COPING AND QUALITY OF LIFE FOR SIBLINGS OF CHILDREN WITH CHRONIC ILLNESS

A dissertation submitted
to Kent State University in partial fulfillment of the requirements for the degree of Doctor of Philosophy

by
Elissa Lampe Deggelman

August, 2011
Dissertation written by
Elissa Lampe Deggelman
B.A., University of Notre Dame, 2005
M.A., Kent State University, 2008
Ph.D., Kent State University, 2011

Approved by

__________________________, Chair, Doctoral Dissertation Committee
Beth G. Wildman, PhD

__________________________, Members, Doctoral Dissertation Committee
Janis Crowther, PhD

William Merriman, PhD

Susan Roxburgh, PhD

Sara Newman, PhD

Accepted by

__________________________, Chair, Department of Psychology
Janis Crowther, PhD

__________________________, Dean, College of Arts and Sciences
John R. D. Stalvey, PhD
# TABLE OF CONTENTS

LIST OF TABLES .................................................................................................................. iv

INTRODUCTION ...................................................................................................................... 1

METHODS ............................................................................................................................... 24
  PARTICIPANTS ..................................................................................................................... 24
  MEASURES .......................................................................................................................... 27
  PROCEDURES ....................................................................................................................... 35

RESULTS ................................................................................................................................. 43

DISCUSSION ............................................................................................................................ 55

REFERENCES ......................................................................................................................... 65

APPENDICES

A  DEMOGRAPHIC QUESTIONNAIRE ................................................................................... 80
B  KIDCOPE ............................................................................................................................ 82
C  PEDSQL PARENT REPORT ............................................................................................... 84
D  PEDSQL CHILD REPORT .................................................................................................. 85
# LIST OF TABLES

<table>
<thead>
<tr>
<th>Table</th>
<th>Description</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Descriptive statistics for participants included and excluded from analysis.</td>
<td>25</td>
</tr>
<tr>
<td>2</td>
<td>The coping strategies and corresponding item numbers for the coping styles assessed by the Kidcope.</td>
<td>31</td>
</tr>
<tr>
<td>3</td>
<td>Correlational table among all study variables.</td>
<td>38</td>
</tr>
<tr>
<td>4</td>
<td>Results of post-hoc power analyses and achieved effect size for all analyses.</td>
<td>44</td>
</tr>
<tr>
<td>5</td>
<td>Results of regression using demographic and illness parameters including and excluding time since diagnosis to predict well-sibling self-report adjustment.</td>
<td>45</td>
</tr>
<tr>
<td>6</td>
<td>Results of regressions using well-sibling age and approach and avoidant coping styles, total coping strategies, and coping ratio of avoidance to total number of coping strategies to predict well-sibling self-report adjustment.</td>
<td>47</td>
</tr>
<tr>
<td>7</td>
<td>Results of regressions using well-sibling age and number of avoidant or approach coping strategies to predict well-sibling self-reported adjustment.</td>
<td>48</td>
</tr>
<tr>
<td>8</td>
<td>Results of regressions using well-sibling age and avoidant or approach coping ratio to predict well-sibling self-reported adjustment.</td>
<td>50</td>
</tr>
<tr>
<td>9</td>
<td>Percentages of coping strategies endorsed by well-siblings.</td>
<td>51</td>
</tr>
<tr>
<td>10</td>
<td>Results of regression using well-sibling report distress to predict well-sibling self-reported adjustment after controlling for well-sibling age.</td>
<td>53</td>
</tr>
</tbody>
</table>
INTRODUCTION

An estimated 30% of children have a long-term, chronic illness including diabetes, cancer, asthma, and cystic fibrosis (McDougall, King, de Wit, Miller, Hong, Offord, LaPorta, & Meyer, 2004). Chronic illness is a disease with “an expected duration of at least three months or the impossibility of a cure” (van der Lee, Mokkink, Grootenhuis, Heymans, & Offringa, 2007, pg. 2749). Physical symptoms and treatment related to the chronic illness interfere with the life of the affected child (hereafter referred to as the sick-child), as well as the lives of his or her parents and siblings (Barlow & Ellard, 2006). Family systems and social ecology theory state that each family member is critical in maintaining balance and equilibrium within the family system, and thus emphasize the importance of the interaction among family members (Kazak, 1992a; Kazak, 1992b). These theories posit that when one member of the family experiences a disruption or change, all family members are influenced by this change. Despite the influence of the illness on all members of the family, most research that studies the impact of a child’s chronic illness on the “family” examines only maternal adjustment and neglects the impact on other family members, including siblings (Wood, 2005).

Though most research on the family impact of chronic illness does not include siblings, there is some research examining the psychosocial adjustment of a child whose sibling has a chronic illness (hereafter referred to as well-sibling). Examination of the available literature revealed a small but significant negative impact on psychosocial
outcomes of well-siblings, such that well-siblings of children with chronic illnesses have more adjustment problems than siblings of healthy children (Sharpe & Rossiter, 2002). In many studies, well-sibling adjustment is assessed through parental reports of well-sibling psychopathology symptoms (e.g. Labay & Walco, 2004; Laufersweiler-Plass, Sudnik-Schöneborn, Zerres, Backes, Lehmkuhl, & von Gontard, 2003). Though well-siblings appear to be at an increased risk of emotional problems compared to healthy controls, most well-siblings’ psychosocial problems are within normal range and are not clinically significant so as to warrant classification as psychopathology (e.g., Sharp & Rossiter, 2002; Taylor et al., 2001; Wood, Sherman, Hamiwka, Blackman, & Wirrell, 2008).

Many studies examining well-sibling adjustment to chronic illness use instruments that measure only internalizing and externalizing psychosocial problems (e.g., Alderfer, Labay, & Kazak, 2004; Batte, Watson, & Amess, 2006; Laufersweiler-Plass et al., 2003). Given that most well-siblings’ adjustment levels are within normal range and are not significant enough to warrant classification as psychopathology, using assessment measures that assess for clinical levels of psychosocial problems may not adequately capture well-sibling adjustment (e.g., Gold, Mahrer, Treadwell, Weissman, & Vichinsky, 2008; Labay & Walco, 2004; Taylor et al., 2001). Additionally, overlap between coping strategies and symptoms of psychopathology are not uncommon (Compas, Connor-Smith, Saltzman, Thomsen, & Wadsworth, 2001). For example, crying is both a symptom of depression (American Psychiatric Association [DSM-IV-TR],
2000) and a coping method. In examining well-sibling adjustment, it is important that the outcome measure is not confounded with coping styles.

An assessment of global adjustment of the well-sibling, or quality of life, measures more domains of functioning than only psychopathology and avoids confounding coping and symptoms of distress. Quality of life is defined as “well-being in multiple domains of life considered salient in one’s culture and time” (Wallander, Schmitt, & Koot, 2001; pg. 574). Quality of life assessments measure multiple domains in which the child interacts, such as physical, academic, social, and emotional functioning. Traditional measurements of well-sibling adjustment that assess psychosocial problems neglect information on some of these areas of well-being, such as physical functioning, social functioning, and emotional functioning. Some research has found that well-siblings have impairments in areas assessed in quality of life, including somatic complaints (Buchbinder, Casillas, & Zeltzer, 2010; Williams, Ridder, Setter, Liebergen, Curry, Piamjariyakul, & Williams, 2009), social problems (Houtzager, Grootenhuis, Caron, & Last, 2004), school problems (Barlow & Ellard, 2006; Read, Kinali, Muntoni, & Garralda, 2010) and emotional reactions (Gold et al., 2008). A quality of life assessment offers a greater understanding of the global impact on the adjustment of the well-sibling rather than measuring only psychopathology symptoms.

Though a meta-analysis of well-sibling adjustment found a negative overall effect (Sharpe & Rossiter, 2002), some research on well-sibling adjustment has yielded mixed results. Specifically, the presence of a chronic illness in a brother or sister was associated
with positive psychosocial outcomes (e.g., MacLeod, Whitsett, Mash, & Pelletier, 2003; Waite-Jones & Madill, 2007) or no significant psychosocial outcomes (e.g., Labay & Walco, 2004). Given the discrepancies in the literature about the positive, negative, or neutral impact of chronic illness on well-sibling adjustment, it is important to examine some possible explanations for these different results. Many factors have been studied in relation to well-sibling adjustment, including illness parameters (such as type of illness, severity of illness, and time since diagnosis), demographic parameters (such as age and gender) and well-sibling process variables (such as coping). It is likely that some of the discrepancies reported in the impact of chronic illness on well-sibling adjustment may be related to the factors each study examined. The next section will investigate the relationship between these factors and well-sibling’s adjustment.

Ilness Parameters

Some studies that examined well-sibling adjustment to chronic illness included a sample that combined several types of chronic illnesses (e.g. Gallo & Szchlnski, 2003; Taylor et al., 2001), while other studies used only a single type of chronic illness in the sample (e.g., Waite-Jones & Madill, 2007; Weiss, Schiaffino, Ilowite, 2001). There is mixed evidence as to whether type of illness differentially impacts the well-sibling’s adjustment. Use of a sample that combines different chronic illnesses is referred to as the non-categorical approach (Stein & Jessop, 1982). This approach acknowledges that each illness creates unique demands but emphasizes that the issues common to all pediatric chronic illnesses, such as disruption of family life and increased medical attention, are more important than specific illness factors. Given the importance of
these common issues, multiple chronic illnesses can be examined together. The
categorical, or disease-specific, approach claims that the specific type of illness
differentially impacts the adjustment of well-siblings (Stein & Jessop, 1982). Many
studies support the categorical approach (e.g., Hames & Appleton, 2009; Gold, Mahrer,
Treadwell, Weissman, & Vinchinsky, 2008; Read et al., 2010; Waite-Jones & Madill,
2007). Additionally, some previous research found well-sibling adjustment to chronic
illness is impacted by specific illness factors. These factors, including time since
diagnosis and illness severity, vary by specific disease types and further support the
categorical approach.

Severity of illness is a specific illness factor related to well-sibling adjustment in
some studies (Barlow & Ellard, 2006; Sharpe & Rossiter, 2002). One study that
examined well-sibling adjustment to having a sibling with muscular dystrophy found a
significant relationship between psychological symptoms of well-siblings and higher
extent of wheelchair use for the sick-child (Read et al., 2010). Wheelchair use is a proxy
for illness severity for sick-children with muscular dystrophy; therefore these results
suggest that more severe illnesses have a greater negative impact on well-siblings. Other
research suggested more intensive treatments for sick-children predict poorer outcomes
for well-siblings (Barlow & Ellard, 2006). Some illnesses (e.g. cancer or cystic fibrosis)
require more complicated and intrusive treatment, such as rigid dietary restrictions or
taking many medications multiple times daily, while other illnesses (e.g., asthma) require
less intensive daily intervention, such as taking only one medication daily. Some data
suggest that well-siblings of sick-children with illnesses that require intensive daily
treatment regimes that restrict the well-siblings’ daily activities, such as school and recreational time, have poorer adjustment (Barlow & Ellard, 2006). Other studies have not found a relationship between illness severity and well-sibling adjustment to chronic illness (e.g., Bellin, Bentley, & Sawin, 2009; Laufersweiler-Plass et al., 2003). One meta-analysis concluded that well-siblings of children with chronic illnesses with higher mortality rates (including HIV/AIDS, cancer, cystic fibrosis, and sickle cell anemia) were no more at risk for poor psychosocial adjustment than well-siblings of children with less severe chronic illnesses with lower mortality rates (including diabetes, rheumatic disease, bowel disease, hearing impairments, and asthma) (Sharpe & Rossiter, 2002). Given the conflicting evidence, research should further examine the impact of severity of illness on well-siblings’ quality of life.

Similar to severity of illness, there is mixed evidence to suggest that the amount of time elapsed since diagnosis is another specific illness factor that impacts well-sibling adjustment (Alderfer, Long, Lown, Marsland, Ostrowski, Hock, & Ewing, 2010). Well-siblings of pediatric cancer patients exhibited the greatest impairments in quality of life in the first month following the diagnosis; negative effects of the diagnosis were significantly reduced six months following the diagnosis (Houtzager et al., 2004). These data suggest that well-siblings may have more opportunities to adjust to the disruption of diagnosis as more time elapses. Time since diagnosis is not consistently related to well-sibling adjustment (Labay & Walco, 2004), and most studies that explored the impact of time since diagnosis on sibling adjustment examined this relationship for well-siblings of pediatric oncology patients. The categorical approach suggests the influence of time since
diagnosis may be different for illnesses other than cancer, such as epilepsy and diabetes. Time since diagnosis was not a significant predictor of well-sibling adjustment to type 1 diabetes mellitus (Jackson, Richer, & Edge, 2008). Well-siblings of sick-children with diabetes and epilepsy have normal adjustment after several years have passed since diagnosis (Hames & Appleton, 2009; Wennick, Lundqvist, Hallström, 2009). Thus, given the disparity about the impact of time since diagnosis according to type of illness, there is a need for further investigation of the impact of time since diagnosis on well-sibling adjustment to chronic illness other than cancer.

The differential impact of type of treatment, time since diagnosis, and severity of illness further supports the categorical approach to understanding the impact of chronic illness on siblings (e.g., Houtzager et al., 2004; Jackson, Richer, & Edge, 2008; Weiss et al., 2006). Working within the categorical approach, the current paper will examine well-sibling adjustment to a specific chronic illness: cystic fibrosis. Cystic fibrosis was selected for the current study because it is a serious, progressive chronic illness that affects the daily lives of the ill children and their family members (Carpenter & Narsavage, 2004). Cystic fibrosis is a genetic disease that causes the body to produce thick and sticky mucus that builds up in the lungs and the pancreas; this collection of mucus causes life-threatening lung infections, and poor growth and weight gain due to difficulty breaking down and absorbing food. The role of family is particularly important in managing cystic fibrosis as there are many at-home requirements for the treatment of the sick-child. Specifically, the sick-child must adhere to strict dietary requirements and follow routines that the family must perform two or three times daily, including
inhalation of medication and chest physiotherapy for the treatment of pulmonary symptoms. Given the conflicting results in the literature, the present study will examine the impact of severity of illness and time since diagnosis in predicting quality of life of well-siblings of children with cystic fibrosis.

