Make sure the start time is appropriate for the participant's time zone.

D. Be sure you have selected the appropriate companion list in the box on the top left.

E. If at any time during the DPD, you would like to refer to a phone script (see pages 10 to 14 in the Manual), select "scripts" in the lower left corner of the screen.

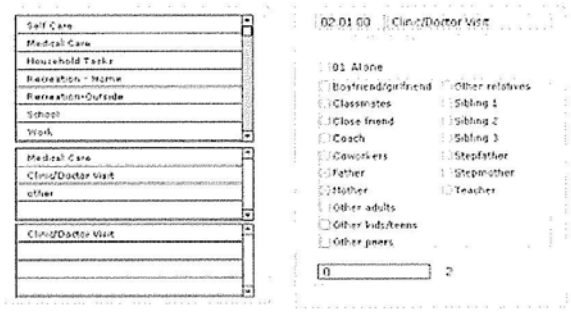




2. Entering Activities

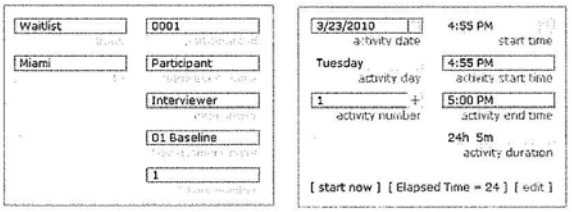
A. Start the DPD by clicking "start now" in the activity box (see screen shot on next page).

B. Enter the first activity, making sure you select an option for all three levels of the activity code (see screen shot). Once you do so, the activity name and number will appear above the text, "selected activity- or enter other activity" (see screen shot).



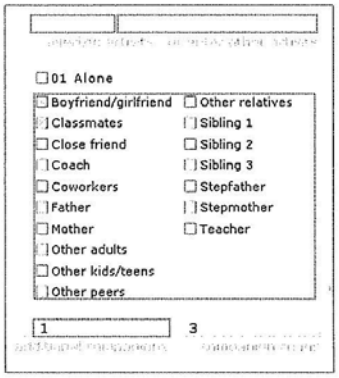
C. Complete the remaining diary fields by selecting the companions, if applicable, as well as the mood and purpose (recreational vs. instrumental) of that activity from the boxes to the right of the activity list.

D. Enter the duration of the activity by either clicking on outer edge of the clock or manually entering the activity "end time." Then select the "+" above "activity number" to enter the next activity. When 24 hours of activities have been entered, the "Elapsed Time" field, which tracks the approximate amount of time covered by the DPD so far, will turn red and read "Elapsed Time = 24" (see screen shot below). If you then try to enter another activity, a message will automatically appear stating that 24 hours are full. I for any reason, the last activity goes over24 hours, the DPD program will automatically truncate the last activity so that the duration equals 24 hours.



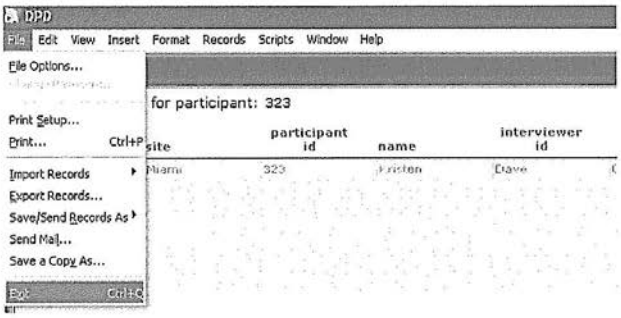
3. Entering Companion Codes

When entering information about companions, please note that if "Alone" is selected for a given activity, then other companions cannot be selected. You can select as many companions as needed for a given activity; please be sure the "companion count" is accurate. If you need to enter additional companions to make the total accurate (for example, if you click "classmates" it will record 1 classmate in the Companion count unless you change the field "additional companions") (see Screen shot).



4. Saving the Data

After the call is completed, select "Exit" from the File menu. You will be directed to a table containing the data from this DPD and given the option to save this as a text file (see below). The program will save the data in the same folder in which the DPD program was installed, with the file name "participant id_date.txt."


DPD
File Edit View Insert Format Records Scripts Window Help
File Options...
Print Setup...
Print... Ctrl+P
Import Records
Export Records...
Save/Send Records As
Send Mail...
Save a Copy As...
Exit Ctrl+Q
for participant: 323
site
participant id
name
interviewer id
Miami
323
Kristen
Dave

Editing the Data

- **To edit an activity or participant information:** To edit information about an activity, choose "edit" in the activity box. You will be directed to the output data screen (see previous page). Click on the item you would like to change and input the corrected information. When finished, click "Interview" to return to the main screen. If you need to change an activity *time*, edit this information while conducting the call. You will have to manually enter the correct time into the duration field (i.e. don't use clock), or you can delete the activity and re-enter it as explained above. If editing the Participant Information, all changes must be made in the main interview screen.

- **To delete a record:** If you wish to delete all of the data from a DPD (e.g. data were entered for training purposes), select "edit" in the activity box. Right click the space directly to the left of the record you would like to delete. Choose "Delete Record..." and then select "Delete" when asked if you would like to "Permanently delete this ENTIRE Record."

Helpful Hints for Completing the DPD

1. If a participant mentions doing two activities at once, ask which activity they were *primarily* engaged in. If they report engaging in two activities simultaneously over a long time interval, consider asking if one of these activities was primary during the first part of that time and another activity was primary for the second part of that time.

2. If the participant reports that his/her mood changed during the course of an activity, enter the same activity information twice, with two different mood ratings, as if they were two separate activities. Ask how long the participant experienced each mood state, and who the participant was with during each mood (companions)

3. Categorize activities as either "recreational" (for fun) or "instrumental" (necessary). If this is not clear (e.g., driving, shopping), then ask: "Was this an activity you were doing for fun or because you needed to"

4. A companion is counted if the person was in the same area and interaction with the participant for at least half of the duration of the activity (adolescent reading in same room as Mom is cooking, and they are talking together).

Sample Scripts:
Beginning and Ending Phone Calls

First Night Script

INTRODUCTION - CALL 1

"Hello, may I please speak with _____participant's name_____? This is ___your first name____ calling from the _____ Study.

As you know, we scheduled today for our first phone call. Is this still an okay time for you?"

NO: Find out what the problem is and determine whether or not you should continue,, call back later in the evening, or reschedule for another day.

YES: "Good." Continue

"You were given a packet containing a mood scale. This packet will help with the call. Do you have it with you?"

NO: "Can you find the packet in a minute or so?" (If it is lost, encourage the participant to try to find it, but let the person know it is not a problem, you will just read the choices over the phone.) You could also ask them to write down the mood scale choices.

YES: "Good." Go to Script – Call 1

SCRIPT - CALL 1

"Today I will be asking you about the types of activities you've done over the past 24 hours. So during this phone call, I'm going to ask you to think about the period of time from __current time p.m. yesterday to __current time__ today."

"Now, to help you remember that whole time better, I'd like you to think back to __current time__ yesterday and tell me what you were doing."

"How about today, right before I called?"

"Those activities will mark the period of time I will be asking you about."

"I will track you through all the activities you did during this time period.
I'm interested in each activity that lasted about 5 minutes or more. Things like making dinner, talking to a friend, or going to the store."

"For each activity, I will ask you how long it took, who else was with you, and how positive or negative your mood was then."

"In your packet, you will find a mood scale. I'd like you to use that scale to rate how positive or negative your mood was during each activity."

"Do you have any questions? All right, let's get started."

Second and Third Night Scripts

INTRODUCTION – CALL 2/3

"Hello, can I please speak with ____participant's name_____? This is ___your first name___ calling from the _____ Study. As you know, we scheduled today for our second/third phone call. Is this still an okay time for you?"

NO: Find out what the problem is and determine whether or not you should continue with the phone call, call back later in the evening, or reschedule for a new day.

YES: "Good." Continue

"Do you have the packet handy?"

NO: "Can you find the packet in a minute or so?" (If it is lost, encourage the participant to try to find it, but let the person know it is not a problem, you will just read the choices over the phone.) You could also ask them to write down the mood scale choices.

