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Parenting-Related Stress, Parental Distress, and Youth Health-Related Quality of Life in Families of Youth with Spina Bifida: Parenting Behaviors As Mediators

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LOYOLA UNIVERSITY CHICAGO

PARENTING-RELATED STRESS, PARENTAL DISTRESS, AND YOUTH HEALTH-RELATED QUALITY OF LIFE IN FAMILIES OF YOUTH WITH SPINA BIFIDA: PARENTING BEHAVIORS AS MEDIATORS

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PROGRAM IN CLINICAL PSYCHOLOGY

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CHAPTER ONE

INTRODUCTION

With advances in medical care, individuals with spina bifida (SB) are now expected to live well into adulthood (Oakeshott, Hunt, Poulton, & Reid, 2010), but they are also susceptible to many potentially life-threatening secondary health conditions, including pressure sores and urinary tract infections. As such, individuals with SB are required to maintain an extensive, complex medical regimen, and adherence to this regimen is critical in preventing medical complications. Given the complexity of this condition, it is not surprising that past research has shown that youth with spina bifida (SB) have poorer psychosocial outcomes compared to typically developing (TD) youth (Ammerman et al., 1998, Holmbeck & Devine, 2010, Holmbeck et al., 2003, 2010, Murray et al., 2015) and poorer quality of life than both typically developing youth and youth with other chronic illnesses (Murray et al., 2015). Studies have also found that many demographic (i.e., SES) and illness-severity (e.g., number of shunt revisions) factors affect quality of life, but modifiable factors affecting quality of life have not yet been identified in this population. Quality of life is an especially important factor for youth with SB as past research has implicated quality of life as an important predictor of health outcomes, including adherence (Rodis & Kibbe, 2010; Loon, Jin, & Jin, 2015; Martinez, Prado-Aguilar, Rascon-Pacheco, & Valdivia-Martinez, 2008).

Parents of youth with chronic illnesses, including SB, are at risk for increased stress and poorer psychosocial functioning (Holmbeck et al., 1997; Wallander, Pitt, & Mellins, 1990). Parents of these children may have difficulty managing all of their responsibilities, including caring for their child and, thus, may experience more stress, worry, and depressive symptoms (Greenley, Holmbeck, & Rose, 2006; Holmbeck et al., 2003; Kelly et al., 2008; Singh, 2003). If parents experience distress and stress, this may negatively impact their child's level of psychosocial adjustment, including quality of life. In fact, in some pediatric populations (e.g., cerebral palsy, diabetes, epilepsy), parent factors have been found to impact QOL above and beyond illness severity (Bolghan-Abadi, Kimiaee, & Amir, 2011; Aran, Shaley, Biran, & Gross-Tsur, 2007). Though more research is needed to assess the impact of parent factors on quality of life, it is possible that this impact may be especially salient in youth with SB, as youth with SB depend on parents for both medical and non-medical caregiving needs.

A review of the current literature reveals a lack of understanding of the parent factors and behaviors that may impact quality of life in youth with SB. The current study seeks to address these gaps by testing longitudinal, multi-method, and multi-informant models of these individual and family factors. The following sections provide an overview of the current research on parent functioning in relation to youth quality of life in families of youth with SB and how parenting behaviors may mediate the relationship between parent and youth functioning. Weaknesses and gaps in the current literature are identified, and a detailed description of the current study is provided.

CHAPTER TWO

REVIEW OF THE RELEVANT LITERATURE

Health-related Quality of Life in Youth with Spina Bifida

Spina bifida (SB) is a relatively common congenital birth defect that occurs in approximately 3 of every 10,000 live births in the United States (Centers for Disease Control and Prevention, 2011). SB occurs in the early weeks of gestation, when the neural tube fails to close completely. SB is associated with a number of complications, including paralyzed lower extremities, urinary and bowel dysfunction, and hydrocephalus (Copp, Adzick, Chitty, Fletcher, Holmbeck, & Shaw, 2015). Additional medical and non-medical difficulties associated with SB include motor, orthopedic, sensory, neurocognitive, self-care, and social issues (Zukerman, Devine, & Holmbeck, 2011). The severity of SB varies, and these variations are partly dependent on the level of the spinal lesion and neurological complications, such as the number of shunt infections and revisions (Copp et al., 2015). Given the risk for secondary medical conditions and complications, individuals with SB often follow an extensive medical regimen, including medications, catheterization, bowel programs, skin checks, and shunt monitoring (Zukerman, et al., 2011). Advances in medical care have increased the life expectancy of individuals with SB, and many live well into adulthood (Oakeshott, et al., 2010). Adherence to these prescribed tasks is critical as these individuals seek to maintain their health in early adulthood.