**Demographic Parameters**

**Well-Sibling Gender.** In addition to cystic fibrosis illness parameters, individual well-sibling factors are important when assessing adjustment to chronic illness (e.g., Alderfer, Labay, & Kazak, 2003; Houtzager et al., 2004; Waite-Jones & Madill, 2007). Brothers and sisters may adjust to the presence of a chronic illness in different ways, therefore well-sibling gender should be considered when predicting well-sibling adjustment to chronic illness (Houtzager et al., 2004; Silver & Frohlinger-Graham, 2000). Well-siblings appear to be at risk for different types of psychopathology; boys are more at risk for externalizing problems such as aggression and hyperactivity, and girls are more at risk for internalizing problems such as anxiety, depression, and loneliness (e.g., Houtzager et al., 2004; Thompson et al., 1994). These gender differences may reflect developmental differences in psychopathology rather than gender differences in adjusting to chronic illness, as boys are generally more at-risk for externalizing symptoms and girls more at-risk for internalizing symptoms (e.g., Ramtekkar, Reiersen, Todorov, & Todd, 2010). Though some studies found that gender influences the way in which well-siblings adjust to chronic illnesses, most of these studies examine psychopathology symptoms of well-siblings and neglect other domains, such as social functioning. Some studies have not found gender differences in well-sibling adjustment in areas other than...
psychopathology such as empathy, emotional symptoms, and peer problems (e.g., Barlow & Ellard, 2006; Labay & Walco, 2004; Taylor et al., 2002). There is a need, therefore, for research to examine the relationship between gender and the quality of life of well-siblings of children with chronic illnesses. Unlike psychopathology, there are no significant gender differences in quality of life (Varni et al., 2003). Therefore, gender differences in quality of life are likely to reflect a response to chronic illness.

Internalizing symptoms of well-sibling females may be attributed to increased domestic responsibilities (Sharpe & Rossiter, 2002). Well-siblings report increased domestic responsibilities, such as housekeeping and caretaking of the sick-child (Hames & Appleton, 2009; Read et al, 2010). Parents also reported that well-siblings participate in more caretaking responsibilities with a sick-child (Lehna, 2001; Williams et al., 2009). Compared to boys, well-sibling girls often have more responsibilities within the family and are more involved in the treatment of the illness (Waite-Jones & Madill, 2007). Female well-siblings appear to assume a more parental role to aid in the support of the sick child, or undertake other domestic roles that the parent vacates to care for the sick child. Taking on parental chores and responsibilities that are developmentally or culturally inappropriate for a child to assume is known as “parentification” (Minuchin, Montalvo, Guerney, Rosman, & Schumer, 1967). For example, a parentified well-sibling might clean the house and prepare daily meals while his or her parents are at the hospital with the sick child. There are long-term, negative outcomes associated with parentification, including internalizing symptoms such as depression and anxiety (Earley & Cushway, 2002; Jacobvitz, Hazen, Curran, & Hitchens, 2004).
**Well-Sibling Age.** The age of the well-sibling is also an important illness parameter to consider when assessing adjustment to chronic illness. Similar to gender, differences in adjustment according to well-sibling age are often assessed by psychopathology measures (Gold, Treadwell, Weissman, & Vichinsky, 2011). The impact of age on well-sibling adjustment is difficult to measure using psychopathology assessments given that younger and older children are vulnerable to different psychological symptoms (Lahey, Schwab-Stone, Goodman, Waldman, Canino, Rathouz, Miller, Dennis, Bird, & Jensen, 2000). Older adolescent well-siblings of children diagnosed with cancer are more at risk for internalizing symptoms and exhibit higher levels of anxiety, more insecurity and loneliness, more traumatic stress, and greater emotional involvement with the illness than middle-school aged well-siblings (Alderfer, Labay, & Kazak, 2003; Houtzager et al., 2004). Younger well-siblings are more likely to exhibit externalizing symptoms such as aggression and hyperactivity (Thompson et al., 1994). Older well-siblings had poorer adjustment to type 1 diabetes than younger well-siblings (Jackson et al., 2008).

Given the age differences in psychopathology assessments (e.g., Lahey et al., 2000), there is a need to examine adjustment using a measure that is not influenced by age, such as quality of life. Quality of life outcomes are not different according to age (Varni et al., 2003). Therefore, if age differences are found in quality of life measures, these differences may be attributable to age differences in adjusting to the specific demands of the sick-child’s chronic illness, such as increased responsibilities with household chores. Some previous research suggested that older children are expected to
help more with domestic duties such as cleaning the house, baby-sitting and preparing meals (Houtzager et al., 2004). Younger well-siblings may feel ignored as a result of parents increased need to spend time with the sick child during treatments or hospital visits; well-siblings might demonstrate behavioral problems to attract attention from their parents (Hijmans, Grootenshui, Oosterlaan, Last, Heijboer, Peters, & Fijnvandraat, 2009). Older and younger well-siblings may adapt to disruptions in the family system in different ways; by measuring only psychopathology it is unclear as to whether age differences are related to specific demands of a chronic illness or are only a reflection of developmental age differences in internalizing and externalizing symptoms (Lahey et al., 2000; Ramtekkar, Reiersen, Todorov, & Todd, 2010). Therefore, there is a need to examine the well-sibling to predict well-sibling adjustment in domains related to quality of life, such as social functioning.

Sibling Process Variables

Though examining demographic and illness parameters alone offers a greater understanding of how a well-sibling might be expected to adjust based on these variables, these parameters, including age and gender, are unalterable. Many well-sibling studies focus on assessing only the direct relationship between a demographic or illness factor and well-sibling adjustment, and therefore do not capture the processes that underlie well-sibling adaptation to chronic illness (e.g., Houtazger, et al., 2004; Wood et al., 2008). Understanding other modifiable factors related to adjustment that predict better well-sibling adjustment, such as well-sibling adaptational processes, will provide information useful in helping children adjust to their brother or sister’s chronic illness.
One model that examines well-sibling processes related to adjustment is the transactional stress and coping model. This model was originally proposed to investigate the adjustment of sick-children and their mothers to the stress of childhood chronic illness, and identify processes underlying their adaptation (Thompson, Gustafson, Hamlett, & Spock, 1992a; Thompson, Gustafson, Hamlett, & Spock, 1992b). Recently this model was adapted to examine processes related to well-sibling adjustment to having a sibling with sickle-cell disease (Gold, Treadwell, Weissman, & Vichinsky, 2008). This adapted well-sibling model includes factors important to understanding well-sibling adjustment, such as illness parameters, demographic parameters, and adaptational processes of both the family and well-sibling. The present study will evaluate the well-sibling adaptational process of coping in predicting well-sibling adjustment to cystic fibrosis.

**Well-Sibling Coping.** Coping was selected as the focus of this study because it is an important, yet not well-understood, process affecting well-sibling adjustment. Compas and colleagues (2001) defined coping as “a goal-directed process in which the individual orients thoughts and behaviors toward the goals of resolving the source of stress and managing emotional reactions to stress” (pg. 88). Coping affects child adjustment to any stressful event (e.g., Eschenbeck, Kohlmann, & Lohaus, 2007; Griffith, Dubow, & Ippolito, 2000), and is therefore important in understanding well-sibling adjustment to chronic illness. Though stressful events in the lives of children and adolescents have been linked to emotional and behavioral problems, there is variability in the extent to which these stressors are associated with psychosocial problems (Compas,
In other words, different individuals may respond and adjust differently to the same stressor. These individual differences can be, at least partially, explained by coping; thus, coping is considered a protective factor for many emotional and behavioral difficulties in childhood and adolescence (Compas, 1987a; Compas et al, 2001). As previously discussed, the literature contains evidence of both positive and negative adjustment to the stressful event of having a sibling with a chronic illness; coping may be a way of understanding and explaining these differences.

Lazarus and Folkman (1984) developed a model to better understand coping. This model distinguishes between two types of coping: problem-focused and emotion-focused. In many studies examining child and adolescent coping, these have been labeled approach and avoidance strategies respectively, and these labels will henceforth be used (e.g., Compas et al., 2001; Griffith, Dubow, & Ippolito, 2000). Approach strategies refer to efforts made by a child or adolescent to master or change the stressful event, such as planned problem solving and seeking social support (Blount, Simons, Devine, Jaaniste, Psychol, Cohen, Chambers, & Hayutin, 2008; Hampel & Petermann, 2005; Taylor & Stanton, 2007). Avoidance strategies refer to attempts made by the individual to manage or regulate negative emotions surrounding an event, and examples include denial, distraction, wishful thinking, resignation and blaming others (Blount et al., 2008; Hampel & Petermann, 2005; Taylor & Stanton, 2007).

Studies of sick children adjusting to their own chronic illness found avoidance coping strategies were associated with negative adjustment, and approach coping strategies were associated with positive adjustment (e.g., Aldridge & Roesch, 2006;
Marsac, Funk, & Nelson, 2006; Wells, Booth-Jones, & Jacobson, 2009). Though these studies were not conducted with well-siblings coping with a sick-child’s chronic illness, one could extrapolate these data to this population as both well-siblings and sick-children are coping with the similar stressor of the illness. Other data suggest that healthy children and adolescents using avoidant coping with stressors other than chronic illness also have poorer adjustment and more symptoms of psychopathology (Griffith, Dubow, & Ippolito, 2000; Seiffge-Krenke, 2000; Seiffge-Krenke & Klessinger, 2000). One might predict that well-siblings would have similar positive outcomes with approach coping strategies, and negative outcomes with avoidant coping strategies. Longitudinal data demonstrate that healthy children can change their coping strategies, and consequently reduce symptoms of anxiety and depression (Hirshfeld-Becker, Masek, Henin, Blakely, Rettew, Dufton, Segool, & Beiderman, 2008). These data suggest modification of the well-siblings’ coping style can improve their adjustment, thus teaching effective coping strategies may be incorporated into interventions targeting well-siblings of children with chronic illness.

Lazarus and Folkman’s (1984) model emphasizes the importance of obtaining assessments of coping and adjustment directly from the individual facing the stressor. In well-sibling research, studies rely on parent proxy-reports of well-sibling’s adjustment and coping (e.g., Labay & Walco, 2004; Laufersweiler-Plass et al., 2003; Weiss et al., 2001), though research suggests that examining data from more than one informant is important when assessing child adjustment (Eiser & Jenney, 2007). Parental-report and well-sibling self-report of adjustment yield different information (Sharpe & Rossiter, 2002). Parent-reports of well-sibling adjustment are influenced by
their personal stress and worries (Cremeens et al, 2006; Houtzager et al., 2004). In general, parents who are depressed have a greater discrepancy with child self-report when reporting on their child’s symptoms (Treutler & Epkins, 2002; Youngstrom et al., 2000), and parents of a child with a chronic illness report emotional distress including anxiety and depression (Glasscoe, Lancaster, Smyth, & Hill, 2007; Vrijmoet-Wiersma, van Klink, Kolk, Koopman, Ball, & Egeler, 2008). Parental distress about the sick-child’s illness influences their assessment of the well-child’s quality of life (Houtzager, Grootenhuis, Caron, & Last, 2005), thus further highlighting the importance of collecting collateral information about the well-sibling’s adjustment from sources other than parents.

**Previous Research on Well-Sibling Coping.** Several studies that have explored coping for well-siblings adjusting to sick-children’s chronic illnesses are qualitative studies. These studies contain interviews with well-siblings regarding ways they handle the stress associated with the sick-child’s chronic illness. In one study, well-siblings were asked to offer advice to other children on ways to cope with having a sibling with cystic fibrosis or asthma (Derouin & Jessee, 1996). Some examples of coping advice included the following: “Talk about what’s going on, or you’ll feel like you’re going to explode” (an example of emotional expression, or an approach style of coping) and “when I’m home, playing helps me forget” (an example of distraction, an avoidant style of coping). In evaluating answers to a sentence-completion list, an additional study found that well-siblings of children with cancer demonstrated cognitive and behavioral coping techniques (Walker, 1988). Well-siblings endorsed cognitive strategies including seeking
information, processing information, and seeking support from others; behavioral strategies included self-soothing activities and distraction. Though these qualitative studies indicated well-siblings’ use of coping strategies to deal with the sick child’s chronic illness, they did not directly measure the relationship between these coping strategies and well-sibling adjustment to chronic illness.

Few quantitative studies have examined well-sibling coping with the sick-child’s chronic illness. Houtzager and colleagues (2004) examined coping and well-siblings’ adjustment to the sick-child’s diagnosis of cancer. Results indicated that cognitive coping strategies were correlated with well-sibling emotions (Houtzager et al., 2004). Specifically, well-siblings who described remaining optimistic about the course of the sick-child’s illness reported lower levels of anxiety, insecurity, and loneliness. Cognitive coping styles were not significantly related to a measure of quality of life of well-siblings of children with chronic illness. This study’s focus was limited to only cognitive coping strategies, and thus neglected other forms of coping that may be important for well-sibling adaptation, such as seeking social support.