YES: "Good." Go to Script

SCRIPT - CALL 2/3

"We'll be doing the same thing tonight that we've done before. I will be asking you about the types of activities you did over the past 24 hours. So during this phone call I'm going to ask you to think about the period of time from __current time__ yesterday to __current time__ today."

"Now, to help you remember that whole time better, I'd like you to think back to __current time__ yesterday and tell me what you were doing."

"How about today right before I called?"

"Those activities will mark out the period of time I will be asking you about."

"Just to remind you, I'm interested in each activity that you did during this time period that lasted 5 minutes or more. For each activity, I'll ask you to tell me how long it took, who you were with and how positive or negative your mood was during that time."

"Do you have any questions? O.K., if you could open your packet to the mood scale we can get started."

Ending Call Scripts

ENDING - CALLS 1/2

"Those are all the questions that I have for tonight. Thank you so much for talking with me. Our next call is scheduled for _____. Is that okay? Do you have any questions? Thank you again- we really appreciate your help."

ENDING - CALL 3

"All right, those are all the questions for tonight and this is our last phone call of this series. Thank you so much for talking with me each evening- we really appreciate your participation in this study. I will be scheduling another series of these phone calls with you in _____, but I may not be the one calling you. However, we will call you in advance to schedule a time that is convenient for you. Do you have any questions?

Thank you again!"

Mood Scale

5- Very positive

Frequently Asked Questions

QUESTION: What should I do if I call and no one answers the phone?

ANSWER: Try calling the participant approximately 5-10 minutes later. If no one answers the second time, try again every 30-60 minutes until you reach the participant or it is too late in the evening to complete the call. If you have other diaries scheduled for that participant in that assessment series, complete those diaries and consider making up the missed one by calling on another weekend or weekday, depending on which day was missed.

QUESTION: What should I do if I am unable to contact a participant at the same time on the second night (ex. - called 8 PM Friday night but not able speak with them until 9 PM on Sat).

ANSWER: Ask the participant to recall their day starting from the time atwhich you are speaking to them the second day. If another DPD is scheduled for the following day (a third day), be sure to begin the third diary so that the time frame being recalled does not overlap with the second day.

QUESTION: What should I do if I am repeatedly unable to reach a participant?

ANSWER: A. Try calling at different times to reschedule the DPDs.

B. Try calling alternative phone numbers (e.g., parent).

C. Contact the participant via their research or medical team.

D. Use texting or email if the participant has given permission.

E. Be sure to project enthusiasm and a sense of urgency about completing the calls.

QUESTION: How can Interviewers train to use the DPD?

ANSWER: We recommend that Interviewers anticipate that diaries will take longer to complete at first, and have found practicing "mock diaries" to be helpful in familiarizing Interviewers with the DPD procedures and program.

QUESTION: What should I do if my computer crashes, or the program will not work during a call?

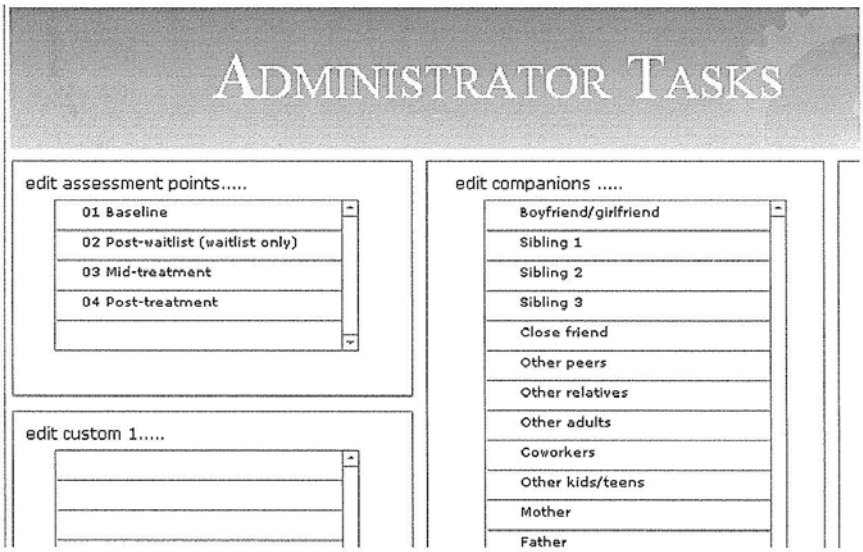
ANSWER: You can always record the DPD information by writing it down during the call. Make sure you ask the following: type of activity, duration, companions, mood, and activity purpose. Later, you can enter the activities into the DPD program using a custom start time (see page 5).

Instructions for Study Administrator

Although most of this manual was written for Interviewers conducting the DPD, there are several options in the DPD program that are designed for use by the Study Administrator. To use these functions, you will need to log in as an "Administrator."

Editing Assessment Points, Companions, Moods and Purposes

Once you are logged in as an Administrator, you can make changes to the assessment point, companions, mood, and purposes menus to tailor the DPD to your study. Simply select "Administrator" in the lower right corner of the interview screen, and you will be directed to the Administrator view where this information can be modified by clicking "edit" (see screen shot below). If you need assistance, select "Help Messages" at the bottom left of the screen. Once you are finished, you can select "Interview Screen" to return to the DPD.



Activity Codes

Activity codes can also be edited within the Administrator view. If the edit is minor (e.g. correcting a spelling error) then these changes can be made by simply selecting "Activities List" at the bottom of the Administrator screen, selecting the activity you want to edit, and making any necessary changes. However, if you want to add, remove, or re-categorize activities, it is recommended to edit the activity codes in the text file and re-import them. To do this, select "Import New Activities" on the Sdministrator activities screen. You will be prompted to back up your current activities list, which you can do by selecting "Backup Current Activities" on the same screen. Next, select the file to import. Importing activities will only work if the activities are in a tab-delimited text file (extension: .txt). There should be tabs between each level of the activity and the activity name. For example, the first few lines of your text file might look like this:

01	00	00	Self Care
01	01	00	Basic Self Care
01	01	01	getting ready for bed
01	01	02	bathing/shampooing

The first column represents the most general category of activity, the second column the second the sub-category, and the third the specific activity.

Custom Codes

Custom codes are codes that you can create depending on the needs of the study. They can be used during every activity or just some activities. For example, if you were interested in anxiety levels throughout the day, you might create a custom code for anxiety with different levels of intensity (high, medium, low) that could be coded for each activity or a subset of activities. These custom codes can be created and modified within the Administrator view.

Analyzing DPD Data

The DPD has been used in a variety of different studies, asking different research questions. Thus, we cannot provide a guide for conducting analyses. One option is to write code within SAS or another statistical program extracts the codes of interest and then potentially aggregates the activities by category. For example, if you were using the DPD to measure medical adherence within cystic fibrosis and you wanted to know how much time participants spend doing treatments each day, you might write a code that would aggregate all of the medical treatment activity codes.

Call Log Sheet

Interviewer _____ Participant Name:_____

Participant ID _____ **Date of Initial DPD**_____

	First Call	*Second Call*	*Third Call*	*Attempts*
Assessment 1				
Assessment 2				
Assessment 3				
Assessment 4				

COMMENTS:

Example Scheduling Timelines

3-day diaries (2 weekdays, 1 weekend day)

	First Call	Second Call	Third Call
Option 1	Thursday PM	Friday PM	Saturday PM
Option 2	Friday AM	Saturday AM	Sunday AM
Option 3	Sunday PM	Monday PM	Tuesday PM
Option 4	Monday AM	Tuesday AM	Wednesday AM

** Keep in mind that teens are not available on weekday mornings during the school year. Make sureall participants can complete the diaries at the scheduled times.