The World Health Organization (WHO) defines quality of life as “an individual’s perception of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, and concerns” (The WHOQOL Group, 1995). Health-related quality of life (HRQOL), specifically, focuses on how individuals with a chronic health condition perceive the impact of their condition on their physical and psychological functioning (Walters, Hays, Spritzer, Fridman, & Carter, 2002). Interest in HRQOL has increased recently due to the increasing number of individuals with chronic illnesses who live longer. HRQOL has also been implicated as an important predictor of health outcomes, including adherence (Rodis & Kibbe, 2010; Loon et al., 2015; Martinez et al., 2008).

Several recent studies have assessed HRQOL in youth with SB. Children and adolescents with SB have been found to have significantly lower HRQOL than both typically developing youth (Murray et al., 2015; Zegers et al., 2015) and youth with other chronic health conditions (e.g., asthma, ADHD, depression, diabetes; Murray et al., 2015). A review of the literature revealed three longitudinal studies assessing HRQOL in youth with SB (Parekh et al., 2006; Bellin et al., 2013; Murray et al., 2015), and each of these studies found that, though overall, HRQOL remains stable within the population, there may be variations within subjects (i.e., for some individuals, HRQOL increases over time, while for others it decreases). This variability of course highlights the importance of identifying factors that may affect HRQOL in individuals with SB.

Research has shown that HRQOL in individuals with SB is affected by factors such as age, gender, socioeconomic status (SES), and severity of medical issues (e.g., number of operations, bladder incontinence, mobility impairment, and pain; Murray et al., 2015; Sawin & Bellin, 2010). While these factors are important to consider, they are also non-modifiable demographic factors or difficult-to-modify illness-specific factors. Given the likelihood that individuals with SB will have poor quality of life and the role that HRQOL seems to play in medical adherence, determining modifiable factors that influence HRQOL is an important step in developing appropriate interventions to improve functioning for individuals with SB. One such modifiable factor is the family environment. Familial correlates of HRQOL have not been studied as frequently as have demographic and individual factors.

Parent Adjustment in the Families of Youth with Spina Bifida

Parent characteristics have a major influence on all (typically developing [TD] or non-TD) children's well-being and adjustment. For example, both maternal and paternal depression have been found to be associated with more child problems, such as behavioral issues and internalizing symptoms (Ringoot et al., 2015). In the context of pediatric chronic illness, research has consistently demonstrated that parenting characteristics can have wide-ranging effects on child and family system adjustment (Thompson & Gustafson, 1996). In fact, in some pediatric populations (e.g., cerebral palsy, diabetes, epilepsy), parent factors have been found to impact QOL above and beyond illness severity (Bolghan-Abadi, Kimiaee, & Amir, 2011; Aran, Shaley, Biran, & Gross-Tsur, 2007).

Parents of youth with chronic health conditions face unique challenges, including the management of a child's medical regimen, stress related to the child's health status, and uncertainty regarding the child's current and future independence (Mullins et al., 2007). The clinical symptoms of SB place considerable physical, psychological, and social demands on both individuals with SB and their families (Greenley et al., 2006; Holmbeck et al., 2003; Kelly et al., 2008; Singh, 2003). Family relationships are particularly important for youth with SB, since these youth tend to be more socially isolated from their peers than TD children (Holmbeck et al., 2003). Additionally, the majority of youth with SB complete multiple daily medical routines with at least some assistance from a parent or other caretaker (Copp et al., 2015). Thus, youth with SB are especially reliant on their parents, and, therefore, may be more affected by parent adjustment and behaviors than TD youth. Three distinct parent adjustment factors will be examined further due to their potential impact on HRQOL in youth with SB – parent distress, parenting stress, and SB-specific parenting stress.