Gold and colleagues (2008) examined the contribution of coping in well-siblings’ adjustment to sickle cell disease. This study measured well-siblings’ coping and divided these strategies into approach and avoidance coping styles. Avoidant styles of coping were significantly correlated with both internalizing and externalizing symptoms in well-siblings. Furthermore, avoidant coping style only predicted well-sibling externalizing behavior problems. Internalizing symptoms of well-siblings were not predicted by avoidant or approach styles of coping. This study used a global measure of coping rather
than specifically assessing well-sibling coping with chronic illness. Global measures of coping assess the general way in which an individual responds to a stressor. Well-siblings may cope with chronic illness in a specific way that is uniquely different than coping with other negative life events. For example, strategies that a well-sibling used to cope with having a fight with a friend or failing a test may be very different than the strategies used to cope with the chronic illness of the sick-child. Using a global measure may not accurately capture well-sibling coping with chronic illness. This study also only analyzed the relationship between coping and parent-reports of well-sibling internalizing and externalizing behavior problems, and consequently neglected other domains of functioning included in quality of life assessments, such as social relationships.

Some quantitative studies found no significant relationships between coping and well-sibling adjustment (e.g., Jackson, Richer, & Edge, 2008; Madan-Swain et al., 1993; Sloper & While, 1999). Madan-Swain and colleagues (1993) examined the coping of well-siblings of sick children with cancer by comparing the coping strategies of three groups: well-siblings of children with cancer, the sick-child with cancer, and a control group of siblings of healthy children. There were no significant differences among the three groups on coping, suggesting that well-siblings of children with chronic illness do not have difficulties coping, and use the same strategies as healthy controls. Another study found most coping strategies used by well-siblings of a sick-child with a chronic illness were not significantly correlated with well-sibling adjustment; only self-criticism, an avoidant style of coping, was significantly positively associated with well-sibling adjustment (Jackson, Richer, & Edge, 2008). Both of these studies used global measures
of coping that assessed the way the well-sibling coped with any stressors (such as fighting with a friend or failing a test); this global measure limited interpretation of the results as it may not have captured the specific processes involved in coping with a chronic illness.

Sloper and While’s (1996) study examined well-sibling’s adjustment to cancer by using a ratio of avoidance coping to total number of coping strategies to predict well-sibling behavior problems. The avoidance coping ratio is established in the adult literature, and allows for consideration of the relative use of a coping style for an individual by comparing how much the individual uses one coping strategy compared to all others (e.g., Hansdottir, Malcarne, Furst, Weisman, & Clements, 2004; Tamres, Janicki, & Helgeson, 2002; Yanos, Primavera, & Knight, 2001). There were no significant differences in the avoidance coping ratio between the two groups: well-siblings with high levels of behavior problems and well-siblings with low levels of behavior problems. One limitation of this study was the use of a measure of behavior problems in assessing well-sibling adjustment, given that previous studies demonstrated that well-siblings might have problems in other areas of functioning, such as somatic complaints, emotional problems, or social problems. Additionally, though there is some evidence to support the use of the avoidance coping ratio in children (e.g., Lipani & Walker, 2006; Mikolajczak, Petrides, & Hurry, 2009), it was originally developed for use with an adult population (Vitaliano, Maiuro, Russo, & Becker, 1987). It is unclear whether using the avoidance coping ratio is appropriate for children and adolescents, and
the utilization of this ratio may explain why coping did not predict psychosocial problems in well-siblings in Sloper and While’s (1996) study.

The discrepancies in the literature regarding the impact of coping on well-sibling adjustment have several possible explanations. One explanation is related to whether or not well-siblings completed global measures of coping rather than specific measures relating to the impact of chronic illness. The impact of coping also might be different depending on illness parameters of the sample used, such as disease severity or time since diagnosis; data on these illness parameters are not reported in every study. It is possible, then, that studies that found no relationship between well-sibling adjustment and coping contained samples of well-siblings of children with long-standing, well-controlled, and less severe illnesses. These siblings would be more likely to experience sufficient time to adjust to the impact of chronic illness. Most quantitative studies that examined coping included only well-siblings of children with cancer (e.g., Houtzager et al., 2004; Madan-Swain et al., 1993; Sloper & While, 1996). There are no known published data examining the impact of coping on well-siblings of children with cystic fibrosis.

Present Study

The present study sought to resolve discrepancies in the literature by examining the ability of demographic and illness parameters to predict the quality of life of well-siblings of children with chronic illness. Using the non-categorical approach, this study investigated the quality of life of well-siblings of children with cystic fibrosis. Specifically, this study examined the predictive value of well-sibling age, well-sibling
gender, illness severity, and time since diagnosis on quality of life of well-siblings of children with cystic fibrosis. Additionally, after controlling for the aforementioned demographic and illness parameters, the present study examined the predictive value of coping strategies’ use on well-sibling quality of life. Total number of coping strategies, avoidance and approach styles of coping, and the avoidance coping ratio (Sloper and While, 1999) were examined as individual predictors of well-sibling adjustment. Use of coping strategies is limited in interpretation as it does not provide information about the well-sibling’s evaluation of the strategy in coping with the chronic illness, thus the present study also examined well-sibling subjective report of helpfulness of coping and well-sibling subjective distress. Finally, well-sibling adjustment was measured by both parent-report and child-report forms; these forms were compared to examine potential differences by informant.

Hypothesis 1:

Demographic parameters of age and gender, as well as illness factors of illness severity and time since diagnosis were hypothesized to statistically predict quality of life of well-siblings of children with cystic fibrosis. It was expected that younger well-siblings and male well-siblings would have better quality of life because they would be less likely to be expected to help with daily chores or routines that may be disrupted by the sick-child’s illness. It was also predicted that well-siblings of sick-children with less severe cystic fibrosis would have better quality of life because the illness would not impact the well-sibling’s daily life as significantly as a more severe illness. Finally, well-
siblings of sick-children diagnosed more than one year ago would have better quality of life as they would have more time to adjust to the illness.

*Hypothesis 2:*

After controlling for demographic and illness parameters, coping was expected to significantly predict quality of well-sibling’s life. Coping included cognitive and behavioral strategies aimed at managing the specific stressful situation of having a sibling with cystic fibrosis. Based on previous research, it was expected that more frequent use of approach coping strategies would predict positive well-sibling quality of life, and more frequent use of avoidant coping strategies would predict negative well-sibling quality of life. Next, number of total coping strategies was predicted to significantly predict well-sibling adjustment. Use of a greater number of coping strategies was expected to predict better well-sibling adjustment than use of fewer coping strategies. This hypothesis was based on the expectation that, in using a variety of coping strategies, well-siblings would be more likely to find a coping style that is effective. Finally, the avoidance coping ratio (Sloper and While, 1999) was hypothesized to predict well-sibling adjustment. Though this ratio was not found to be significant in Sloper and While’s (1999) study, it was hypothesized that it would be predictive of well-sibling quality of life given the numerous studies in the adult literature that support use of this ratio. Well-siblings with a higher ratio of avoidance coping strategies to total number of coping strategies were predicted to have poorer adjustment.

*Hypothesis 3:*
In examining well-sibling subjective ratings, approach styles of coping were predicted to be more helpful than avoidant styles of coping. Approach styles of coping were more active and focused on managing the stressor, and therefore were predicted to be appraised as more helpful to well-siblings. Avoidance styles of coping were aimed at regulating negative emotions about the stressor and were not as active as approach styles of coping, and therefore were thought to be rated as less helpful to well-siblings.

*Hypothesis 4:*

Well-sibling subjective reports of distress were hypothesized to predict well-sibling adjustment after controlling for significant well-sibling demographic factors or illness factors from Hypothesis 1. Specifically, well-sibling reports of greater anxiety, sadness, and anger were expected to predict poorer well-sibling adjustment. Well-siblings distress was expected to impair well-sibling quality of life in domains such as emotional and social functioning.

*Hypothesis 5:*

Well-siblings of sick-children with more severe cystic fibrosis were predicted to be more distressed by the illness than well-siblings of sick-children with less severe cystic fibrosis. Well-siblings of sick-children with greater severity were expected to be more worried, anxious, and sad about their brother or sister.

*Hypothesis 6:*

Self-report and parent-report of well-sibling adjustment were hypothesized to be significantly different from each other. Specifically, parent-report was predicted to be
significantly lower as their own quality of life was expected to negatively impact their ratings of well-sibling report.
METHODS

Participants

Fifty-four families agreed to participate in the study. Of those 54 families, eight well-siblings were lost to follow-up after they could not be reached by telephone. An additional three families did not complete all assessments and were consequently excluded from analyses. After these 11 families were removed, 43 participants were included in study analyses and descriptive statistics are displayed in Table 1; missing data analyses were conducted using chi-square analyses and independent t-tests to determine if there were significant differences between those included and excluded from analyses. Results of chi-squared analyses revealed no significant differences between those participants with missing data and those with complete data on well-sibling gender, well-sibling insurance type, sibling relationship type, single parent status, parent education, or parent relationship type. There were significant differences in well-sibling ethnicity, as the only two African-American well-siblings in the study were both lost to follow-up ($\chi^2(2, N = 52) = 13.459, \ p < .01$). Results of independent t-test indicated no significant differences between those participants with missing data and those with complete data for well-sibling age, sick-child age, parent age, time since diagnosis, and severity of illness.

Of participants with complete data, well-siblings were identified as biological siblings (79.1%), half-siblings (9.3%), stepsiblings (9.3%), and foster sibling (2.3%).
Table 1. Descriptive statistics for participants included and excluded from analysis.

<table>
<thead>
<tr>
<th></th>
<th>Included (n = 46)</th>
<th>Excluded (n = 11)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Well-sibling Gender</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Female</td>
<td>23 (51.1%)</td>
<td>5 (62.5%)</td>
</tr>
<tr>
<td>Male</td>
<td>22 (48.9%)</td>
<td>3 (37.5%)</td>
</tr>
<tr>
<td><strong>Well-Sibling Ethnicity</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>African-American</td>
<td>0 (0%)</td>
<td>2 (28.6%)</td>
</tr>
<tr>
<td>Caucasian</td>
<td>44 (97.8%)</td>
<td>5 (71.5%)</td>
</tr>
<tr>
<td>Multiracial</td>
<td>1 (.02%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td><strong>Mean (SD) Well-Sibling Age</strong></td>
<td>12.04 (3.4)</td>
<td>12.88 (3.9)</td>
</tr>
<tr>
<td><strong>Mean (SD) Sick-Child Age</strong></td>
<td>9.89 (4.6)</td>
<td>11.25 (6.6)</td>
</tr>
<tr>
<td><strong>Mean (SD) Time Since Diagnosis</strong></td>
<td>117.78 (61.0)</td>
<td>158.6 (86.0)</td>
</tr>
<tr>
<td><strong>Mean (SD) Severity of Illness</strong></td>
<td>80.1 (13.2)</td>
<td>82.2 (14.5)</td>
</tr>
<tr>
<td><strong>Well-Sibling Insurance</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Medicaid</td>
<td>11 (24.4%)</td>
<td>2 (25%)</td>
</tr>
<tr>
<td>Medicaid-HMO</td>
<td>8 (33.3%)</td>
<td>2 (25%)</td>
</tr>
<tr>
<td>Private Insurance</td>
<td>24 (53.3%)</td>
<td>2 (25%)</td>
</tr>
<tr>
<td>Self-Pay</td>
<td>2 (.04%)</td>
<td>2 (25%)</td>
</tr>
<tr>
<td><strong>Number of Single Parents</strong></td>
<td>10 (22.72%)</td>
<td>1 (12.5%)</td>
</tr>
<tr>
<td><strong>Mean (SD) Parent Age</strong></td>
<td>39.76 (7.4)</td>
<td>38.13 (7.4)</td>
</tr>
</tbody>
</table>
At the time of the study, all well-siblings lived at home with the sick-child for at least one year. In the family that included a foster sibling, the parent completing the forms was the biological mother of the well-sibling and the sick-child was a two year-old foster child who had lived with the family for over a year. Those participants who were biological siblings were compared to participants of all other types of sibling relationships (including half-siblings, stepsiblings, and foster sibling) to determine if there were any group differences based on relationship type using paired-samples t-tests. There were no significant group differences on well-sibling demographic factors (age and gender), well-sibling adjustment (quality of life of well-siblings, both parent-report and self-report), and well-sibling self-reported distress about the sick-child’s illness. Per parental report, almost all well-siblings were identified as Caucasian (97.7%), with only one well-sibling identified as multiracial (2.3%). These ethnicity data are largely consistent with estimates of cystic fibrosis, which is a disease that is more predominant in Caucasians than other ethnicities (Cystic Fibrosis Foundation, 2006). The demographic population typically served by the Cystic Fibrosis Clinic might also explain the large proportion of Caucasian participants. Though the present study did not collect data on the exact number of Amish patients enrolled, the Clinic serves many Amish patients.

The mean age of well-siblings was 12.05 years (range from 7 to 19 years), and 51.2% were female. Well-siblings’ insurance types included Medicaid (27.9%), Medicaid-HMO (18.6%), private insurance (48.8%), and self-pay (4.7%). One must exercise caution in interpreting insurance as a proxy for socioeconomic status in this
study, as patients of Amish culture are usually self-pay (Weyer, Hustey, Rathbun, Armstrong, Reed, Ronyak, & Savrin, 2003). Sick-children were a mean age of 10.26 years (range 2 to 18 years), and 46.5% were female. Parents were a mean age of 39.74 years (range 25 to 62 years), and 23.3% identified as single parents. Parents or guardians completing the measures included biological mothers (69.8%), biological fathers (16.3%), stepmothers (9.3%), and grandmothers (4.7%). All participants identified as primary care givers for the well-sibling. Parents reported the following levels of education: some high school (9.3%); high school or GED (32.6%); some college or professional school (32.6%); bachelor, associates, professional degree (14.0%); some graduate school (2.3%), and graduate or advanced degree (4.7%).

**Measures**

*Demographic Questionnaire* (See Appendix A). A demographic questionnaire was created for this study. This form was completed by parents, and included demographic information about the parent (marital status, educational attainment, and relationship to well-sibling), well-sibling (age, ethnicity, gender, and insurance type), and sick-sibling (age, ethnicity, and gender). On the Demographic Questionnaire, the parent provided the month and year of diagnosis of cystic fibrosis in the sick-child. This date was used to calculate the amount of time since diagnosis.