Sample Daily Phone Diary Activity Codes

01 00 00 "Self Care"
- 01 01 00 "Basic Self Care"
  - 01 01 01 "getting ready for bed"
  - 01 01 02 "bathing/shampooing"
  - 01 01 03 "getting ready for day/activity (dressing, etc.)"
  - 01 01 04 "getting a haircut"
  - 01 01 05 "going to beautician (getting nails done, etc.)
  - 01 01 06 "getting a massage"
  - 01 01 07 "napping/resting"
  - 01 01 08 "other"
- 01 02 00 "Facilitating Own Activities"
  - 01 02 01 "talking to teacher, babysitter, coach"
  - 01 02 02 "arranging transportation"
  - 01 02 03 "planning own activities"
  - 01 02 04 "using the internet not for recreation"
  - 01 02 05 "arranging finances/doing bills"
  - 01 02 06 "other"
- 01 03 00 "Self-Focused Activities (Non-Play)"
  - 01 03 01 "talking with parent"
  - 01 03 02 "taking a lesson (e.g., driving or music lesson)"
  - 01 03 03 "practicing for a lesson"
  - 01 03 04 "church/Sunday school"
  - 01 03 05 "praying/reading the Bible/other religious rituals"
  - 01 03 06 "youth group"
  - 01 03 07 "meditation/yoga"
  - 01 03 08 "thinking about own interests or problems"
  - 01 03 09 "other"

02 00 00 "Medical Care"
- 02 01 00 "Clinic/Doctor Visit"
- 02 02 00 "other"

03 00 00 "Household Tasks"
- 03 01 00 "Chores"
  - 03 01 01 "cleaning"
  - 03 01 02 "laundry"
  - 03 01 03 "yard work"
  - 03 01 04 "dishes"
  - 03 01 05 "repairs"
  - 03 01 06 "pet care"
  - 03 01 07 "washing car"
  - 03 01 08 "putting groceries away"

03 01 09 “shopping (e.g. for school supplies)”
03 01 10 "other"
03 02 00 "Errands"
03 02 01 "banking"
03 02 02 "grocery shopping"
03 02 03 "transporting siblings/family"
03 02 04 "errands for parents/family (not driving)"
03 02 05 "buying gifts"
03 02 06 "other"
03 03 00 "Preparing Meals"
03 03 01 "cooking"
03 03 02 "ordering food"
03 03 03 "picking up food at restaurant/drive-thru"
03 04 00 "Eating Meals"
03 04 01 "with TV or Videos"
03 04 02 "Snack"
03 04 03 "Eating at Drive-Thru or in Car"
03 05 00 "Driving for Errands"
03 06 00 "Talking and Discussing Household Plans"
03 07 00 “Other”
04 00 00 "Recreation - Home"
04 01 00 "TV or Videos"
04 02 00 "Reading"
04 02 01 "books"
04 02 02 "newspaper"
04 02 03 "magazines"
04 02 04 “comics”
04 02 05 "other"
04 03 00 "Computer/Internet/Nintendo Games"
04 04 00 "Talking on Phone for Pleasure"
04 05 00 “Texting for pleasure”
04 06 00 “Chatting online for pleasure”
04 07 00 "Listening to Music"
04 08 00 “Playing/Creating Music”
04 09 00 "Baking"
04 10 00 "Arts & Crafts (coloring/drawing/sewing, etc.)"
04 11 00 “Writing for Pleasure”
04 11 01 “creative writing (stories, poems, etc.)”
04 11 02 “writing in a journal/diary”
04 11 03 “writing a letter for pleasure”
04 12 00 "Gardening"
04 13 00 "Card/Board Games/Other Indoor Games (ping pong, pool, darts, etc.)"
04 14 00 "Having People Over"
04 14 01 "party/BBQ"

04 14 02 "sleep over"
04 14 03 "friend(s) for dinner"
04 14 04 "other"
04 15 00 "Talking & Discussing for Pleasure"
04 16 00 "Exercising at Home"
04 17 00 "Playing Sports at Home"
04 18 00 "Playing with Pet"
04 19 00 "Making Out/Sexual Activities"
04 20 00 "Eating for pleasure inside"
04 21 00 "Dancing"
04 22 00 "Other"
05 00 00 "Recreation-Outside"
05 01 00 "Shopping"
05 02 00 "Party"
05 03 00 "Attending a Wedding, Baptism, or Confirmation"
05 04 00 "Attending a Funeral or Visiting a Graveyard"
05 05 00 "Eating at a Restaurant"
05 06 00 "Movies"
05 07 00 "Concert/Play"
05 08 00 "Going to a Museum or Exhibit"
05 09 00 "Watching a Sporting Event"
05 09 01 "basketball"
05 09 02 "football"
05 09 03 "baseball"
05 10 00 "Playing a Sport"
05 10 01 "individual"
05 10 02 "team"
05 11 00 "Hiking, Hunting, Fishing, Camping"
05 12 00 "Boating, Swimming, Other Water Sports"
05 13 00 "Exercise"
05 13 01 "going to the gym"
05 13 02 "lifting weights"
05 13 03 "jogging"
05 13 04 "aerobics"
05 13 05 "walking"
05 13 06 "skateboarding"
05 13 07 "biking"
05 13 08 "skating/rollerblading"
05 13 09 "other"
05 14 00 "Going to a Park/Picnic"
05 15 00 "Going to the Beach"
05 16 00 "Going to a Carnival, Circus, Fair, Zoo, or Amusement Park"
05 17 00 "Visiting Friends"
05 17 01 "eating snack/meal while visiting"

05 18 00 "Visiting Relatives"
05 18 01 "eating snack/meal while visiting"
05 19 00 "Hobby or Club Meetings"
05 20 00 "Attending a Convention or Revival"
05 21 00 "Walking or Playing with Pet"
05 22 00 "Volunteer Work in the Community"
05 23 00 "Making Out/Sexual Activities Outside Home"
05 24 00 "Driving FOR Recreation"
05 25 00 "Driving TO/FROM Recreational Activities"
05 26 00 "Traveling TO/FROM Recreational Activities (on bus, plane, etc.)"
05 27 00 "Making a Phone Call to Home"
05 28 00 "Eating for pleasure outside"
05 29 00 "Going on a Date"
05 30 00 "Appreciating Nature"
05 31 00 "Tanning"
05 32 00 "Competitive Racing"
05 33 00 "Going to Lectures/Hearing Speakers for Recreation"
05 34 00 "Having Tea/Coffee with Friends"
05 35 00 "Protesting Social, Political, or Environmental Conditions"
05 36 00 "Being Honored/Attending a Banquet"
05 37 00 "Other"
06 00 00 "School"
06 01 00 "Getting To/From School"
06 01 01 "Getting to/from school in a car"
06 01 02 "Getting to/from school on a school bus"
06 02 00 "Attending School"
06 02 01 "Listening in class"
06 02 02 "Doing in-class assignments (not homework) individually"
06 03 03 "Working in a group"
06 02 04 "Gym class"
06 02 05 "Spending time with friends"
06 02 06 "Eating meal at School"
06 02 07 "Taking at test"
06 02 08 "Talking to the counselor/school nurse"
06 02 09 "Giving a presentation"
06 02 10 "Going to a vocational school"
06 02 11 "Other activities at school"
06 03 00 "Doing Homework (at school or home)"
06 04 00 "After School Activities"
06 04 01 "Clubs"
06 04 02 "Practicing Sports"
06 04 03 "Competing in Sports"
06 04 04 "Practicing performing arts"
06 04 05 "Putting on a Performance"

06 04 06 “Tutoring”
06 04 07 “School Function (Dance, etc.)
06 04 08 “Internship/Research”
06 04 09 “Other after school activities”
07 00 00 "Work"
07 01 00 "Getting To/From Work"
07 02 00 "Working"
07 03 00 "Taking a Break at Work"
07 04 00 "Eating Meal at Work"
08 00 00 "Other Interactions/Activities"
08 01 00 "Being Lectured/Yelled At by Parents or Adults"
08 02 00 “Being in an Argument/Fight”
08 03 00 “Resolving an Argument/Fight”
08 04 00 "Crying/Feeling Sad or Upset"
08 05 00 "Smoking"
08 06 00 "Using Alcohol/Drugs"
08 07 00 “Shoplifting”
08 08 00 “Gambling”
08 09 00 “Meeting New People”
08 10 00 “Military Activities (JROTC, etc.)”
08 11 00 "other"
09 00 00 "Research/Treatment Related Activities"
09 01 00 "Doing the DPD"
09 02 00 "Completing Forms"
09 02 01 “Doing mood ratings”
09 02 02 “Doing cognitive reframing exercises”
09 02 03 “Doing other forms”
09 03 00 “Doing exposure homework”
09 03 01 “Exposure- at home”
09 03 02 “Exposure- at school”
09 03 03 “Exposure-elsewhere”
09 04 00 “Individual Therapy Session”
09 05 00 “Group Therapy Session”
09 06 00 “Psychiatrist Visit”
09 07 00 “Other”
10 00 00 "Sleep"
10 01 00 “Trying to sleep”
10 02 00 “Sleeping”
11 00 00 "Other"

Previous Studies Using the DPD

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Quittner, A.L., Espelage, D.L., Opipari, L.C., Carter, B., Eid, N., & Eigen, H. (1998). Role Strain in Couples With and Without a Child with a Chronic Illness: Associations with Marital Satisfaction, Intimacy, and Daily Mood. *Health Psychology, 17*(2), 112-124.