Parent Distress

Parent distress includes personal distress experienced by a person (who is also a parent). Parent distress is operationalized as the parent's psychosocial functioning or degree of extreme anxiety, sorrow, or pain an individual experiences. Global psychological distress includes internalizing symptoms such as depression, anxiety, and somatic symptoms (Friedman et al., 2004). Compared to parents of TD children, parents of youth with chronic health conditions have been found to report higher levels of distress (Power & Franck, 2008). Specifically, parents of children living with a chronic illness have reported elevated rates of depression, anxiety, and post-traumatic stress symptoms (Breslau, Staruch, & Mortimer, 1982; Hauenstein, 1990; Quittner & DiGirolamo, 1998).

Some parents of children with SB have been found to experience clinical levels of global psychological distress (e.g., depressive symptoms, anxiety, somatic complaints; Holmbeck et al., 1997). A meta-analysis of 15 studies revealed a consistent negative impact of SB on the psychological adjustment of parents (Vermaes, Janssens, Bosman, & Gerris, 2005). Parents of youth with SB are at risk for feeling more isolated, being less adaptable to change, and holding less optimistic views about the future. Demographic factors that contribute to the experience of anxiety and depression in the parents of youth with SB include caregiver and child age and employment status/income (Malm-Buatsi, et al., 2015). Illness-specific factors, such as lesion level (a proxy for illness severity) and shunt status, have also been found to be associated with the experience of anxiety and depression in these parents (Malm-Buatsi et al., 2015; Grosse et al., 2009). It possible that the burden of monitoring for shunt malfunctions – a task that requires input from the child and is without clear physical indicators – may increase parental anxiety. Additionally, caring for a more severely affected child may include increased frequency of doctor's visits and medical responsibilities for parents. These increased responsibilities, as well as the uncertainty of the course of illness, may cause increased parental distress. However, it is unclear how the experience of personal distress by a parent impacts youth adjustment, specifically HRQOL. It is possible that increased parent distress may impact HRQOL in youth with SB. In fact, one study found that maternal psychological distress predicted lower HRQOL in youth with SB (Abad, 2007). Still, more research is necessary to elucidate this relationship.

Parenting Stress

Parents are often faced with balancing many responsibilities, and, therefore, may experience an increased amount of stress (when compared to non-parents). Parenting stress is operationally defined as the mental or emotional strain or pressure an individual experiences as a direct result of being a parent. For example, this includes the stress a caretaker experiences due to enforcing bedtimes, preparing meals, or arranging after-school activities. Studies of mothers of youth with SB have found that more than one-third of these mothers experience clinically significant levels of parenting stress (Kanaheswari, Razak, & Ong, 2011).

Increased stress may be experienced by parents of youth with SB for a number of reasons. While increased stress for parents of youth with SB may be related to disease-related factors (discussed below), there are a number of non-disease-related factors that impact the experience of stress for parents. For example, ethnic minority parents report significantly higher levels of parenting stress due to societal disadvantages such as lower income, single parenthood, and assimilation/acculturation (Nomaguchi & House, 2013). One study comparing parenting stress between mothers of youth with SB and mothers of able-bodied children found that mothers of youth with SB had lower educational levels, were more likely to be single parents, and were more likely to be unemployed (Ong, Norshireen, & Chandran, 2010). These life stressors as well as maternal mental health status and mother-report of child's adaptive skills were proposed to moderate the impact of SB on parenting stress in these individuals (Ong, Norshireen, & Chandran, 2010).

Parenting stress is associated with both parenting behaviors and child adjustment. Parenting stress decreases the quality of the parent-child relationship (Turner et al., 2010) and is predictive of non-optimal social-emotional and cognitive outcomes for children and adolescents (Deater-Deckard, Chen, & El Mallah, 2013). While increased parenting stress has been found to be associated with decreased youth HQOL in other illness populations (e.g., obesity; Frontini, Moreira, & Canavarro, 2016), studies investigating the specific impact of parenting stress on HRQOL for youth with SB are necessary.