Time since diagnosis and well-sibling age were significantly positively correlated ($r = .454, p < .01$), such that older well-siblings were related to sick-children diagnosed longer ago. The mean time since diagnosis for the current study was 111.4 months, with a minimum of 23 and a maximum of 219 months. Those well-siblings of sick-children
diagnosed less than one year were hypothesized to have poorer adjustment, but this hypothesis was unable to be tested due to the limited range of time since diagnosis. It is likely that well-siblings had sufficient time to stabilize since the minimum time since diagnosis was almost two years. Given this stabilization and the significant correlation between well-sibling age, time since diagnosis was removed from future analyses and well-sibling age was retained.

*Kidcope* (Spirito et al., 1988) (See Appendix B). The Kidcope was created to measure cognitive and behavioral coping strategies in children and adolescents. The Kidcope is a 15-item inventory that assesses ten coping strategies: blaming others, distraction, problem solving, self-criticism, emotional regulation, social withdrawal, wishful thinking, cognitive restructuring, seeking social support, and resignation. Children are asked to identify both the use of an individual coping strategy (“Did you do this?”) and the effectiveness of the strategy (“How much did it help?”). Participants’ responses on these two questions are categorized into the Use Scale or Helpfulness Scale respectively. The Use Scale is scored as follows: a “yes” response is scored as 1 and a “no” response is scored as 0. In the Helpfulness Scale, children select from the following responses to indicate how much the coping strategy helped them: “not at all” (scored as 0), “a little” (scored as 1), and “a lot” (scored as 2). Similar to other researchers using the Kidcope (e.g., Frank, Blount, & Brown, 1997; Gold et al., 2008), Use Scale scores were used in primary analyses of the current study.

The Kidcope allows for presentation of a standard problem to all children. In this study, the following script related to coping with having a sibling with a chronic illness
was presented to each well-sibling: “We are trying to find out how children deal with having a brother or sister with cystic fibrosis. Please think about the way you deal with having a brother or sister with cystic fibrosis and answer the questions.” To assess well-siblings’ distress, the authors of the Kidcope suggest participants rate their anxiety, sadness, and anger about the stressor (here, the sick-child’s cystic fibrosis) on a 5-point Likert scale. The well-siblings were asked the following questions that were written by the authors of the Kidcope: “Did this situation [having a brother/sister with cystic fibrosis] make you nervous?”; “Did this situation [having a brother/sister with cystic fibrosis] make you sad?”; “Did this situation [having a brother/sister with cystic fibrosis] make you angry or mad?” Respondents rated each question on a 5-point Likert scale: “not at all (scored as 0),” “a little (scored as 1),” “somewhat (scored as 2),” “pretty much (scored as 3),” and “very much (scored as 4).”

There are two versions of the Kidcope available: one for children aged 7 to 12 years, and one for adolescents aged 13 to 18 years. The most significant difference between the two versions is the Use Scale. In the younger version, children indicate “yes” or “no” to use of a strategy; in the older version, adolescents indicate how often they use the strategy (“not at all,” “sometimes,” “a lot of the time,” and “almost all of the time”). The authors suggest researchers using samples with both age groups use the younger version of the scale. Construct validity in the initial standardization study was assessed by comparing this measure with other measures of coping, such as the Coping Strategies Inventory (CSI) and the Adolescent-Coping Orientation from Problem
Experiences Inventory (ACOPE) (Spirito et al., 1988); correlations with the CSI ranged from 0.55 to 0.77, and with the ACOPE ranged from 0.43 to 0.62.

Past factor analyses of the Kidcope yielded both one-factor and two-factor structure solutions for sick-children coping with their own illness (Spirito et al., 1988; Spirito, Stark, & Tyc, 1994). There are no available factor analyses of the Kidcope conducted with well-siblings, therefore the present study examined both the single and two-factor scores of the Kidcope. The two-factor structure of approach and avoidance of the Kidcope was identified for sick-children coping with a discrete episode, such as an aversive medical procedure (Spirito et al., 1988). This two-factor solution was also previously used in well-sibling studies (Gold et al., 2008; Sloper & While, 1999). The approach coping style factor includes coping strategies of problem solving, cognitive restructuring, social support, and emotional regulation. The avoidance coping style factor consists of blaming others, distraction, wishful thinking, and resignation. The coping strategies identified as approach and avoidant coping were summed to yield a total score ranging from 0 and 4 for each coping style. Table 2 illustrates the items that load onto each of the coping style factors. The single factor structure was supported when the proposed stressor for the sick-child was experienced over a longer period of time, such as a hospital stay over several days (Spirito et al., 1994). The single factor included all items on the Kidcope. The internal consistencies of the one- and two-factor structures of the Kidcope were examined using Cronbach’s alpha. The internal consistency of the one-factor structure in the present study was acceptable ($\alpha = 0.70$). For the two-factor scale, internal consistency of the avoidance coping style factor in the present study was very
Table 2. The coping strategies and corresponding item numbers for the coping styles assessed by the Kidcope.

<table>
<thead>
<tr>
<th>Coping Strategy</th>
<th>Item Number</th>
<th>Coping Style</th>
</tr>
</thead>
<tbody>
<tr>
<td>Distraction</td>
<td>1,2</td>
<td>Avoidance</td>
</tr>
<tr>
<td>Cognitive Restructuring</td>
<td>5</td>
<td>Approach</td>
</tr>
<tr>
<td>Blaming Others</td>
<td>7</td>
<td>Avoidance</td>
</tr>
<tr>
<td>Problem Solving</td>
<td>8, 9</td>
<td>Approach</td>
</tr>
<tr>
<td>Emotional Expression</td>
<td>10, 11</td>
<td>Approach</td>
</tr>
<tr>
<td>Wishful Thinking</td>
<td>12, 13</td>
<td>Avoidance</td>
</tr>
<tr>
<td>Social Support</td>
<td>14</td>
<td>Approach</td>
</tr>
<tr>
<td>Resignation</td>
<td>15</td>
<td>Avoidance</td>
</tr>
</tbody>
</table>
poor ($\alpha = 0.18$), and approach coping style factor was acceptable ($\alpha = 0.78$). Given the very poor internal consistency of the avoidance coping style factor, it was used in subsequent analyses with caution and results from these analyses were considered exploratory.

*Pediatric Quality of Life Inventory (PedsQL™)* (Varni, Seid, & Kurtin, 2001) (See Appendices C and D). The PedsQL™ is a 23-item measure that evaluates children’s and adolescents’ quality of life. It examines the domains of Physical Functioning, Emotional Functioning, Social Functioning, and School Functioning. Some items on the School Functioning scale include: “paying attention in class,” “missing school because of not feeling well,” and “missing school to go to the doctor or hospital.” The School Functioning scale of the PedsQL™ has previously been eliminated because the samples included children too young to attend school (Varni, Burwinkle, Seid, & Skarr, 2003). Of the forty-three participants in the current study, four indicated they were home-schooled. Home-schooled children may interpret questions on the School Functioning scale differently than children in public or private schools. Therefore, the School Functioning scale was removed from analyses because including this domain of functioning for some participants but not others would result in an unequal and different composition of quality of life across subjects. For example, a participant in a public school with attention or organization difficulties would demonstrate impaired quality of life if the School Functioning scale was included in the assessment. Quality of life for this same participant would not appear impaired if the participant was enrolled in home-school, as the School Functioning scale would be eliminated. This unequal assessment would create inequitable
quality of life scores, thus limiting interpretation. Consequently the outcome measure of well-sibling adjustment in the present study was a composite variable created from the Physical Functioning, Emotional Functioning, and Social Functioning scales on the well-sibling self-report PedsQL™. Severity of illness was created from the Physical Functioning, Emotional Functioning, and Social Functioning for the parent-report PedsQL™ of the sick-child.

The PedsQL™ has parallel forms available for self-report and proxy-report by the parent. Respondents are asked to indicate how much of a problem each item has been for them in the past month. The ratings are made on a 5-point Likert scale including “never” (scored as 0), “almost never” (scored as 1), “sometimes” (scored as 2), “often” (scored as 3), and “almost always” (scored as 4). To score the PedsQL™, items are reverse scored such that higher scores indicate better quality of life. The items are linearly transformed to a 0 to 100 scale; therefore, a score of 0 is transformed to 100, 1 to 75, 2 to 50, 3 to 25, and 4 to 0 (Varni et al., 2001). The total scale score is the mean of all the responses and is computed by calculating the sum of all the items answered and dividing by the number of items answered. Using the mean of the scale also accounts for missing data. There are different forms available for four age ranges: toddler (2 to 4 years), young child (5 to 7 years), child (8 to 12 years), and adolescent (13 to 18 years). The difference between the forms is the use of more age-appropriate wording to label participants in the self-report, such as “teens” rather than “kids.” For example, the child version reads, “I have trouble getting along with other kids” and the adolescent version reads, “I have trouble getting along with other teens.”
To interpret scores of the PedsQL™, Varni and colleges (2003) established cut-offs. These cut-off scores are one standard deviation below the mean of the total population sample of healthy children and adolescents, and well-siblings with self-reported total scores of less than 71.44 or with parent-report scores of less than 67.44 are considered at-risk for impaired quality of life (Varni et al, 2003). The PedsQL™ demonstrated adequate convergent, discriminant, and construct validity (Varni et al., 2001). The internal consistencies of the quality of life in the present study were examined using Cronbach’s alpha. After removing the School Functioning scale, internal consistencies were good for parent-report of the sick-child (α = 0.84), parent-report of the well-sibling (α = 0.85), and well-sibling self-report (α = 0.86).

Severity of Illness. The survivorship score was collected from the sick-child’s medical records to estimate severity of illness. This score is intended to be calculated annually, and is derived from a formula that includes many factors, including pulmonary functioning, weight for age, pancreatic sufficiency, and annual number of acute pulmonary exacerbations (Liou, Adler, FitzSimmons, Cahill, Hibbs, & Marshall, 2001). Only 35 out of 53 (66.1%) sick-children that participated in the study had a survivorship score. Inability to complete the pulmonary functioning tests was the most common reason for lack of a survivorship score. Of the children with survivorship scores, 25 out of 35 (71.4%) were completed over a year ago, making current severity of illness impossible to accurately assess using survivorship score. Therefore, severity of illness was assessed using a PedsQL™ for the sick-child that was completed by the parents. The PedsQL™ is...
a valid measure that was designed to assess severity of illness (Varni et al., 1999), such that sick-children with poorer quality of life scores have a greater severity of illness.

Procedure

Children and adolescents with cystic fibrosis and their parents were approached at the Cystic Fibrosis Clinic at Akron Children’s Hospital during routine outpatient visits. The principal investigator or an undergraduate research assistant trained in the research protocol collected the data. These data collectors followed a protocol that was approved by the Institutional Review Boards of Kent State University and Akron Children’s Hospital. The Cystic Fibrosis Clinic met once per week, and children with cystic fibrosis and their families presented to the clinic for routine follow-up with several specialists involved in their care, including pulmonologists, dieticians, nurses, physical therapists, and social workers. In order to avoid contact between sick-children with cystic fibrosis who might incidentally infect one another with various strains of bacteria, sick-children and their families were immediately escorted to individual exam rooms upon check-in at the clinic. Each health care provider rotated through the individual exam rooms. The researcher also worked within this model, and parents were recruited while waiting in exam rooms for health care providers’ visits.

To be eligible for the study, individuals with cystic fibrosis were required to have a well-sibling without a chronic illness between the ages of 7 and 18 years. The age range selected includes both children and adolescents, and the minimum age of participants was selected to ensure that all children were able to read the forms necessary for the study. In an effort to include a greater number of participants, the upper range of data collection
was expanded to 19 years if the well-sibling still lived at home with the sick-child; expanding the age range added one participant. Only one well-sibling per family was included to avoid violating assumptions related to independence of data. If the sick-child had more than one well-sibling within the qualifying age range, the sibling closest in age to the individual with cystic fibrosis was selected for participation.

Those parents who consented to participate in the study completed the Demographic Questionnaire and two parent-report forms of the PedsQL™ (for both the well-sibling and sick-child). The instruments that the parents completed (Demographic Questionnaire, PedsQL™ for sick-child and well-sibling) took parents approximately ten minutes to complete. The parent was asked to complete the appropriate measures during his or her time in the clinic, and then provided the name of the well-child and a phone number at which he or she could be reached. The researcher contacted the well-sibling to administer the appropriate instruments by phone. The researcher called the well-sibling within two business days of the office visit; if the well-sibling was not reachable at this time, the researcher tried several times more over the next several weeks. Of the 54 participants, 8 well-siblings could not be reached by phone for follow-up; all well-siblings that were reached for follow-up consented to the study. After the well-sibling was reached by phone, the study was explained to him or her using the following script: “You are being asked to be in this study because you have a brother or sister with cystic fibrosis. We want to learn more about how this makes you feel, and what you do to deal with this. We hope that we can find ways to help children who have a brother or sister who is sick.” After the study was explained and they had an opportunity to ask questions,
well-siblings were asked for verbal assent to the study. The researcher then administered
the Kidcope and the self-report form of the PedsQL™ to the well-sibling over the phone.
The self-report form of the PedsQL™ was used as the outcome measure of sibling
adjustment. Following completion of these assessments, the researcher mailed the well-
sibling a $5 gift card to a Target® Store. Table 3 contains the correlations among all
variables used in the present study.