Wiener, L., Riekert, K., Ryder, C., & Wood, L.V. (2004). Assessing Medication Adherence in Adolescents with HIV when Electronic Monitoring is not Feasible. *AIDS Patient Care and STDs, 18*(9), 527-538.

Modi, A.C., Lim, C.S., Yu, N., Geller, D., Wagner, M.H., & Quittner, A.L. (2006). A Multi-Method Assessment of Treatment Adherence for Children with Cystic Fibrosis. *Journal of Cystic Fibrosi, 5*(3), 177-185.

Modi, A.C. & Quittner, A.L. (2006). Barriers to Treatment Adherence for Children with Cystic Fibrosis and Asthma: What Gets in the Way? *Journal of Pediatric Psychology, 31*(8), 846-858.

Quittner, A.L., Modi, A.C., Lemanek, K.L., Ievers-Landis, C.E., & Rapoff, M.A. (2007). Evidence-based Assessment of Adherence to Medical Treatments in Pediatric Psychology. *Journal of Pediatric Psychology, 33*(9), 916-936.

Modi, A.C., Marciel, K.K., Slater, S.K., Drotar, D., & Quittner, A.L. (2008). The Influence of Parental Supervision on Medical Adherence in Adolescents with Cystic Fibrosis: Developmental Shifts from Pre to Late Adolescence. *Children's Health Care, 37*(1), 78-92.

Appendix D

Study questionnaires

1

ID # _____ GROUP_____

Sibling Relationship Questionnaire - Revised (Child) 3/90

My name is _____(completed by)

The phrase "this sibling" refers to _____(completed about)

---

1. Some siblings do nice things for each other a lot, while other siblings do nice things for each other a little. How much do both you and this sibling do nice things for each other?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
2. Who usually gets treated better by your mother, you or this sibling?	[ ]My sibling almost always gets treated better [ ]My sibling often gets treated better [ ]We get treated about the same [ ]I often get treated better [ ]I almost always get treated better
3. How much do you show this sibling how to do things he or she doesn't know how to do?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
4. How much does this sibling show you how to do things you don't know how to do?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
5. How much do you tell this sibling what to do?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much

6. How much does this sibling tell you what to do?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
7. Who usually gets treated better by your father, you or this sibling?	[ ]My sibling almost always gets treated better [ ]My sibling often gets treated better [ ]We get treated about the same [ ]I often get treated better [ ]I almost always get treated better
8. Some siblings care about each other a lot while other siblings don't care about each other that much. How much do you and this sibling care about each other?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
9. How much do you and this sibling go places and do things together?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
10. How much do you and this sibling insult and call each other names?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
11. How much do you and this sibling like the same things?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
12. How much do you and this sibling tell each other everything?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much

13. Some siblings try to out-do or beat each other at things a lot, while other siblings try to out-do each other a little. How much do you and this sibling try to out-do each other at things?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
14. How much do you admire and respect this sibling?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
15. How much does this sibling admire and respect you?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
16. How much do you and this sibling disagree and quarrel with each other?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
17. Some siblings cooperate a lot, while other siblings cooperate a little. How much do you and this sibling cooperate with each other?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
18. Who gets more attention from your mother, you or this sibling?	[ ]My sibling almost always gets more attention [ ]My sibling often gets more attention [ ]We get about the same amount of attention [ ]I often get more attention [ ]I almost always get more attention
19. How much do you help this sibling with things he or she can't do by him or herself?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much

20. How much does this sibling help you with things you can't do by yourself?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
21. How much do you make this sibling do things?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
22. How much does this sibling make you do things?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
23. Who gets more attention from your father, you or this sibling?	[ ]My sibling almost always gets more attention [ ]My sibling often gets more attention [ ]We get about the same amount of attention [ ]I often get more attention [ ]I almost always get more attention
24. How much do you and this sibling love each other?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
25. Some siblings play around and have fun with each other a lot, while other siblings play around and have fun with each other a little. How much do you and this sibling play around and have fun with each other?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
26. How much are you and this sibling mean to each other?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much

27. How much do you and this sibling have in common?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
28. How much do you and this sibling share secrets and private feelings?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
29. How much do you and this sibling compete with each other?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
30. How much do you look up to and feel proud of this sibling?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
31. How much does this sibling look up to and feel proud of you?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
32. How much do you and this sibling get mad at and get in arguments with each other?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
33. How much do both you and your sibling share with each other?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
34. Who does your mother usually favor, you or this sibling?	[ ]My sibling almost always is favored [ ]My sibling is often favored [ ]Neither of us is favored [ ]I am often favored [ ]I am almost always favored

35. How much do you teach this sibling things that he or she doesn't know?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
36. How much does this sibling teach you things that you don't know?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
37. How much do you order this sibling around?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
38. How much does this sibling order you around?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
39. Who does your father usually favor, you or this sibling?	[ ]My sibling almost always is favored [ ]My sibling is often favored [ ]Neither of us is favored [ ]I am often favored [ ]I am almost always favored
40. How much is there a strong feeling of affection (love) between you and this sibling?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
41. Some kids spend lots of time with their siblings, while others don't spend so much. How much free time do you and this sibling spend together?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
42. How much do you and this sibling bug and pick on each other in mean ways?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much

43. How much are you and this sibling alike?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
44. How much do you and this sibling tell each other things you don't want other people to know?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
45. How much do you and this sibling try to do things better than each other?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
46. How much do you think highly of this sibling?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
47. How much does this sibling think highly of you?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much
48. How much do you and this sibling argue with each other?	[ ]Hardly at all [ ]Not too much [ ]Somewhat [ ]Very much [ ]EXTREMELY much

ID No._____

Name:_____

Age:_____ Birthdate:_____

Grade in school:_____ Sex:_____

Today's date:_____

CDI

by Maria Kovacs, Ph.D.

Kids sometimes have different feelings and ideas.

This form lists the feelings and ideas in groups. From each group of three sentences, pick one sentence that describes you ***best*** for the past two weeks. After you pick a sentence from the first group, go on to the next group.

There is no right or wrong answer. Just pick the sentence that best describes the way you have been recently. Put a mark like this ☒ next to your answer. Put the mark in the box next to the sentence that you pick.

Here is an example of how this form works. Try it. Put a mark next to the sentence that describes you ***best***.

Example:

- ☐ I read books all the time.
- ☐ I read books once in a while.
- ☐ I never read books.

Remember, pick out the sentences that describe you best in the PAST TWO WEEKS.

Item 1
- ☐ I am sad once in a while.
- ☐ I am sad many times.
- ☐ I am sad all the time.

Item 2
- ☐ Nothing will ever work out for me.
- ☐ I am not sure if things will work out for me.
- ☐ Things will work out for me O.K.

Item 3
- ☐ I do most things O.K.
- ☐ I do many things wrong.
- ☐ I do everything wrong.

Item 4
- ☐ I have fun in many things.
- ☐ I have fun in some things.
- ☐ Nothing is fun at all.

Item 5
- ☐ I am bad all the time.
- ☐ I am bad many times.
- ☐ I am bad once in a while.