SB-specific Parenting Stress

Parenting a child with a longstanding or life-threatening illness is challenging and can have a negative impact on many aspects of the parent’s life. Parents of these children often have difficulty balancing caring for their child with other responsibilities such as work life, social life, managing finances, and other household tasks. As a result, they may feel overwhelmed or incompetent and, therefore, experience more stress and worry (Power & Franck, 2008). Parenting a child with SB has negative effects on parent stress levels. In fact, parents of children with SB appear to experience more stress than parents of TD children (Holmbeck et al., 1997; Wallander et al., 1990). One qualitative study found that parents of youth with SB consistently describe adhering to daily medical regimen as a major challenge in their everyday lives (Sawin, Belling, Roux, Buran, Brei, & Fastenau, 2003). This same study found that balancing independence-dependence needs of youth with SB was a significant daily stressor for these parents. Stress that is a direct result of these disease-related factors can be described as SB-specific parenting stress.

One factor found to significantly impact SB-specific parenting stress experienced by mothers of youth with SB is the need for clean intermittent catheterization (CIC; Kanaheswari, Razak, & Ong, 2011). CIC is a technique used in the management of bladder dysfunction for many individuals with SB, and needs to be performed as often as once ever 4-6 hours each day. Maintaining this schedule, or reminding a child to maintain this schedule, significantly impacts the level of parenting stress experienced by mothers of youth with SB. Another SB-specific factor found to affect parenting stress is ambulatory status of the child (Antiel et al., 2016). Parents of youth with SB who are able to walk independently report lower parenting stress than parents of youth who are wheel-chair bound (Antiel et al., 2016). Mobility and bladder and bowel dysfunction in individuals with SB are ongoing stressors for these individuals and their caregivers.

Spina bifida is a condition that can affect both physical and cognitive functioning, and the severity of deficits in functioning likely impacts parenting stress. One study found that the severity of physical dysfunctions, but not cognitive deficits, was associated with increasing parenting stress in mothers of youth with SB (Vermaes, Janssens, Mullaart, Vinck, & Gerris, 2008). Given the heterogeneity of impairments associated with SB, it is likely the SB-specific parenting stress is not uniform across all parents of youth with SB. However, given the negative impact of general parenting stress on youth outcomes (Turner et al., 2010; Deater-Deckard et al., 2013), it can be hypothesized that increased SB-specific parenting stress may also lead to poorer child outcomes, including HRQOL. Again, more research on SB-specific parenting stress and HRQOL is necessary to fully understand this relationship.

Summary

In summary, parenting a child with a chronic illness (e.g., SB) can profoundly impact parents' personal experiences. Parents of youth with SB are at an increased risk for experiencing personal distress (anxiety, depression, internalizing symptoms) and parenting stress, both general and SB-specific. Youth with SB may be especially affected by these three distinct parent factors due to their increased reliance on their parents as medical caregivers. However, it is unclear how parental distress, parenting stress, and SB-specific parenting stress impact youth adjustment, specifically youth HRQOL.

Parenting Behaviors in Families of Youth with Spina Bifida

Research has shown that parents of youth with SB exhibit differences in parenting behaviors as compared to parents of TD youth. Specifically, mothers of children with SB have been found to display more psychological control than mothers of TD children (Holmbeck, Shapera, & Hommeyer, 2001). Psychological control – psychological methods of controlling a child that prevent the child from developing as an autonomous individual – is associated with negative psychosocial outcomes for all children (Barber, 1996). Additionally, parents of children with SB have been found to exhibit higher levels of intrusiveness and authoritarian parenting than parents of TD youth (Holmbeck et al., 2002; Sawin et al., 2003; Seefeldt et al., 1997; Vermaes et al., 2005).

As would be expected, adaptive parenting behaviors are associated with positive psychosocial adjustment outcomes in youth; similarly, maladaptive parenting behaviors are associated with negative psychosocial adjustment outcomes in youth (Lamb & Lewis, 2010). For example, increases in these psychological control and authoritarian parenting may limit a child’s autonomy (Holmbeck et al., 2002; Sawin et al., 2003; Seefeldt et al., 2007; Vermaes et al., 2005). However, parental acceptance – the degree to which a parent is affectionate, approving, emotionally supportive, and involved – is associated with positive outcomes (Holmbeck et al., 2001). Higher levels of parental acceptance and parental behavioral control – the degree to which the parent makes rules and regulations, sets limits on the child’s activities, and enforces these rules and limits – have been found to be associated with higher levels of medical adherence in youth with SB (O’Hara and Holmbeck, 2012). As in other chronic illness populations (e.g., cerebral palsy, Aran et al., 2007; Tezcan & Simsek, 2013), these behaviors may be linked to less desirable child outcomes, including lower levels of HRQOL, for youth with SB.