Data Analysis Plan

Pearson point-biserial correlations were used to determine bivariate relationships
of all study variables (see Table 3). Linear regressions and hierarchical linear regression
analyses were utilized to evaluate study hypotheses. Linear regressions were selected
because these analyses explain the relationship between predictor variables entered
simultaneously on a dependent variable (Tabachnick & Fidell, 2001). Linear regressions
yielded the degree of association of demographic parameters (well-sibling age and
gender) and illness parameter (severity of illness) with well-sibling self-reported quality
of life. Those demographic and illness characteristics significantly associated with well-
sibling quality of life ($p \leq .05$) that were identified from linear regressions were
subsequently used as control variables in the hierarchical linear regressions.

Hierarchical linear regression was selected because it allows for exploration of the
impact of several predictor variables on a dependent variable, while controlling for the
impact of other variables (Tabachnick & Fidell, 2001). Predictor variables are entered in
progressive steps, and consulting the $R^2$ change statistic after each step yields information
on whether inclusion of these new predictor variables significantly improves the
Table 3. Correlational table among all study variables.

<table>
<thead>
<tr>
<th></th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
<th>9</th>
<th>10</th>
<th>11</th>
<th>12</th>
<th>13</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>--</td>
<td>.03</td>
<td>.06</td>
<td>.01</td>
<td>-.01</td>
<td>-.30</td>
<td>-.04</td>
<td>-.13</td>
<td>-.18</td>
<td>.47†</td>
<td>.05</td>
<td>.78†</td>
<td>-.15</td>
</tr>
<tr>
<td>2</td>
<td>.03</td>
<td>--</td>
<td>.17</td>
<td>-.15</td>
<td>-.20</td>
<td>.09</td>
<td>.14</td>
<td>-.26</td>
<td>.10</td>
<td>-.05</td>
<td>.02</td>
<td>-.02</td>
<td>-.15</td>
</tr>
<tr>
<td>3</td>
<td>.06</td>
<td>.17</td>
<td>--</td>
<td>.17</td>
<td>-.04</td>
<td>.35*</td>
<td>-.09</td>
<td>.03</td>
<td>-.06</td>
<td>.27</td>
<td>-.15</td>
<td>.10</td>
<td>.06</td>
</tr>
<tr>
<td>4</td>
<td>.00</td>
<td>-.15</td>
<td>.12</td>
<td>--</td>
<td>.32*</td>
<td>.07</td>
<td>-.29</td>
<td>.37</td>
<td>.08</td>
<td>.01</td>
<td>-.02</td>
<td>-.10</td>
<td>-.07</td>
</tr>
<tr>
<td>5</td>
<td>-.01</td>
<td>-.20</td>
<td>-.04</td>
<td>.32*</td>
<td>--</td>
<td>-.09</td>
<td>.13</td>
<td>.06</td>
<td>.49†</td>
<td>.25</td>
<td>.21</td>
<td>.01</td>
<td>.34*</td>
</tr>
<tr>
<td>6</td>
<td>-.30</td>
<td>.09</td>
<td>.35*</td>
<td>.07</td>
<td>-.09</td>
<td>--</td>
<td>.05</td>
<td>.09</td>
<td>.03</td>
<td>-.10</td>
<td>-.04</td>
<td>-.10</td>
<td>.06</td>
</tr>
<tr>
<td>7</td>
<td>-.04</td>
<td>.14</td>
<td>-.09</td>
<td>-.29</td>
<td>.13</td>
<td>.05</td>
<td>--</td>
<td>-.13</td>
<td>.15</td>
<td>.22</td>
<td>.36*</td>
<td>.03</td>
<td>.33*</td>
</tr>
<tr>
<td>8</td>
<td>-.13</td>
<td>-.26</td>
<td>.03</td>
<td>.37*</td>
<td>.06</td>
<td>.09</td>
<td>-.13</td>
<td>--</td>
<td>-.17</td>
<td>-.27</td>
<td>.13</td>
<td>-.02</td>
<td>-.12</td>
</tr>
<tr>
<td>9</td>
<td>-.18</td>
<td>.10</td>
<td>-.06</td>
<td>.08</td>
<td>.49†</td>
<td>.03</td>
<td>.15</td>
<td>-.17</td>
<td>--</td>
<td>.00</td>
<td>.32*</td>
<td>-.04</td>
<td>.20</td>
</tr>
<tr>
<td>10</td>
<td>.47†</td>
<td>-.05</td>
<td>.27</td>
<td>.01</td>
<td>.25</td>
<td>-.10</td>
<td>.22</td>
<td>-.27</td>
<td>.00</td>
<td>--</td>
<td>.14</td>
<td>.45†</td>
<td>.09</td>
</tr>
<tr>
<td>11</td>
<td>.05</td>
<td>.02</td>
<td>-.15</td>
<td>-.02</td>
<td>.21</td>
<td>-.04</td>
<td>.36*</td>
<td>.13</td>
<td>.32*</td>
<td>.14</td>
<td>--</td>
<td>.16</td>
<td>.01</td>
</tr>
<tr>
<td>12</td>
<td>.78†</td>
<td>-.02</td>
<td>.10</td>
<td>.10</td>
<td>.01</td>
<td>-.10</td>
<td>.03</td>
<td>-.02</td>
<td>-.04</td>
<td>.45†</td>
<td>.16</td>
<td>--</td>
<td>.04</td>
</tr>
<tr>
<td>13</td>
<td>-.15</td>
<td>-.15</td>
<td>.06</td>
<td>-.07</td>
<td>.34*</td>
<td>.06</td>
<td>.33*</td>
<td>-.12</td>
<td>.19</td>
<td>.09</td>
<td>.01</td>
<td>.04</td>
<td>--</td>
</tr>
<tr>
<td>14</td>
<td>.04</td>
<td>.25</td>
<td>.31*</td>
<td>.01</td>
<td>.05</td>
<td>.07</td>
<td>.23</td>
<td>-.23</td>
<td>.03</td>
<td>.28</td>
<td>.13</td>
<td>.15</td>
<td>.35*</td>
</tr>
<tr>
<td>15</td>
<td>-.17</td>
<td>-.18</td>
<td>-.11</td>
<td>-.04</td>
<td>-.12</td>
<td>.00</td>
<td>.22</td>
<td>-.11</td>
<td>-.19</td>
<td>-.17</td>
<td>-.04</td>
<td>-</td>
<td>.28</td>
</tr>
<tr>
<td>16</td>
<td>.10</td>
<td>-.10</td>
<td>-.03</td>
<td>.01</td>
<td>.12</td>
<td>.00</td>
<td>-.15</td>
<td>-.08</td>
<td>.11</td>
<td>.06</td>
<td>-.06</td>
<td>-.08</td>
<td>-.43†</td>
</tr>
<tr>
<td>17</td>
<td>-.15</td>
<td>.03</td>
<td>-.01</td>
<td>.06</td>
<td>-.08</td>
<td>.02</td>
<td>.14</td>
<td>.17</td>
<td>-.12</td>
<td>-.05</td>
<td>.11</td>
<td>.03</td>
<td>.36*</td>
</tr>
</tbody>
</table>
Table 3. Continued

<table>
<thead>
<tr>
<th></th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
<th>9</th>
<th>10</th>
<th>11</th>
<th>12</th>
<th>13</th>
</tr>
</thead>
<tbody>
<tr>
<td>18</td>
<td>-.08</td>
<td>.22</td>
<td>.08</td>
<td>.00</td>
<td>.00</td>
<td>-.01</td>
<td>.24</td>
<td>-.13</td>
<td>.06</td>
<td>-.12</td>
<td>-.06</td>
<td>-.03</td>
<td>.41†</td>
</tr>
<tr>
<td>19</td>
<td>-.11</td>
<td>.17</td>
<td>.05</td>
<td>.06</td>
<td>.16</td>
<td>.07</td>
<td>.16</td>
<td>-.10</td>
<td>.18</td>
<td>-.12</td>
<td>-.07</td>
<td>-.15</td>
<td>.14</td>
</tr>
<tr>
<td>20</td>
<td>-.15</td>
<td>.31*</td>
<td>-.06</td>
<td>.04</td>
<td>-.04</td>
<td>-.08</td>
<td>-.11</td>
<td>.15</td>
<td>.16</td>
<td>-.40†</td>
<td>.11</td>
<td>-.19</td>
<td>-.16</td>
</tr>
<tr>
<td>21</td>
<td>-.12</td>
<td>.13</td>
<td>.04</td>
<td>.04</td>
<td>-.06</td>
<td>-.02</td>
<td>.23</td>
<td>-.02</td>
<td>-.04</td>
<td>-.08</td>
<td>.02</td>
<td>.02</td>
<td>.46†</td>
</tr>
<tr>
<td>22</td>
<td>-.13</td>
<td>-.15</td>
<td>-.07</td>
<td>.06</td>
<td>-.04</td>
<td>-.22</td>
<td>.13</td>
<td>-.03</td>
<td>-.03</td>
<td>-.06</td>
<td>-.01</td>
<td>-.06</td>
<td>.51†</td>
</tr>
<tr>
<td>23</td>
<td>-.27</td>
<td>.06</td>
<td>.14</td>
<td>.36*</td>
<td>-.04</td>
<td>.17</td>
<td>-.13</td>
<td>.09</td>
<td>.10</td>
<td>-.30*</td>
<td>.07</td>
<td>-.17</td>
<td>-.27</td>
</tr>
<tr>
<td>24</td>
<td>.23</td>
<td>.12</td>
<td>.05</td>
<td>.08</td>
<td>.27</td>
<td>.24</td>
<td>-.01</td>
<td>-.26</td>
<td>.19</td>
<td>.10</td>
<td>.12</td>
<td>.09</td>
<td>-.10</td>
</tr>
<tr>
<td>25</td>
<td>.14</td>
<td>-.12</td>
<td>.16</td>
<td>.17</td>
<td>.42†</td>
<td>.07</td>
<td>-.13</td>
<td>.05</td>
<td>.09</td>
<td>.45†</td>
<td>.00</td>
<td>.14</td>
<td>-.02</td>
</tr>
</tbody>
</table>

*p<.05; †p<.01

Table 3. Continued

<table>
<thead>
<tr>
<th></th>
<th>14</th>
<th>15</th>
<th>16</th>
<th>17</th>
<th>18</th>
<th>19</th>
<th>20</th>
<th>21</th>
<th>22</th>
<th>23</th>
<th>24</th>
<th>25</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>.04</td>
<td>-.17</td>
<td>.10</td>
<td>-.15</td>
<td>-.08</td>
<td>-.11</td>
<td>-.15</td>
<td>-.12</td>
<td>-.13</td>
<td>-.27</td>
<td>.23</td>
<td>.14</td>
</tr>
<tr>
<td>2</td>
<td>.25</td>
<td>-.18</td>
<td>-.10</td>
<td>.03</td>
<td>.22</td>
<td>.17</td>
<td>.31*</td>
<td>.13</td>
<td>-.15</td>
<td>.06</td>
<td>.12</td>
<td>-.12</td>
</tr>
<tr>
<td>3</td>
<td>.31*</td>
<td>-.11</td>
<td>-.03</td>
<td>-.01</td>
<td>.08</td>
<td>.05</td>
<td>-.06</td>
<td>.04</td>
<td>-.07</td>
<td>.14</td>
<td>.05</td>
<td>.16</td>
</tr>
<tr>
<td>4</td>
<td>.01</td>
<td>-.04</td>
<td>.01</td>
<td>.06</td>
<td>.00</td>
<td>.06</td>
<td>.04</td>
<td>.04</td>
<td>.06</td>
<td>.36*</td>
<td>.08</td>
<td>.17</td>
</tr>
<tr>
<td>5</td>
<td>.05</td>
<td>-.12</td>
<td>.12</td>
<td>-.08</td>
<td>-.01</td>
<td>.16</td>
<td>-.04</td>
<td>-.04</td>
<td>-.04</td>
<td>-.04</td>
<td>.27</td>
<td>.42†</td>
</tr>
<tr>
<td>6</td>
<td>.07</td>
<td>.00</td>
<td>.00</td>
<td>.02</td>
<td>-.02</td>
<td>.07</td>
<td>-.08</td>
<td>-.02</td>
<td>-.22</td>
<td>.17</td>
<td>.24</td>
<td>.07</td>
</tr>
<tr>
<td>7</td>
<td>.23</td>
<td>.22</td>
<td>-.15</td>
<td>.14</td>
<td>.24</td>
<td>.16</td>
<td>-.11</td>
<td>.23</td>
<td>.13</td>
<td>-.13</td>
<td>-.01</td>
<td>-.13</td>
</tr>
<tr>
<td>8</td>
<td>-.23</td>
<td>-.11</td>
<td>-.08</td>
<td>.17</td>
<td>-.13</td>
<td>-.10</td>
<td>.15</td>
<td>-.02</td>
<td>-.03</td>
<td>.10</td>
<td>-.26</td>
<td>.05</td>
</tr>
<tr>
<td>9</td>
<td>.03</td>
<td>-.19</td>
<td>.11</td>
<td>-.12</td>
<td>.06</td>
<td>.18</td>
<td>.16</td>
<td>-.04</td>
<td>-.03</td>
<td>.10</td>
<td>.20</td>
<td>.09</td>
</tr>
<tr>
<td>10</td>
<td>.28</td>
<td>-.17</td>
<td>.06</td>
<td>-.05</td>
<td>-.12</td>
<td>-.12</td>
<td>-.40†</td>
<td>-.08</td>
<td>-.06</td>
<td>-.30†</td>
<td>.10</td>
<td>.45†</td>
</tr>
<tr>
<td>11</td>
<td>.13</td>
<td>-.04</td>
<td>-.06</td>
<td>.11</td>
<td>-.06</td>
<td>-.07</td>
<td>.11</td>
<td>.02</td>
<td>-.01</td>
<td>.07</td>
<td>.20</td>
<td>.00</td>
</tr>
<tr>
<td>12</td>
<td>.15</td>
<td>-.31*</td>
<td>.08</td>
<td>.03</td>
<td>-.03</td>
<td>-.15</td>
<td>-.19</td>
<td>.02</td>
<td>-.06</td>
<td>-.17</td>
<td>.09</td>
<td>.14</td>
</tr>
<tr>
<td>13</td>
<td>.35*</td>
<td>.28</td>
<td>-.43†</td>
<td>.36*</td>
<td>.41†</td>
<td>.14</td>
<td>-.16</td>
<td>.45†</td>
<td>.51†</td>
<td>-.27</td>
<td>-.10</td>
<td>-.02</td>
</tr>
<tr>
<td>14</td>
<td>--</td>
<td>.10</td>
<td>-.13</td>
<td>.16</td>
<td>.31*</td>
<td>.26</td>
<td>.16</td>
<td>.31*</td>
<td>.22</td>
<td>.00</td>
<td>-.08</td>
<td>-.13</td>
</tr>
<tr>
<td>15</td>
<td>.10</td>
<td>--</td>
<td>-.35*</td>
<td>.34*</td>
<td>.37*</td>
<td>.14</td>
<td>.02</td>
<td>.46†</td>
<td>.33*</td>
<td>-.19</td>
<td>-.05</td>
<td>-.44†</td>
</tr>
<tr>
<td>16</td>
<td>-.13</td>
<td>-.35*</td>
<td>--</td>
<td>-</td>
<td>-</td>
<td>.17</td>
<td>-.07</td>
<td>-.84†</td>
<td>-</td>
<td>.09</td>
<td>.20</td>
<td>.14</td>
</tr>
<tr>
<td></td>
<td>.96†</td>
<td>.54†</td>
<td>.715†</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>17</td>
<td>.16</td>
<td>.34*</td>
<td>-.96†</td>
<td>--</td>
<td>.47†</td>
<td>-.13</td>
<td>.1</td>
<td>.82†</td>
<td>.68†</td>
<td>-.05</td>
<td>-.17</td>
<td>-.07</td>
</tr>
</tbody>
</table>
Table 3. *Continued*