Item 6
- ☐ I think about bad things happening to me once in a while.
- ☐ I worry that bad things will happen to me.
- ☐ I am sure that terrible things will happen to me.

Item 7
- ☐ I hate myself.
- ☐ I do not like myself.
- ☐ I like myself.

Item 8
- ☐ All bad things are my fault.
- ☐ Many bad things are my fault.
- ☐ Bad things are not usually my fault.

Item 9
- ☐ I do not think about killing myself.
- ☐ I think about killing myself but I would not do it.
- ☐ I want to kill myself.

Turn over and fill out the other side.

Remember, pick out the sentences that describe you best in the PAST TWO WEEKS.

Item 10
- ☐ I feel like crying every day.
- ☐ I feel like crying many days.
- ☐ I feel like crying once in a while.

Item 11
- ☐ Things bother me all the time.
- ☐ Things bother me many times.
- ☐ Things bother me once in a while.

Item 12
- ☐ I like being with people.
- ☐ I do not like being with people many times.
- ☐ I do not want to be with people at all.

Item 13
- ☐ I cannot make up my mind about things.
- ☐ It is hard to make up my mind about things.
- ☐ I make up my mind about things easily.

Item 14
- ☐ I look O.K.
- ☐ There are some bad things about my looks.
- ☐ I look ugly.

Item 15
- ☐ I have to push myself all the time to do my schoolwork.
- ☐ I have to push myself many times to do my schoolwork.
- ☐ Doing schoolwork is not a big problem.

Item 16
- ☐ I have trouble sleeping every night.
- ☐ I have trouble sleeping many nights.
- ☐ I sleep pretty well.

Item 17
- ☐ I am tired once in a while.
- ☐ I am tired many days.
- ☐ I am tired all the time.

Item 18
- ☐ Most days I do not feel like eating.
- ☐ Many days I do not feel like eating.
- ☐ I eat pretty well.

Item 19
- ☐ I do not worry about aches and pains.
- ☐ I worry about aches and pains many times.
- ☐ I worry about aches and pains all the time.

Item 20
- ☐ I do not feel alone.
- ☐ I feel alone many times.
- ☐ I feel alone all the time.

Item 21
- ☐ I never have fun at school.
- ☐ I have fun at school only once in a while.
- ☐ I have fun at school many times.

Item 22
- ☐ I have plenty of friends.
- ☐ I have some friends but I wish I had more.
- ☐ I do not have any friends.

Item 23
- ☐ My schoolwork is alright.
- ☐ My schoolwork is not as good as before.
- ☐ I do very badly in subjects I used to be good in.

Item 24
- ☐ I can never be as good as other kids.
- ☐ I can be as good as other kids if I want to.
- ☐ I am just as good as other kids.

Item 25
- ☐ Nobody really loves me.
- ☐ I am not sure if anybody loves me.
- ☐ I am sure that somebody loves me.

Item 26
- ☐ I usually do what I am told.
- ☐ I do not do what I am told most times.
- ☐ I never do what I am told.

Item 27
- ☐ I get along with people.
- ☐ I get into fights many times.
- ☐ I get into fights all the time.

ID No. _____

Social Skills

Student Form
Elementary Level

Rating System

Grades 3-6

Social Skills Questionnaire

Frank M. Gresham and Stephen N. Elliott

Directions

First write the information about yourself in the box below. Then turn to page 2.

Student Information

Name _____
First Middle Last

☐ Boy ☐ Girl Today's date _____
Month Day Year

Grade _____ Age _____ Birth date _____
Month Day Year

School _____

Teacher's name _____

Form: SE

This paper lists a lot of things that students your age may do. Please read each sentence and think about yourself. Then decide **how often** you do the behavior described.

If you **never** do this behavior, circle the **0.**
If you **sometimes** do this behavior, circle the **1.**
If you **very often** do this behavior, circle the **2.**

Here are two examples:

	How Often?		
	Never	Sometimes	Very Often
I start conversations with classmates.	0	1	(2)
I keep my desk clean and neat.	0	(1)	2

*This student **very often** starts conversations with classmates. This student keeps his or her desk clean and neat **sometimes.***

If you change an answer, be sure to erase completely. Please answer all questions. When you are finished, wait for further directions from your teacher.

Be sure to ask questions if you do not know what to do. There are no right or wrong answers, just your feelings of how often you do these things.

Begin working when told to do so.

FOR OFFICE USE ONLY How Often? C	A	E	S	Social Skills	How Often? Never	Sometimes	Very Often
				1. I make friends easily.	0	1	2
				2. I smile, wave, or nod at others.	0	1	2
				3. I ask before using other people's things.	0	1	2
				4. I ignore classmates who are clowning around in class.	0	1	2
				5. I feel sorry for others when bad things happen to them.	0	1	2
				6. I tell others when I am upset with them.	0	1	2
				7. I disagree with adults without fighting or arguing.	0	1	2
				8. I keep my desk clean and neat.	0	1	2
				9. I am active in school activities such as sports or clubs.	0	1	2
				10. I do my homework on time.	0	1	2
				11. I tell new people my name without being asked to tell it.	0	1	2
				12. I control my temper when people are angry with me.	0	1	2
				13. I politely question rules that may be unfair.	0	1	2
				14. I let friends know I like them by telling or showing them.	0	1	2
C	A	E	S	SUMS OF HOW OFTEN COLUMNS			

FOR OFFICE USE ONLY How Often? C	A	E	S	Social Skills (cont.)	How Often? Never	Sometimes	Very Often
				15. I listen to adults when they are talking with me.	0	1	2
				16. I show that I like compliments or praise from friends.	0	1	2
				17. I listen to my friends when they talk about problems they are having.	0	1	2
				18. I avoid doing things with others that may get me in trouble with adults.	0	1	2
				19. I end fights with my parents calmly.	0	1	2
				20. I say nice things to others when they have done something well.	0	1	2
				21. I listen to the teacher when a lesson is being taught.	0	1	2
				22. I finish classroom work on time.	0	1	2
				23. I start talks with class members.	0	1	2
				24. I tell adults when they have done something for me that I like.	0	1	2
				25. I follow the teacher's directions.	0	1	2
				26. I try to understand how my friends feel when they are angry, upset, or sad.	0	1	2
				27. I ask friends for help with my problems.	0	1	2
				28. I ignore other children when they tease me or call me names.	0	1	2
				29. I accept people who are different.	0	1	2
				30. I use my free time in a good way.	0	1	2
				31. I ask classmates to join in an activity or game.	0	1	2
				32. I use a nice tone of voice in classroom discussions.	0	1	2
				33. I ask adults for help when other children try to hit me or push me around.	0	1	2
				34. I talk things over with classmates when there is a problem or an argument.	0	1	2
C	A	E	S	SUMS OF HOW OFTEN COLUMNS			

Stop. Please check to be sure that all items have been marked.

FOR OFFICE USE ONLY

SUMMARY

SOCIAL SKILLS

	HOW OFTEN? TOTAL (sums from p. 2)	+	(sums from p. 3)	=	BEHAVIOR LEVEL (see Appendix A) Fewer	Average	More
C		+		=			
A		+		=			
E		+		=			
S		+		=			
Total (C + A + E + S)							

(see Appendix D)

Standard Score ____ Percentile Rank ____

(see Appendix E)

SEM ± ____ Confidence Level 68% ☐ 95% ☐

Confidence Band (standard scores) ____ to ____

ID No. _____

Social Skills

Student Form
Secondary Level

Rating System

Grades 7-12

Social Skills Questionnaire

Frank M. Gresham and Stephen N. Elliott

Directions

First write the information about yourself in the box below. Then turn to page 2.

Student Information

Name _____
First Middle Last

☐ Male ☐ Female Today's date _____
Month Day Year

Grade _____ Age _____ Birth date _____
Month Day Year

School _____

Teacher's name _____

Form: SS

This paper lists a lot of things that students your age may do. Please read each sentence and think about yourself. Decide **how often** you do the behavior described.

If you **never** do this behavior, circle the **0**.
If you **sometimes** do this behavior, circle the **1**.
If you **very often** do this behavior, circle the **2**.

Then, decide how important the behavior is to your relationships with others.