A number of factors may impact parenting behaviors. Child-specific factors, such as temperament, affect the behaviors demonstrated by parents (Deater-Deckard et al., 2013). However, factors unrelated to the child, including levels of parent distress, parenting stress, and SB-specific parenting stress may influence parenting behaviors. High emotional distress and high levels of parenting stress may impair parents’ abilities to manage the demands of complex medical treatments, make medical decisions, and, affect the way an individual behaviorally parents their child. For example, personal distress and high levels of parenting stress have been found to decrease parental warmth and provoke harsh, reactive caregiving (Power & Franck, 2008; Deater-Deckard & Scarr, 1996). Parents reporting higher levels of parenting stress are more likely to be authoritarian, harsh, and negative in their interactions with their children (Deater-Deckard & Scarr, 1996). However, empirical studies comparing parents of chronically ill and TD children have demonstrated mixed findings. A meta-analysis of parenting characteristics in the context of chronic illness found that parents of children with chronic illnesses – parents who presumably are under an increased level of stress and are at greater risk for experiencing personal distress – are more caring and accepting, while others have shown the opposite – parents of chronically ill children are less caring, sensitive, and positive (Pinquart, 2013). One recent study found that parents of children with a chronic medical condition (obesity) were more likely to demonstrate permissive parenting, and that this permissive parenting style mediated the relationship between parenting stress and youth HRQOL (Frontini, Moreira, & Canavarro, 2016). Parenting behaviors have potential to play a mediating role between parent factors and youth HRQOL. More research focused on how parent factors influence parenting behaviors and, subsequently, how these behaviors impact child adjustment (including HRQOL) is necessary.

Limitations of the Current Literature

Several methodological issues exist in studies that have been conducted to date. The use of (a) single methods (e.g., questionnaire report only), (b) single reporters (e.g., child-report only), (c) cross-sectional designs, and (d) bivariate analytic strategies are among the most prominent weaknesses of current literature in this area.

Use of multiple methods and reporters has been encouraged within research in general, and research with individuals with SB specifically (Holmbeck et al., 2006). However, studies assessing parent factors that may affect child functioning of youth with SB often include only parent report on both parent and youth factors (e.g., Ong, Norshireen, & Chandran, 2011). The use of a single reporter introduces the limitation of common-method variance. This limitation is especially important to address in this population as stress experienced by parents may bias their perception of their child’s functioning (especially psychological functioning). Therefore, the use of multiple methods (e.g., questionnaires and observational methods) and multiple reporters is imperative when studying the effect of parent functioning on youth functioning.

Additionally, a review of the literature revealed that fathers are rarely included in data collection and/or analyses. Many studies have focused on maternal adjustment to chronic illness, as mothers are often the child’s main caregiver (Thompson & Gustafson, 1996). However, differences may exist between mothers and fathers in their adjustment and coping with chronic illness. There may also be differences in how children respond to mother behaviors versus father behaviors. In fact, the same parenting behaviors (acceptance, behavioral control, psychological control) were found to be associated with different outcomes in youth with SB depending on whether the behavior was exhibited by a mother or father (O’Hara & Holmbeck, 2012). It has been hypothesized that mothers may experience more psychological distress than fathers, given their higher exposure to illness-related situations in the role of primary caregiver (Vermaes, 2005). However, more studies including fathers of youth with SB will need to be conducted to test this hypothesis.