<table>
<thead>
<tr>
<th></th>
<th>14</th>
<th>15</th>
<th>16</th>
<th>17</th>
<th>18</th>
<th>19</th>
<th>20</th>
<th>21</th>
<th>22</th>
<th>23</th>
<th>24</th>
<th>25</th>
</tr>
</thead>
<tbody>
<tr>
<td>18</td>
<td>.31*</td>
<td>.37*</td>
<td>-.54†</td>
<td>.47†</td>
<td>--</td>
<td>.75†</td>
<td>.48†</td>
<td>.88†</td>
<td>.74†</td>
<td>.07</td>
<td>-.05</td>
<td>-.43†</td>
</tr>
<tr>
<td>19</td>
<td>.26</td>
<td>.14</td>
<td>.17</td>
<td>-.13</td>
<td>.75†</td>
<td>--</td>
<td>.53†</td>
<td>.43†</td>
<td>.25</td>
<td>.15</td>
<td>.14</td>
<td>-.33*</td>
</tr>
<tr>
<td>20</td>
<td>.16</td>
<td>.02</td>
<td>-.07</td>
<td>.10</td>
<td>.48*</td>
<td>.53†</td>
<td>--</td>
<td>.35*</td>
<td>.28</td>
<td>.37*</td>
<td>.02</td>
<td>-.35*</td>
</tr>
<tr>
<td>21</td>
<td>.31</td>
<td>.46†</td>
<td>-.84†</td>
<td>.82†</td>
<td>.88†</td>
<td>.43†</td>
<td>.35*</td>
<td>--</td>
<td>.86†</td>
<td>.03</td>
<td>-.13</td>
<td>-.32*</td>
</tr>
<tr>
<td>22</td>
<td>.22</td>
<td>.33*</td>
<td>-.72†</td>
<td>.68†</td>
<td>.74†</td>
<td>.25</td>
<td>.28</td>
<td>.86†</td>
<td>--</td>
<td>.00</td>
<td>-.18</td>
<td>-.20</td>
</tr>
<tr>
<td>23</td>
<td>.00</td>
<td>-.19</td>
<td>.09</td>
<td>-.05</td>
<td>.07</td>
<td>.15</td>
<td>.37*</td>
<td>.03</td>
<td>.00</td>
<td>--</td>
<td>.30*</td>
<td>-.07</td>
</tr>
<tr>
<td>24</td>
<td>-.08</td>
<td>-.05</td>
<td>.20</td>
<td>-.17</td>
<td>-.06</td>
<td>.14</td>
<td>.02</td>
<td>-.13</td>
<td>-.18</td>
<td>.30*</td>
<td>--</td>
<td>.24</td>
</tr>
<tr>
<td>25</td>
<td>-.13</td>
<td>-.44†</td>
<td>.14</td>
<td>-.07</td>
<td>-.43†</td>
<td>-.33*</td>
<td>-.35*</td>
<td>-.32*</td>
<td>-.20</td>
<td>-.07</td>
<td>.24</td>
<td>--</td>
</tr>
</tbody>
</table>

*p<.05; † p<.01

prediction of the outcome variable (Cohen & Cohen, 1975). Hierarchical linear regressions examined the addition of the predictor variables of coping (approach and avoidant, total number, and avoidance coping ratio) after controlling for well-sibling demographic factors and illness parameters.

To determine the sample size, G*Power (Erdfelder, Faul, & Buchner, 1996) was used. For all power analyses, alpha level was set as $p = 0.05$ and desired Power as 0.80. The estimate of effect size of $f^2 = 0.15$ was selected to reflect a medium effect size (Cohen, 1992). The analysis with the greatest number of predictors was a hierarchical linear regression that examined an increase from three to five predictors; a sample size of 77 participants is required.
RESULTS

Given the small sample size, post-hoc power analyses and effect sizes were calculated. Observed effect sizes are based on the small sample of the present study; therefore, the a priori estimate of medium effect size was used as these were based on population estimates (Faul, Erdfelder, Lang, & Buncher, 2007). The sample size used in all calculations was 43 participants, and alpha level was set as $p = 0.05$. Post-hoc power analyses were conducted using G*Power (Faul, Erdfelder, Lang, & Buncher, 2007). For effect sizes, Cohen’s d was calculated for regression analyses (Cohen, 1988) and paired samples t-tests (Dunlap, Cortina, Vaslow, & Burke, 1996). Results from the post-hoc power and effect size analyses are displayed in Table 4.

**Hypothesis 1:**

To test the hypothesis that well-sibling demographic factors and illness factors would predict well-sibling quality of life, a linear regression was conducted. Well-sibling self-reported quality of life was the dependent variable, and well-sibling age, well-sibling gender, and severity of illness were predictor variables (hereafter labeled Regression 1a for ease of comparison with other regressions). Well-sibling demographic variables and illness factors significantly predicted well-sibling quality of life ($R^2 = .21$, $F(3,39) = 3.45$, $p < .05$), and the results are displayed in Table 5. Well-sibling age was the only significant predictor of well-sibling quality of life ($\beta = .48$, $p < .01$). Therefore, well-sibling age was used as a control variable in subsequent hierarchical linear regression
Table 4. Results of post-hoc power analyses and achieved effect size for all analyses.

<table>
<thead>
<tr>
<th>Predictor Variables (Number)</th>
<th>Achieved Power</th>
<th>Effect Size</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hypothesis 1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Age, gender, severity of illness (3)</td>
<td>.51</td>
<td>.27</td>
</tr>
<tr>
<td>Age (1)</td>
<td>.70</td>
<td>.25</td>
</tr>
<tr>
<td>Hypothesis 2</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Age, approach coping, avoidance coping (3)</td>
<td>.51</td>
<td>.46</td>
</tr>
<tr>
<td>Age, total number of coping strategies (2)</td>
<td>.58</td>
<td>.53</td>
</tr>
<tr>
<td>Age, avoidant coping ratio (2)</td>
<td>.58</td>
<td>.29</td>
</tr>
<tr>
<td>Age, approach coping ratio (2)</td>
<td>.58</td>
<td>.27</td>
</tr>
<tr>
<td>Hypothesis 3</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Avoidance Helpfulness – Approach Helpfulness</td>
<td>.89</td>
<td>.49</td>
</tr>
<tr>
<td>Hypothesis 4</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Well-sibling anxiety, sadness, anger (3)</td>
<td>.51</td>
<td>.29</td>
</tr>
<tr>
<td>Age, well-sibling anxiety, sadness, anger (4)</td>
<td>.45</td>
<td>.66</td>
</tr>
<tr>
<td>Hypothesis 5</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Parent-report vs. self-report of well-sib adjustment</td>
<td>.89</td>
<td>1.07</td>
</tr>
</tbody>
</table>
Table 5. Results of regression using demographic and illness parameters including and excluding time since diagnosis to predict well-sibling self-report adjustment.

<table>
<thead>
<tr>
<th>Variable</th>
<th>Regression 1a</th>
<th></th>
<th></th>
<th>Regression 1b</th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>B</td>
<td>SE B</td>
<td>β</td>
<td>B</td>
<td>SE B</td>
<td>β</td>
</tr>
<tr>
<td>Well-Sibling Age</td>
<td>2.31</td>
<td>.72</td>
<td>.48**</td>
<td>2.14</td>
<td>.67</td>
<td>.45**</td>
</tr>
<tr>
<td>Well-Sibling Gender</td>
<td>-2.43</td>
<td>4.77</td>
<td>-.07</td>
<td>--</td>
<td>--</td>
<td>--</td>
</tr>
<tr>
<td>Severity of Illness</td>
<td>.11</td>
<td>.19</td>
<td>.082</td>
<td>--</td>
<td>--</td>
<td>--</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th></th>
<th>Regression 1a</th>
<th></th>
<th></th>
<th>Regression 1b</th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>R²</td>
<td>.21</td>
<td></td>
<td></td>
<td>.20</td>
<td></td>
<td></td>
</tr>
<tr>
<td>F</td>
<td>3.45*</td>
<td></td>
<td></td>
<td>10.24*</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

* p < .05, ** p < .01
analyses. Older well-siblings had better quality of life than younger well-siblings, which is the opposite relationship than predicted. The hypotheses that male well-siblings and well-siblings of sick-children with less severe cystic fibrosis would have better quality of life were not supported.

Hypothesis 2

To evaluate the hypothesis that well-sibling coping would predict well-sibling quality of life after controlling for demographic and illness parameters, a hierarchical linear regression was conducted. Self-reported well-sibling quality of life was the outcome variable, well-sibling age was entered as a predictor variable at the first step of the regression, and approach and avoidant styles of coping were entered as predictor variables in the second step (hereafter labeled Regression 2a). Well-sibling age, and avoidant and approach styles of coping significantly predicted well-sibling quality of life ($R^2 = .31, F (3,39) = 5.92, p < .01$), and results are displayed in Table 6. The addition of approach and avoidant coping styles to well-sibling age predicted more variance in well-sibling quality of life than well-sibling age alone ($F_{change} (2, 39) = 3.21, p = .051$). Contrary to the study’s hypothesis, number of approach styles of coping and number of avoidant styles of coping were not individual significant predictors of well-sibling quality of life. Avoidant coping and approach coping were entered in to separate regressions, and results are displayed in Table 7. Avoidant coping ($\beta = -.28, p < .05$) and approach coping ($\beta =-.29, p < .05$) were both significant individual predictors of well-sibling adjustment when entered into separate regressions.
Table 6. Results of regressions using well-sibling age and approach and avoidant coping styles, total coping strategies, and coping ratio of avoidance to total number of coping strategies to predict well-sibling self-report adjustment.

<table>
<thead>
<tr>
<th>Variable</th>
<th>Regression 2 a</th>
<th></th>
<th></th>
<th>Regression 2 b</th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>B</td>
<td>SE B</td>
<td>B</td>
<td>SE B</td>
<td>B</td>
</tr>
<tr>
<td><strong>Step 1</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Well-Sibling Age</td>
<td>1.95</td>
<td>.64</td>
<td>.41**</td>
<td>1.91</td>
<td>.62</td>
<td>.40**</td>
</tr>
<tr>
<td><strong>Step 2</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Approach Coping</td>
<td>-2.84</td>
<td>2.22</td>
<td>-.19</td>
<td>--</td>
<td>--</td>
<td>--</td>
</tr>
<tr>
<td>Avoidant Coping</td>
<td>-2.17</td>
<td>1.51</td>
<td>-.21</td>
<td>--</td>
<td>--</td>
<td>--</td>
</tr>
<tr>
<td>All Coping Styles</td>
<td>--</td>
<td>--</td>
<td>--</td>
<td>-2.33</td>
<td>.78</td>
<td>-.38**</td>
</tr>
<tr>
<td>( R^2 )</td>
<td>.31</td>
<td></td>
<td></td>
<td>.35</td>
<td></td>
<td></td>
</tr>
<tr>
<td>( F )</td>
<td>5.92</td>
<td></td>
<td></td>
<td>10.54**</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

* \( p < .05 \), ** \( p < .01 \)
Table 7. Results of regressions using well-sibling age and number of avoidant or approach coping strategies to predict well-sibling self-reported adjustment.