If it is **not important** to your relationships, circle the **0**.
If it is **important** to your relationships, circle the **1**.
If it is **critical** to your relationships, circle the **2**.

Here are two examples:

	How Often?			How Important?		
	Never	Sometimes	Very Often	Not Important	Important	Critical
I start conversations with classmates.	0	1	(2)	0	(1)	2
I keep my desk clean and neat.	0	(1)	2	(0)	1	2

*This student **very often** starts conversations with classmates, and starting conversations with classmates is **important** to this student. This student **sometimes** keeps his or her desk clean and neat but a clean and neat desk is **not important** to this student.*

If you change an answer, be sure to erase completely. Please answer all questions. When you are finished, wait for further directions from your teacher. Be sure to ask questions if you do not know what to do. There are no right or wrong answers, just your feelings of how often you do these things and how important they are to you.

Begin working when told to do so.

FOR OFFICE USE ONLY How Often? C	A	E	S	Social Skills	How Often? Never	Sometimes	Very Often	How Important? Not Important	Important	Critical
				1. I make friends easily.	0	1	2	0	1	2
				2. I say nice things to others when they have done something well.	0	1	2	0	1	2
				3. I ask adults for help when other children try to hit me or push me around.	0	1	2	0	1	2
				4. I am confident on dates.	0	1	2	0	1	2
				5. I try to understand how my friends feel when they are angry, upset, or sad.	0	1	2	0	1	2
				6. I listen to adults when they are talking with me.	0	1	2	0	1	2
				7. I ignore other children when they tease me or call me names.	0	1	2	0	1	2
				8. I ask friends for help with my problems.	0	1	2	0	1	2
				9. I ask before using other people's things.	0	1	2	0	1	2
				10. I disagree with adults without fighting or arguing.	0	1	2	0	1	2
				11. I avoid doing things with others that may get me in trouble with adults.	0	1	2	0	1	2
				12. I feel sorry for others when bad things happen to them.	0	1	2	0	1	2
C	A	E	S	SUMS OF HOW OFTEN COLUMNS						

FOR OFFICE USE ONLY How Often? C	A	E	S	Social Skills (cont.)	How Often? Never	Sometimes	Very Often	How Important? Not Important	Important	Critical
				13. I do my homework on time.	0	1	2	0	1	2
				14. I keep my desk clean and neat.	0	1	2	0	1	2
				15. I do nice things for my parents like helping with household chores without being asked.	0	1	2	0	1	2
				16. I am active in school activities such as sports or clubs.	0	1	2	0	1	2
				17. I finish classroom work on time.	0	1	2	0	1	2
				18. I compromise with parents or teachers when we have disagreements.	0	1	2	0	1	2
				19. I ignore classmates who are clowning around in class.	0	1	2	0	1	2
				20. I ask someone I like for a date.	0	1	2	0	1	2
				21. I listen to my friends when they talk about problems they are having.	0	1	2	0	1	2
				22. I end fights with my parents calmly.	0	1	2	0	1	2
				23. I give compliments to members of the opposite sex.	0	1	2	0	1	2
				24. I tell other people when they have done something well.	0	1	2	0	1	2
				25. I smile, wave, or nod at others.	0	1	2	0	1	2
				26. I start conversations with opposite-sex friends without feeling uneasy or nervous.	0	1	2	0	1	2
				27. I accept punishment from adults without getting mad.	0	1	2	0	1	2
				28. I let friends know I like them by telling or showing them.	0	1	2	0	1	2
				29. I stand up for my friends when they have been unfairly criticized.	0	1	2	0	1	2
				30. I invite others to join in social activities.	0	1	2	0	1	2
				31. I use my free time in a good way.	0	1	2	0	1	2
				32. I control my temper when people are angry with me.	0	1	2	0	1	2
				33. I get the attention of members of the opposite sex without feeling embarrassed.	0	1	2	0	1	2
				34. I take criticism from my parents without getting angry.	0	1	2	0	1	2
				35. I follow the teacher's directions.	0	1	2	0	1	2
				36. I use a nice tone of voice in classroom discussions.	0	1	2	0	1	2
				37. I ask friends to do favors for me.	0	1	2	0	1	2
				38. I start talks with classroom members.	0	1	2	0	1	2
				39. I talk things over with classmates when there is a problem or an argument.	0	1	2	0	1	2
C	A	E	S	SUMS OF HOW OFTEN COLUMNS						

Stop. Please check to be sure all items have been marked.

FOR OFFICE USE ONLY

SUMMARY

SOCIAL SKILLS

	HOW OFTEN? TOTAL (sums from p. 2) + (sums from p. 3) =	BEHAVIOR LEVEL (see Appendix A) Fewer	Average	More
C	+ =			
A	+ =			
E	+ =			
S	+ =			
Total (C + A + E + S)				

(see Appendix D)

Standard Score ____ Percentile Rank ____

(see Appendix E)

SEM ± ____ Confidence Level 68% ☐ 95% ☐

Confidence Band (standard scores) ____ to ____

Appendix E

Transcript of sibling responses to the open-ended questions

Question 1:

S001- Age 11 years	We go to the hospital every day
S002- Age 7 years	I go with my nan or aunty – it's fun
S003-Age 14 years	I stay at home and mum and dad shared staying at home and hospital
S004-Age 15 years	Last admission was a number of years ago when I was in primary school. I did like him being in hospital and I enjoyed the change in routine, just being at home with dad and my sister (mum stayed at the hospital)
S005-Age 15 years	Stayed home with grandparents or dad or go with mum to Sale while my sister was in hospital
S006-Age 10 years	1 admission last year. When I woke up in the morning my brother wasn't there – he'd gone into hospital during the night and I skipped school and went with dad to the hospital to be with my brother
S007-Age 10 years	N/A
S008-Age 10 years	N/A
S009-Age 15 years	Mum stayed with my brother. My sister and I stayed at home with dad. When my brother was in surgery (bowel obstruction) I played with friends to take my mind off things
S010-Age 8 years	Mum goes to the hospital and dad is at work so nanna looks after me
S011-Age 14 years	When I was little I would get carted around a lot to different friends. Now I'm older I'll stay at home till dad gets home
S012-Age 14 years	N/A
S013-Age 12 years	I feel a bit left out (not that I'm complaining) I go off to grandmas – I'm shipped off to peoples houses. I don't get to see him for long only when mum and dad change over and I usually see him for longer
S014-Age 15 years	My sibling has HITH and comes home usually on day 2 of a tune-up. I stay at home with my dad and brother. If she stays in we do a lot of travelling to Melbourne. I come down on weekends and sometimes after school
S015-Age 15 years	I stayed with my grandma and dad was there too while my sister was in hospital for a few weeks after she was burnt in a kerosene heater fire. I visited regularly on weekends
S016-Age 13 years	N/A
S017-Age 11 years	It was when I was in grade 2. I went to a friend's house to stay and I visited them in hospital. I remember getting McDonalds and I chipped my tooth and mum was worried about it. Mum stayed at the hospital and dad was working
S018-Age 10 years	N/A
S019-Age 10 years	Mum's boyfriend looks after us.
S020-Age16 years	Last admission was 10 years ago – I can't remember much – visiting a few times only
S021-Age 12 years	Dad stays at home to care for the family and mum stays in hospital with my brother. We visit lots of times
S022-Age 11 years	I can't remember a time when he was in hospital
S023-Age 12 years	I went to school and dad looked after me. I'd go and visit my sibling.