<table>
<thead>
<tr>
<th>Variable</th>
<th>( B )</th>
<th>( SE ) ( B )</th>
<th>( \beta )</th>
<th>( B )</th>
<th>( SE ) ( B )</th>
<th>( \beta )</th>
</tr>
</thead>
<tbody>
<tr>
<td>Well-sibling age</td>
<td>1.98</td>
<td>.65</td>
<td>3.06**</td>
<td>2.03</td>
<td>.64</td>
<td>.43</td>
</tr>
<tr>
<td>Avoidant coping</td>
<td>-4.20</td>
<td>2.03</td>
<td>-.28*</td>
<td>--</td>
<td>--</td>
<td>--</td>
</tr>
<tr>
<td>Approach coping</td>
<td>--</td>
<td>--</td>
<td>--</td>
<td>-3.00</td>
<td>1.38</td>
<td>-.29*</td>
</tr>
</tbody>
</table>

\( R^2 = .28 \) \hspace{1cm} \( F = 7.66** \) \hspace{1cm} \( F = 7.94** \)

* \( p < .05 \), ** \( p < .01 \)

The hypothesis that total number of coping strategies would predict well-sibling quality of life was examined with a hierarchical linear regression (labeled Regression 2b). Self-reported well-sibling quality of life was the outcome variable. The predictor variable of well-sibling age was entered in the first step and total number of coping strategies used was entered in the second step. Well-sibling age and total number of coping styles used significantly predicted well-sibling quality of life (\( R^2 = .35 \), \( F(2,42) = 10.54, p < .01 \)), and results are displayed in Table 6. Coping strategies and well-sibling age predicted more variance in well-sibling quality of life than well-sibling age alone (\( F \) change \( (1, 40) = 8.88, p < .01 \)). Well-sibling age (\( \beta = .40, p < .01 \)) and total number of coping styles used (\( \beta = -.38, p < .01 \)) were both significant predictors of well-sibling quality of life. Greater number of coping strategies endorsed was associated with poorer well-sibling quality of life, which is the opposite direction of the hypothesized relationship.
To test the hypothesis that the avoidance coping style ratio would predict better well-sibling self-reported quality of life, a hierarchical linear regression was conducted with well-sibling age as a predictor in the first step and the avoidance coping ratio in the second step (labeled Regression 2c). Well-sibling age and the avoidance coping ratio significantly predicted well-sibling quality of life ($R^2 = .22$, $F(2,41) = 5.64$, $p < .01$), and results are displayed in Table 8. The avoidance coping ratio was not a significant individual predictor of well-sibling quality of life, which did not support the study’s hypothesis. Given the poor internal consistency of the avoidance coping style factor and the adequate internal consistency of approach coping style factor, this relative coping ratio was modified to measure approach styles of coping to total number of coping strategies. This approach ratio of coping was entered in the second step of a separate hierarchical linear regression to predict well-sibling self-reported quality of life, and well-sibling age was included in the first step ($R^2 = .21$, $F(2,41) = 5.27$, $p < .01$). Similar to the avoidance coping ratio, the approach coping ratio was also not a significant predictor of well-sibling quality of life, and results are displayed in Table 8.

The percentages of coping strategies endorsed by well-siblings are displayed in Table 9. Well-siblings used an average of 3.58 approach styles of coping (minimum of 0 and maximum of 6), and an average of 3.65 avoidant styles of coping (minimum of 0 and maximum of 6). Well-siblings endorsed using an average of 8.56 total coping strategies (minimum of 0 and maximum of 13). Very few participants endorsed “Blaming Others” as a coping strategy. “Blaming Others” was removed from the avoidance factor structure,
Table 8. Results of regressions using well-sibling age and avoidant or approach coping ratio to predict well-sibling self-reported adjustment.

<table>
<thead>
<tr>
<th>Variable</th>
<th>B</th>
<th>SE B</th>
<th>β</th>
<th>B</th>
<th>SE B</th>
<th>β</th>
</tr>
</thead>
<tbody>
<tr>
<td>Well-sibling age</td>
<td>2.11</td>
<td>.66</td>
<td>.45**</td>
<td>2.14</td>
<td>.66</td>
<td>.46**</td>
</tr>
<tr>
<td>Avoidant ratio</td>
<td>14.15</td>
<td>16.72</td>
<td>.12</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Approach ratio</td>
<td>-.73</td>
<td>19.38</td>
<td>-.053</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

\[ R^2 \quad .22 \quad .21 \]

\[ F \quad 5.64** \quad 5.27** \]

* p < .05, ** p < .01
Table 9. Percentages of coping strategies endorsed by well-siblings

<table>
<thead>
<tr>
<th>Coping Strategy</th>
<th>Percentage</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Avoidance</strong></td>
<td></td>
</tr>
<tr>
<td>Distraction (1,2)</td>
<td>86.0%</td>
</tr>
<tr>
<td>Blaming Others (7)</td>
<td>4.7%</td>
</tr>
<tr>
<td>Wishful Thinking (12,13)</td>
<td>88.4%</td>
</tr>
<tr>
<td>Resignation (15)</td>
<td>60.5%</td>
</tr>
<tr>
<td><strong>Approach</strong></td>
<td></td>
</tr>
<tr>
<td>Cognitive Restructuring (5)</td>
<td>86.0%</td>
</tr>
<tr>
<td>Problem Solving (8,9)</td>
<td>65.1%</td>
</tr>
<tr>
<td>Emotional Expression (10,11)</td>
<td>74.4%</td>
</tr>
<tr>
<td>Social Support (14)</td>
<td>88.4%</td>
</tr>
</tbody>
</table>

and internal reliability was reexamined; however, reliability analysis still showed very poor internal consistency (changed from .18 with “Blaming Others” to .16 without it).

**Hypothesis 3**

To evaluate whether approach styles of coping were rated as more helpful than avoidant styles of coping, a paired-samples t-test was conducted. On average, well-siblings rated approach styles of coping ($M = 6.07, SE = 2.50$) as more helpful than avoidance styles of coping ($M = 4.74, SE = 1.93$), ($t(40) = -3.06$, $p < .01$).

**Hypothesis 4**
To test that hypothesis that well-sibling subjective reports of distress would predict well-sibling self-reported quality of life, a hierarchical linear regression was conducted with well-sibling anxiety, sadness, and anger about the illness as predictor variables; well-sibling age was entered in the first step as a control variable. Well-sibling distress significantly predicted well-sibling quality of life \( R^2 = .39, F(4,39) = 6.22, p < .01 \), and Table 10 contains the results of this regression. Anger \( (\beta = -.38, p < .01) \) and sadness \( (\beta = -.26, p < .05) \) were both significant predictors of well-sibling quality of life, such that well-siblings who were less angry and sad about the cystic fibrosis reported better quality of life. The hypothesis that greater anxiety, sadness, and anger would predict poorer well-sibling adjustment was mostly supported, as anger and sadness were both significant individual predictors.

**Hypothesis 5:**

To test the hypothesis that well-sibling reports of distress would be significantly correlated with severity of illness, Pearson coefficients were examined. There were no significant correlations between severity of illness and well-siblings’ anxiety, sadness, or anger (see Table 3).

**Hypothesis 6:**

A paired samples t-test was conducted to evaluate the hypothesis that well-sibling self-report of quality of life would be significantly different from parent-report. Parent-report of well-sibling quality of life \( (M = 89.97, SE = 1.45) \) was significantly better than well-sibling self-report \( (M = 71.48, SE = 2.54) \) \( t(42) = -7.08, p < .01 \), which was the opposite of the relationship predicted by the study. Using Varni and colleagues (2003)
Table 10. Results of regression using well-sibling report distress to predict well-sibling self-reported adjustment after controlling for well-sibling age.

<table>
<thead>
<tr>
<th>Variable</th>
<th>$B$</th>
<th>$SE\ B$</th>
<th>$\beta$</th>
</tr>
</thead>
<tbody>
<tr>
<td>Well-Sibling Age</td>
<td>2.13</td>
<td>8.11</td>
<td>.44**</td>
</tr>
<tr>
<td>Nervous about CF</td>
<td>1.78</td>
<td>1.80</td>
<td>.14</td>
</tr>
<tr>
<td>Sad about CF</td>
<td>-3.28</td>
<td>1.76</td>
<td>-.26*</td>
</tr>
<tr>
<td>Mad about CF</td>
<td>-7.66</td>
<td>2.72</td>
<td>-.38**</td>
</tr>
</tbody>
</table>

$R^2$ .39

$F$ 6.22**

* $p < .05$, ** $p < .01$

cut-off scores of 71.44 for self-report, the present study identified 19 (44.2%) out of 43 well-siblings as at-risk for impaired quality of life. Using parent-report and the cut-off score of 67.44, only 3 of 43 (6.9%) of well-siblings were identified as at-risk. The percentages of well-siblings classified as at-risk of quality of life impairments by self-report and parent-report were significantly different ($\chi^2(1, N = 43) = 15.63, \ p < .001$).

Qualitative Comments

Though most children did not expand or elaborate their answers beyond the suggested item responses, those that did provided additional insight into their emotions about living with the sick-child with cystic fibrosis. These comments are included here to give additional clarification or understanding of the basic Likert-style responses of distress ratings. One eight-year-old girl said that she wished that she could make things
different for her sister. She said, “My heart breaks into little pieces when I hear my sister
coughing.” A twelve-year-old girl said that she felt “somewhat nervous” about having a
sibling with cystic fibrosis because “I don’t know what’s happening [with the sick-child’s
cystic fibrosis].” Another seven-year-old girl discussed the parental attention shift that
occurred with the sick-child’s illness; she said that having a sister with cystic fibrosis
made her “a little mad because she takes a lot of attention.” This same girl also discussed
a sense of responsibility in trying to help her family adjust to this illness, and said, “I try
to calm my family down because they’re always stressed about what you have to do
[about the sick-child’s illness and treatments].”
DISCUSSION

Despite the pervasive effect of childhood chronic illness on all family members, there is a relative dearth of information on the experience of well-siblings. Those previously published empirical studies yielded mixed results with regard to the relationship between well-sibling adjustment, demographic factors, and illness parameters, and to the overall impact of the chronic illness on well-sibling adjustment. The present study found that almost half of the well-siblings assessed were at-risk for impaired quality of life. Past studies found well-sibling adjustment, as measured by psychopathology, was usually within normal limits (e.g., Labay & Walco, 2004; Taylor et al., 2001). Previous research has demonstrated well-sibling impairments extend beyond symptoms of psychopathology to areas of physical functioning (Buchbinder et al., 2010; Williams et al., 2009), social functioning (Houtzager et al., 2004) and emotional functioning (Gold et al., 2008b). Rather than limiting the focus to internalizing and externalizing symptoms as previous studies have done, the present study included other domains of functioning in an assessment of quality of life. Inclusion of many domains of functioning may account for the larger proportion of well-siblings identified as significantly impacted by chronic illness in the present study. Well-siblings in previous studies were usually not impaired in internalizing and externalizing symptoms and consequently were not identified as at-risk, but when areas
of adjustment were expanded to domains such as physical and social functioning, more impairment was evident.

Parents’ reports of well-siblings’ adjustment were significantly different than self-reported quality of life. Many previous studies relied on parent-report of well-sibling’s adjustment, and thus these previous studies may have underestimated the impact of chronic illness on the well-sibling’s adjustment (e.g., Gold et al., 2008a; Jackson et al., 2008). Parents rated well-siblings’ quality of life as significantly better than well-siblings’ report, contrary to the present study’s hypothesis. There are several possible explanations for higher parent-report ratings of well-sibling’s quality of life compared to self-report. Parents caring for a child with cystic fibrosis may be more preoccupied with caring for the sick-child, and consequently may not be as sensitive to the well-sibling’s functioning (Williams et al., 2009). Well-siblings may avoid communicating concerns about the illness with their parents to protect their parents from experiencing more distress (Grootenhuis & Last, 2006). Well-siblings may also purposefully hide their problems from their parents in social, emotional, and physical functioning to protect them from additional burden (Houtzager et al., 2005). This study highlights the importance of obtaining well-sibling self-reported adjustment because if a parent does not perceive quality of life impairments, they are not likely to obtain treatment for the well-sibling (Varni et al., 2001). These results also suggest that interventions aimed at increasing parental awareness of well-sibling quality of life may be important. Previous research found that greater maternal awareness of the well-sibling’s attitudes and perceptions about the sick-child’s illness predicted better well-
sibling adjustment (Taylor et al., 2001). Future research should continue to collect data directly from well-siblings for a more accurate understanding of adjustment, rather than rely solely on parent report.

The present study found that older well-siblings had better self-reported quality of life than younger well-siblings, which was the opposite of the hypothesized relationship. Though the current study found older well-siblings of sick-children with cystic fibrosis had better quality of life, previous research found that older well-siblings had poorer adjustment in other disease populations including cancer, sickle cell disease, and diabetes (e.g., Houtzager et al, 2004; Jackson et al., 2008). Time since diagnosis was also hypothesized to predict well-sibling adjustment based on past research in well-sibling adaptation to cancer (Houtazger et al., 2004), but this hypothesis was not supported in the present study’s population of cystic fibrosis patients. These discrepancies offer support for a categorical approach, as it appears that specific diseases may create unique experiences for the well-sibling which ultimately affects adjustment. For example, sick-children with cancer spend more time in the hospital than sick-children with cystic fibrosis and, consequently, parents of a sick-child with cancer may be required to spend more time away from the home (Chabra & Chávez, 2000). Older well-siblings of sick-children with cancer are required to spend more time in domestic chores that cause greater daily disruption, and this may be related to the time parents spend out of the home (Houtzager et al., 2004). However, cystic fibrosis is usually diagnosed in infancy, thus limiting time since diagnosis variability as compared to cancer that can be diagnosed at various ages of childhood (Hudson, Mulrooney,
In pediatric cancer, it is important to assess the amount of time since diagnosis, as it offers more unique about how much time well-siblings have to adjust to the diagnosis than well-sibling age alone. Well-sibling age is a more important construct in assessing well-sibling adjustment to cystic fibrosis because diagnosis is usually made in infancy. These discrepancies between well-sibling age and time since diagnosis in well-sibling adjustment offer support for the categorical approach as specific diseases may create unique experiences for the well-sibling, which ultimately impacts adjustment.

Contrary to the present study’s hypotheses, well-sibling gender was not predictive of well-sibling adjustment. Differences in boys’ and girls’ adjustment in previous studies may be attributed to the outcome measures used that assessed internalizing and externalizing symptoms. These discrepancies, then, may be related to gender disparities in presentation of psychopathology symptoms rather than differences in adjustment to chronic illness (e.g., Barlow & Ellard, 2006; Ramtekkar et al., 2010). There are no normative significant differences in quality of life ratings by gender (Varni et al., 2001). The data from the present study suggest that well-sibling boys and girls adjust in similar ways to the sick-child’s cystic fibrosis.

The present study did not find the hypothesized relationship between severity of illness and well-sibling adjustment. The discrepancy between the present study and previous studies that did find severity of illness important in predicting well-sibling adjustment may be related to methodology (Gold et al., 2008a; Read et al., 2010). To assess severity of illness, the present study used a quality of life measure of the sick-
child (Varni et al., 2001). Though quality of life is a valid instrument that has utility in assessing disease severity (Varni et al., 1999), parents complete this measure and well-siblings may not understand or rate severity of illness in the same way as parents. Though the present study did not include a rating of severity of illness completed by the well-sibling, well-siblings did rate distress about the sick-child’s illness. These distress ratings were not significantly correlated with more severe cystic fibrosis as rated by parents, which indicates that well-siblings may not be aware of the disease severity of the sick-child. Qualitative data from the present study indicated that some well-siblings did not “know what is happening” with the sick-child, and lack of knowledge about the sibling’s illness was associated with well-sibling subjective distress, particularly anxiety. Assessing illness severity may be better understood with an accurate appraisal of well-sibling knowledge of the illness. Knowledge of illness is particularly important in well-sibling adjustment as it can be improved though psychoeducation (Labato & Kao, 2002). Previous research found that providing the well-sibling with information about the sick-child’s illness significantly reduced emotional distress about the sick-child’s hospitalization (Gursky, 2007). Future research should incorporate a measure of well-sibling knowledge about the illness to better understand its impact on well-sibling adjustment. Well-sibling anger and sadness about cystic fibrosis predicted quality of life. Well-sibling distress should be assessed, and future intervention studies should determine whether reduction of distress about the cystic fibrosis improves well-sibling adjustment.
Other methodological differences between the present study and previous studies may explain discrepant results about severity of illness as a predictor of well-sibling adjustment. Previous research used different assessments of severity of illness, such as number of trips to the emergency room, number of nights the sick-child spent in the hospital, greater extent of wheelchair use for the sick-child, and total number of hospitalizations (e.g., Gold et al., 2008b; Gold et al., 2011; Read et al., 2010). These measures of illness severity are more concrete and, as such, likely have a greater and more tangible impact on well-siblings’ daily lives than an assessment of illness severity completed by parents. Future research should incorporate well-siblings’ self-appraisals of the severity of illness in predicting well-sibling adjustment, as well as other more objective measures of severity of illness such as recent number of days in the hospital.

The present study is one of few quantitative studies to examine the relationship between coping and well-sibling adjustment. The addition of approach and avoidance styles of coping or the avoidance coping ratio accounted for more variance in predicting well-sibling quality of life than well-sibling age alone. However, contrary to the study’s hypotheses, these coping variables were not significant individual predictors of well-sibling adjustment. Modifying this relative coping ratio to reflect number of approach styles of coping to total number of coping strategies was also not significant in predicting well-sibling quality of life. These data are consistent with Sloper and While’s (1999) study with well-siblings’ adjustment to cancer. Well-siblings rated approach styles of coping as more helpful than avoidant styles of coping. These data suggest that well-siblings value approach styles of coping more than avoidant styles,
despite the lack of significant difference between the two coping styles in predicting adjustment.

The relatively small sample size of the present study may not have been large enough to accurately test the unique contribution of approach and avoidant coping styles. These coping styles were significantly correlated predictor variables, thus make interpreting findings difficult (Mason & Perreault, 1991); this interpretation is supported with the data that avoidant and approach styles of coping were both significant individual predictors when entered in to separate regressions. Avoidant styles of coping and the avoidance coping ratio may not be significant individual predictors as a result of measurement error given the poor reliability of the avoidant style of coping factor. Interpretations using the two-factor structure of the Kidcope for well-siblings should be made cautiously given the poor internal reliability for the avoidance factor. The lack of data on the factor structure of the Kidcope for well-siblings is a limitation of the present study. Future research with a larger population should examine the factor structure of this coping instrument with well-siblings.

Well-siblings who used a greater number of total coping strategies had poorer adjustment. Though these data did not support this study’s hypothesis, past research with sick-children coping with their own diagnosis of cancer found that fewer, more frequently used coping strategies were more effective in improving adjustment than using many different strategies (Worchel, Copeland, & Barker, 1987). Also, children who were diagnosed with posttraumatic stress disorder (PTSD) following a car accident endorsed a greater number of coping strategies than those in a car accident without
PTSD (Stallard, Velleman, Langsford, & Baldwin, 2001). These data from other populations suggest that well-siblings who were experiencing more difficulty in adjusting to the sick-child’s illness may have attempted many coping strategies in an effort to find the most effective strategy. Well-siblings who are better adjusted have likely identified a few particularly helpful coping strategies, and therefore do not need to try additional coping strategies. Future studies should address limitations of the present study by including additional information about well-sibling coping, such as an interview with the well-siblings about their coping strategies. A structured or semi-structured interview would allow for more interpretations about the complexities of coping, including frequency of use and ways in which the well-sibling selects strategies for coping with a sick-child’s illness compared to coping with other life stressors (such as test-taking or fighting with a friend). Additionally, future studies should collect longitudinal data to determine how, if at all, well-siblings change their strategies for coping with chronic illness over time.

One limitation of the present study is the sample size, which was smaller than that proposed from a priori power analyses due to difficulty with recruiting participants in a small clinical population. Small samples limit statistical power and may lead to Type II error or falsely rejecting a true hypothesis (Freiman, Chalmers, Smither, & Kuebler, 1978). Post hoc power analyses confirmed that power was limited in the present study. Effect size was calculated in the present study as these statistics offers information about the associations between variables, and is not greatly influenced by sample size (Ferguson, 2009). Using Cohen’s (1988) size conventions, regression and t-
test analyses of the present study yielded medium to large effect sizes. These effect size data demonstrate that the present study’s findings are robust and meaningful. With a larger sample size, it is possible that some of the variables entered into regressions would emerge as significant individual predictors of well-sibling adjustment. Therefore, though the present study eliminated non-significant predictors, future studies should include all variables tested to determine if these variables emerge as significant with a larger sample size. The small sample size may have also limited the variability in the sample population. Missing data analyses revealed that African-American well-siblings were less likely to have complete data. Given confounds between ethnicity and socioeconomic status (e.g., Kaufman, Cooper, & McGee, 1997), these participants may have been lost to follow up due to limited resources. The two African-American well-sibling participants in the study both had Medicaid insurance and limited parental education and, therefore, socioeconomic status may explain the difficulty in reaching the participants by phone for follow-up.

Older well-siblings of sick-children with cystic fibrosis and those who used fewer coping strategies reported better self-reported quality of life. This study highlighted the importance of future studies examining factors that influence well-sibling adjustment, such as well-sibling rating of the severity of illness, and well-sibling knowledge of the illness. Finally, in addition to investigating demographic and illness factors that may help explain and predict well-sibling adjustment, future studies should incorporate these factors in interventions to improve well-sibling adjustment. Though studies have investigated an intervention for well-siblings to improve well-sibling
quality of life (Besier, Hölling, Schlack, West, & Goldbeck, 2010; Packman, Chesterman, vanZutphen, Golan, & Amylon, 2005) and knowledge of the illness (Sidu, Passmore, & Baker, 2006; Williams, Williams, Graff, Hanson, Stanton, Hafeman, Liebergen, Leuenberg, Setter, Ridder, Curry, Barnard, & Sanders, 2003), future interventions should incorporate curriculum aimed at improving well-sibling coping styles. Parents should also be included in the interventions to promote better communication between the well-sibling and the parent. This increased communication may decreased the discrepancies between parent-report and self-report of well-sibling quality of life. The present study underscores the impact of chronic illness on the quality of life of the well-sibling, and future research should be devoted to better understanding and helping these children.
REFERENCES


Besier, T., Hölling, H., Schlack, R., West, C., Goldbeck, L. (2010). Impact of a family-


disease-related stressors by chronically ill children and adolescents. *Journal of the

for use with pediatric populations. *Journal of Pediatric Psychology, 13*, 555-574.

Heights, MA: Allyn & Bacon.

Tamres, L.K., Janicki, D., & Helgeson, V.S. (2002). Sex differences in coping behavior:
A meta-analytic review and an examination of relative coping. *Personality and

chronic physical disorder in a sibling: How important is maternal awareness of
their illness attitudes and perceptions? *Journal of Child Psychology and
Psychiatry, 42*(7), 953-962.


of well siblings of chronically ill children. *Children’s Health Care, 23*(3), 211-
226.


Youngstrom, E., Loeber, R., & Southamer-Loeber, M. (2000). Patterns and correlates of
APPENDIX A

Date:__/__/____

Answer the following questions (1-4) about your child with cystic fibrosis (CF):

1. Age of child _____
2. Child’s gender: ☐ Female ☐ Males
3. Date child was diagnosed with CF (month and year): __/__
4. How would you describe the racial/ethnic background of your child with CF?
   (Check all that apply)
   ☐ African-American ☐ Asian/Pacific Islander ☐ Caucasian
   ☐ Latino/Hispanic ☐ Other (please specify) _______________

Answer the following questions (5-8) about your child without CF (sibling of child with CF): Note: If your child with CF has more than one sibling without CF, please complete the questions about the sibling closest in age to your child with CF

5. Age of sibling _____
6. Sibling’s gender: ☐ Female ☐ Males
7. How would you describe the racial/ethnic background of your child with CF?
   (Check all that apply)
   ☐ African-American ☐ Asian/Pacific Islander ☐ Caucasian
   ☐ Latino/Hispanic ☐ Other (please specify) _______________
8. What is the type of medical insurance coverage of your child without CF?
   ☐ Medicaid ☐ Medicaid-HMO
   ☐ Private Insurance ☐ Self Pay

Answer the following questions (9-13) about yourself:

9. Your age _____
10. Your gender: ☐ Female ☐ Male
11. Are you a single parent? ☐ Yes ☐ No
12. What is the highest level of education that you have completed?
   ☐ Some High School ☐ High School or GED
   ☐ Some College/Professional School ☐ Bachelor/Associates/Professional Degree
   ☐ Some Graduate School ☐ Graduate or Advanced Degree
13. What is your current relationship to the child without CF?

- [ ] Biological Mother
- [ ] Biological Father
- [ ] Step-Mother
- [ ] Step-Father
- [ ] Adoptive Mother
- [ ] Adoptive Father
- [ ] Other Relative
- [ ] Other (please specify) __________
**APPENDIX B**

**Instructions:** We are trying to find out how children deal with having a brother or sister with cystic fibrosis. Please think about the way you deal with having a brother or sister with cystic fibrosis and answer the questions below:

1. Did this situation make you nervous?
   - Not at all
   - A little
   - Somewhat
   - Pretty much
   - Very much

2. Did this situation make you sad?
   - Not at all
   - A little
   - Somewhat
   - Pretty much
   - Very much

3. Did this situation make you angry or mad?
   - Not at all
   - A little
   - Somewhat
   - Pretty much
   - Very much

Now, please circle whether you used any of the following ways to deal with having a sibling with cystic fibrosis:

<table>
<thead>
<tr>
<th>Did You Do This?</th>
<th>How Much Did It Help?</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Not at all</td>
</tr>
<tr>
<td>1. I just tried to forget it</td>
<td>Yes</td>
</tr>
<tr>
<td>2. I did something like watch TV or played a game to forget it</td>
<td>Yes</td>
</tr>
<tr>
<td>3. I stayed by myself.</td>
<td>Yes</td>
</tr>
<tr>
<td>4. I kept quiet about the problem.</td>
<td>Yes</td>
</tr>
<tr>
<td>5. I tried to see the good side of things.</td>
<td>Yes</td>
</tr>
<tr>
<td>6. I blamed myself for causing the problem.</td>
<td>Yes</td>
</tr>
<tr>
<td>7. I blamed someone else for causing the problem.</td>
<td>Yes</td>
</tr>
<tr>
<td>8. I tried to fix the problem by thinking of answers.</td>
<td>Yes</td>
</tr>
<tr>
<td>9. I tried to fix the problem by doing something or talking to someone.</td>
<td>Yes</td>
</tr>
<tr>
<td>10. I yelled, screamed, or got mad.</td>
<td>Yes</td>
</tr>
<tr>
<td>11. I tried to calm myself down.</td>
<td>Yes</td>
</tr>
<tr>
<td>Question</td>
<td>Yes</td>
</tr>
<tr>
<td>-------------------------------------------------------------------------</td>
<td>-----</td>
</tr>
<tr>
<td>12. I wished the problem had never happened.</td>
<td></td>
</tr>
<tr>
<td>13. I wished I could make things different.</td>
<td></td>
</tr>
<tr>
<td>14. I tried to feel better by spending time with others like family, grownups, or friends.</td>
<td></td>
</tr>
<tr>
<td>15. I didn’t do anything because the problem couldn’t be fixed.</td>
<td></td>
</tr>
</tbody>
</table>
APPENDIX C

PedQL™ Parent Report

There are 23 items in total that measure physical, emotional, social, and school functioning on the PedQL™ Parent Report. The full instrument is available from:

MAPI Research Trust
27, rue de la Villette
69003 Lyon
France
APPENDIX D

*PedsQL™ Child Report*

There are 23 items in total that measure physical, emotional, social, and school functioning on the PedsQL™ Child Report. The full instrument is available from:

MAPI Research Trust  
27, rue de la Villette  
69003 Lyon  
France