Question 1: (continued)

S024-Age 15 years	Mum goes into the hospital to stay and we visit on weekends. Dad stays at home with us.
S025-Age 9 years	N/A
S026-Age 13 years	Stayed with great grandmother - mum came to stay at RCH with sibling
S027-Age 14 years	Mum and dad both stay at home as sibling is a teenager. When I go to the hospital to visit I muck up and get into trouble because I talk to everyone else except my brother
S028-Age 11 years	Stay at grandparents with mum and dad stays with my sister
S029-Age 10 years	I usually look after the baby when mum is busy with my sister. If she goes in RCH I stay with dad or nanna unless it's in the holidays
S030-Age 15 years	N/A – sibling only in hospital at time of diagnosis and I can't remember it
S031-Age 9 years	No – N/A
S032-Age 13 years	N/A
S033-Age 16 years	Never had an admission
S034-Age 13 years	Last time I went to Melbourne with the family and we all stayed together – my brother was having investigations as an O/P – can't remember him having to stay in hospital
S035-Age 11 years	N/A
S036-Age 11 years	I stayed at my nan's house and had to catch the bus to school because they live in Mathoura. Dad had to work and mum stayed in Melbourne with my sister
S037-Age 14 years	He goes into Geelong Hospital. When he was younger mum or dad would stay at the hospital but recently they've stayed at home
S038-Age 14 years	Mum sometimes stays in at the hospital. Dad is at home and I go about my own things
S039-Age 12 years	I stay home with the rest of the family and I go to school. Mum usually stays at the hospital

Question 2:

S001- Age 11 years	- She can have lollies and I can too - I get to ride my bike more because she needs exercise - We play poison ball on the trampoline to give my sister exercise and it's fun
S002- Age 7 years	We get to jump on the trampoline and walk to school because it's good for him
S003-Age 14 years	N/A
S004-Age 15 years	We moved to a house with a bigger block as my parents thought it would be good for my brother and also good for us
S005-Age 15 years	We went to America with Make a Wish
S006-Age 10 years	- I get to play lots of games like soccer with him as it's good for him to nun around - I like helping with his physio – patting and help him count with his blowing - When I was younger I enjoyed the activities in clinic with the play specialist as I'd get out of school earlier to go to my brother's appointments.
S007-Age 10 years	She takes me to clinic with her and when I go there it's fun
S008-Age 10 years	You get to take days off school to come into clinic and play on the Nintendo
S009-Age 15 years	- Having junk food around the house - I know all about the GI system because we talk at home and it helps with my schooling - It stops middle child syndrome and gives him 'his special thing'
S010-Age 8 years	That I can always take care of her every day and help her like doing her jobs when she is sick
S011-Age 14 years	- Meeting all the hospital staff - You learn a lot more and help others understand about CF - Getting involved in charities and helping them e.g. Starling Foundation, TLC for Kids, CF Research Trust
S012-Age 14 years	- Get to come to Melbourne and go shopping and to McDonalds - Getting to see the rural area around Melbourne - Missing school and having a day out
S013-Age 12 years	- Not that I want him to go into hospital but when he does it's so good to see him and we have lots of laughs - It has taught me to think about what I say around him and others as sometimes I get angry – this a good thing - I've met lots of new people at the hospital e.g. VSK sibling days
S014-Age 15 years	It makes us closer as siblings as I have a different understanding about things because of CF

Question 2: (continued)

S015-Age 15 years	- When I was in primary school I'd come to Melbourne for her checkups
	- Understanding the disease – I did CF for Biology (year 10) genetics
	- We get spoilt with fattening treats more often.
S016-Age 13 years	- I usually come to Melbourne for clinic visits
	- I get to go to McDonalds sometimes because my brother needs to eat it
	- Because he's well; he is a normal kid
S017-Age 11 years	- I got a Creon milkshake maker and it has been very handy
	- Access to experiences we wouldn't have otherwise had e.g. football matches and Made A Wish
	- I go to McDonalds when I come into hospital
S018-Age 10 years	- We get free movie tickets
	- We get to sell red rose ribbons at school and to friends
	- We get things like the milk shake maker and expandable face washer
S019-Age 10 years	- The whole family are involved in his wish with Starlight or Make A Wish
	- We went to a fun park because my brother wanted to go there (Ballan Park)
S020-Age16 years	- I wouldn't be as good friends with my best friend (who has CF) because of having CF in common
	- I get an afternoon to myself at home on clinic days
S021-Age 12 years	- We get to know the hospital a lot more and get to know the doctors I see when I'm sick
S022-Age 11 years	- Good to hold on to when they are alive
	- It's brought us closer together
S023-Age 12 years	He can't be bothered fighting me back. He's not stupid because he feels sick. When he's sick he's quiet
S024-Age 15 years	- Get to know more people like staff and doctors at the hospital
	- Get to spend time as a family in Melbourne for clinic visits and hospitalisation
	- Helps with school projects as you can do CF – a lot of info you can learn e.g. for science
S025-Age 9 years	- Sometimes I get to go to the hospital and have McDonalds or go shopping after
	- I get to share the trampoline with my brother
S026-Age 13 years	Get to come to Melbourne and go shopping after clinic
S027-Age 14 years	I get more attention when he's in hospital

Question 2: (continued)

S028-Age 11 years	- I get to see friends I've met at VSK sibling days
	- I get to go to the Starlight Room
	- I like seeing the helicopters land in the park at the back of RCH
S029-Age 10 years	- I get extra junk food
	- I get to meet a lot of new people
	- I get time to do things on my own when she's doing nebs and her treatments
S030-Age 15 years	- I get experience coming to the hospital and seeing/meeting people who have chronic illness
	- She is determined to be classed as "normal" and not to be left out
S031-Age 9 years	- I'm happy I don't have it
	- I don't want him to die from it
	- My brother is happy about coming to the hospital
S032-Age 13 years	- Travelling to Melbourne and Adelaide a lot for clinic (Adelaide when family lived in Darwin)
S033-Age 16 years	- It made it easy doing my health project on CF
	- Can't think of any other things
S034-Age 13 years	You get to learn a bit more about life
S035-Age 11 years	I enjoy going to McDonalds and playing Nintendo
S036-Age 11 years	We have McDonalds for lunch when my sister comes to clinic (usually I come too). I enjoy the trips to Melbourne and seeing the city
S037-Age 14 years	Get to go to some good places with Make A Wish. We got to go to Hamilton Island and the Davis Cup
S038-Age 14 years	- You give her a bit more respect
	- She looks up to you and asks you questions about things
	- She likes to play a lot of sport with me which I like and it helps her
S039-Age 12 years	No idea

Question 3:

S001- Age 11 years	- She has to do physio every day and I can't take my friends into the room while she is doing it as she doesn't like anyone seeing her do it
	- Can't think of anything else
S002- Age 7 years	- He always vomits when he is sick
	- We have to stop in the car if we are going somewhere and he feels unwell
	- If he forgets his enzymes it's disgusting
S003-Age 14 years	She's always sick and coughs at night and keeps me awake and I am concerned if it sounds bad that she's ok.
S004-Age 15 years	- He has a lot of time with mum and dad and is spoilt (also compounded by being youngest)
	- He does not appreciate things
	- We don't know if he's going to get really sick and have a shorter life
	- His treatment probably stops us from doing things as a family and it comes before enjoyable things
S005-Age 15 years	- Having to remind her to take her tablets
	- Waking up in the middle of the night because she's coughing and you can't sleep. I go and check sometimes that she's ok
S006-Age 10 years	- When he goes into hospital – worried he'll be ok
	- He doesn't like taking the medicine and has tantrums and it's hard for him – I don't have to do the treatments so it's not the same for me
	- He has to have tests at clinic sometimes and I don't know how that feels – it's hard for him and it's hard for me as he has it and I don't
S007-Age 10 years	Can't think of anything
S008-Age 10 years	Cant' think of anything
S009-Age 15 years	- When you go out and he forgets his enzymes it's annoying
	- When he's in hospital and I don't talk to my friends as he's coming to the high school next year so I don't want everyone to know about it – he can tell
	- The nebuliser makes a lot of noise if you are watching TV
S010-Age 8 years	- Loud coughing and I get headaches
	- We are sometimes late for school because she sleeps in because she was coughing at night
S011-Age 14 years	- Being transported around when she's in hospital
	- People ask more about my sibling than me
	- When I come to visit her sometimes I bear the brunt of feeling bad/having a bad day and staff emphasise her – What about me?

Question 3: (continued)

S012-Age 14 years	- Having to pick up his tablets lying around
	- He's lighter than other kids his age and he doesn't eat as much as he should
	- Has to come for checkups all the time. I shouldn't be able to pick him up
S013-Age 12 years	- I have to watch what I say around him sometimes when I'm angry with him, I'd like to say I wish you'd die but that would be awful
	- There can be arguments about treatment that we wouldn't have if he didn't have CF
	- It's hard when you tell someone you have a brother with CF and they are being sympathetic and you are trying to normalise it – it's a reminder to me about how awful it is to have CF, his button (PEG) and port are also reminders that he's not well.
S014-Age 15 years	- Having her staying in hospital and not being at home - I miss her not being there!
	- We aren't able to enjoy the same activities that other siblings can e.g. going swimming and running, because of CF
S015-Age 15 years	- Sometimes she refuses to have her physio or neb and that's frustrating
	- She's not into sport much – my sib dropped out of martial arts as it was too much for her but it would have been good to practice together and have her company there
	- She's fussy with food and that's difficult
S016-Age 13 years	- Sometimes I miss out on Melbourne clinics
	- Having chocolate and fatty foods in the house which I can't have
	- He's a bit spoilt because of his CF but that doesn't worry me at all
S017-Age 11 years	- She screams at the GP when she needs a flu injection as she hates needles; it's embarrassing
	- She's favoured by my grandma e.g. she doesn't get into trouble as much as if I did the same things
	- She wakes up in the night and comes to get me or cries out for mum as she's afraid of the dark, especially when she was little
S018-Age 10 years	- To remind him to have his tablets at school
	- Sometimes I have to go to a friend's house if they aren't back from Melbourne clinic – I might get home to get a message on the table
S019-Age 10 years	- He has to stop to rest when he is riding his bike to school, so I have to wait a long time
	- He is always coughing at night when I'm trying to sleep
	- He has to cough a lot when we play tiggy and it stops the game
S020-Age16 years	- Clinic visits impact on my ability to be picked up/dropped off
	- Sibling is mild so doesn't have a huge impact

Question 3: (continued)

S021-Age 12 years	- I don't get to see my brother and mum as much when my brother is in hospital
	- It makes it hard to do homework
	- I can't do after school activities as mum can't pick me up and dad is at work
S022-Age 11 years	- He might die at a young age
	- If he goes I'm only going to have a stepbrother not a (real) brother
	- I won't have anyone to look out for at school and look after if he goes
S023-Age 12 years	- I get bored if he's sick and can't muck around with me
	- His physio can interrupt us playing
S024-Age 15 years	- We can't go to the pool sometimes because it's too cold for my sister and I'd like to take her
	- We can't go to the snow either because of the cold and it's effect on her
	- Hassle of travelling to Melbourne sometimes is a negative
S025-Age 9 years	- Sometimes people stare when he takes his medicine
	- Have to be careful not to pass on colds to him
S026-Age 13 years	- Having admissions and checkups at the Base Hospital all the time. We have to stay there late and it gets boring
S027-Age 14 years	- You can't tease him when he's doing his treatments
	- He gets away with everything and is treated differently by our parents when he's sick
S028-Age 11 years	- Sometimes I'm late to school because dad is doing my sister's physio
	- We have to go into hospital and it is a long trip
	- Have to see other sick kids in the hospital and I feel a bit sad
S029-Age 10 years	- We have to travel a lot
	- A lot of money we have is spent on my sister's medicines
	- My sister needing to have operations because of CF (PEG and port)
S030-Age 15 years	- When she doesn't want to have her medication she gets cross at me if I tell her to do it
	- When she's sick she groans in the night and I her hear (her bedroom is next door)
	- She leaves tissues she has coughed into everywhere and the new puppy eats them
S031-Age 9 years	- Having enzymes and a pump
	- Getting sick
	- Having to go to the RCH

Question 3: (continued)

S032-Age 13 years	- Having him get fed up taking his medications and then the arguments with mum and dad
	- When he gets unwell he gets really sick
S033-Age 16 years	- Its annoying when he plays his saxophone which he plays to keep fit
	- It s annoying when he forgets to take his enzymes when we go out – he usually takes his food home
S034-Age 13 years	- The money it costs to buy pumps and equipment
	- I feel miserable for my brother because its hard for him and not fair he has a disease
	- If he is sick you have to stop everything to care for him – it can interrupt family plans/activities
S035-Age 11 years	No, just used to it – I don't have any problems
S036-Age 11 years	My sister has to have physio, pumps and tablets
S037-Age 14 years	He has to have medication all the time and go into hospital
S038-Age 14 years	- You've got to be careful with her when you are mucking around if she starts coughing
	- When she goes into hospital, not seeing her
	- The attention she gets (but she needs it) from mum and dad
S039-Age 12 years	Nothing

Question 4:

S001- Age 11 years	She could do her physio in the room at the back of the house so I could bring my friend's inside
S002- Age 7 years	If when he swallows his food, it didn't get stuck
S003-Age 14 years	Finding a cure for CF
S004-Age 15 years	Nothing I can think of
S005-Age 15 years	Sometimes it helps to talk to other siblings
S006-Age 10 years	If he didn't have to do the physio and medications it would be easier i.e. if we forget the enzymes he can't have food if we are out until we get home so I wait to eat too to make it easier for him
S007-Age 10 years	No
S008-Age 10 years	Make her run slower as she's a faster runner than me! She's in the top 3 at school and I'm in the top 7
S009-Age 15 years	When I was in grade 4, he was sick and that was hard – felt left out. It would be nice to know other CF siblings as they understand. When I was younger it would have helped to speak to people who understood. My sister was a baby then – she helped – mum and dad were worried about my brother but the baby didn't change – constant there – I was that to her this last admission as I was older. You're not jealous because you don't want CF
S010-Age 8 years	If she had a room further away from me as the comes into my room when I'm asleep! She sometimes com to my room to play in the night
S011-Age 14 years	Doing studies like this for siblings. Camps like VSK for sibs to escape the pumps and coughing – get to talk to others who know how you feel. Keep in touch with sibs via MSN
S012-Age 14 years	If we lived closer to Melbourne or there was a CF Centre closer to Shepparton
S013-Age 12 years	A cure would be great obviously. Something for teenage sibs – regular monthly meeting and go to movies, etc.
S014-Age 15 years	Being able to talk to someone else who is the sibling of a person with CF
S015-Age 15 years	- Provide more information about the disease - Activities room for siblings or programme - games or movie room - It would be good to talk to other siblings to know what their experiences are
S016-Age 13 years	To have something like the Starlight Room for siblings as lots of things are for patients
S017-Age 11 years	If she wasn't favoured, scared of needles and didn't have to go to hospital
S018-Age 10 years	If I didn't have to share toys with him – but I'd have to anyway even if he didn't have CF
S019-Age 10 years	Can't think of anything
S020-Age16 years	Very little effect on sibling as brother has very mild CF
S021-Age 12 years	Talking to other siblings may help

Question 4: (continued)

S022-Age 11 years	- If he didn't have CF I wouldn't have to worry so much - If he lived as long as me - If there was someone to talk to at the hospital.
S023-Age 12 years	Nothing
S024-Age 15 years	To have a meeting with a group of siblings to see their perspective of what it's like
S025-Age 9 years	Have a cure
S026-Age 13 years	Can't think of anything
S027-Age 14 years	Provide something to do when siblings are visiting RCH to meet other siblings
S028-Age 11 years	If there was a cure for CF
S029-Age 10 years	- If medicine was cheaper and we didn't have to travel as much - The hospital could have a games room for brothers and sisters so they don't get sick when they are in the hospital
S030-Age 15 years	- Sibling social groups would be good – good way to meeting new people
S031-Age 9 years	If my brother wasn't sick and we could go underwater without him needing earplugs and if he didn't have CF
S032-Age 13 years	If he understood how bad it was maybe he'd take his medicines
S033-Age 16 years	It doesn't affect me as much as he is like every other little brother (annoying!)
S034-Age 13 years	- If they could find a cure - Groups for siblings run by the hospital
S035-Age 11 years	Can't think of anything
S036-Age 11 years	Can't think of anything as not a negative impact
S037-Age 14 years	If he didn't have to take enzymes and we didn't need to remind him all the time
S038-Age 14 years	Finding out some more about CF and what it's like to have it
S039-Age 12 years	No effect, so nothing we could be doing