Research with youth and families is improved when grounded in a developmental framework. One way to establish a developmental framework is to examine these processes over time using longitudinal data. Much of the literature concerning parent mental health and stress and child outcomes to date is cross-sectional (e.g., Malm-Buatsi et al., 2015). Though significant relationships have been found between parent stress and mental health concerns and child outcomes, these significant relationships are correlational and cannot permit causal conclusions. Three studies were identified that specifically examine changes in HRQOL in youth with SB over time (Parekh et al., 2006; Bellin et al., 2013; Murray et al., 2015). These longitudinal studies mark an improvement in study methodology. However, the literature on quality of life in youth with SB would be improved with longitudinal studies that include more assessment points extending over a longer period of time, as each of the three identified studies included only two time points, with time between assessment points ranging from six months to two years. The use of longitudinal moderation and mediation designs to assess outcomes has been recommended for research with families of youth with SB (Holmbeck et al., 2006; Holmbeck & Devine, 2010). By studying potential mechanisms that underlie the relationship between parent functioning and youth functioning over time, findings can reveal both the nature of this relationship and why it exists.

The current study, therefore, seeks to address these limitations of the current literature by including (a) questionnaire and observational measures of family functioning, (b) youth-, mother-, and father- reports, (c) longitudinal data to examine the impact of parent factors (distress, parenting stress, and SB-specific parenting stress) on youth HRQOL over time, and (d) the use of a mediation design to assess the relationship between these parent factors and youth HRQOL as mediated by parenting behaviors for youth with SB.

The Current Study

The current study aimed to investigate parent factors and parenting behaviors in relation to child HRQOL in youth with SB. Three distinct parent variables, parent distress (distress as an individual), parenting stress (stress as a parent), and SB-specific parenting stress (stress of parenting a child with a SB) were examined in this study. (For a related discussion of distinctions among these parent factors, see Friedman et al., 2004. Please note that in this manuscript, parent distress is referred to as “psychosocial functioning.”) Specifically, this study aimed to differentiate the impacts of parent distress, parenting stress, and SB-specific parenting stress on youth HRQOL (Figure 1). The current study also aimed to investigate the role of parenting behaviors (acceptance, psychological control, and behavioral control) as mediators of the relationship between the aforementioned parent factors and youth HRQOL (Figure 2). It is believed that findings from this study will inform future research, as well as the development of evidence-based family interventions aimed at improving psychosocial functioning and quality of life in this population.

Study Hypotheses

The present study had three objectives. The first objective was to identify relationships between the parent variables and youth HRQOL. It was hypothesized that higher levels of parent distress, parenting stress, and SB-specific parenting stress will be associated with lower levels of youth HRQOL (*Hypotheses 1a-1c;* Figure 1). These hypotheses were tested both cross-sectionally and longitudinally, with parent variables at Time 1 predicting youth HRQOL at Time 1, Time 2, and Time 3. It was hypothesized that the relationship between parent variables and youth HRQOL will gradually become less robust as time is extended.

The second objective was to determine which of the three parent variables (parent distress, parenting stress, and SB-specific parenting stress) best predicts levels of youth HRQOL. It was hypothesized that parent distress will be the most robust predictor, with higher levels of parent distress at Time 1 being associated with lower levels of youth-reported HRQOL at Time 2 and Time 3 (*Hypothesis 2).*

The third objective was to examine the parent variables (parent distress, parenting stress, SB-specific parenting stress), parenting behaviors (acceptance, behavioral control, psychological control), and youth HRQOL in a single model (Figure 2). Specifically, it was predicted that higher levels of parent distress would predict higher levels of parental psychological control and lower levels of parental acceptance and behavioral control, which will, in turn, predict lower levels of youth HRQOL (*Hypothesis 3a*). The same predictions were made for other parent variables (i.e., parenting stress and SB-specific parenting stress; *Hypothesis 3b-3c*).

Figure 1. Model for Objective 1: The Association Between Parent Distress and Stress and Youth HRQOL in Youth with Spina Bifida

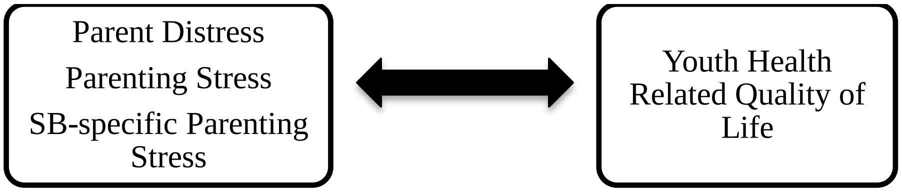
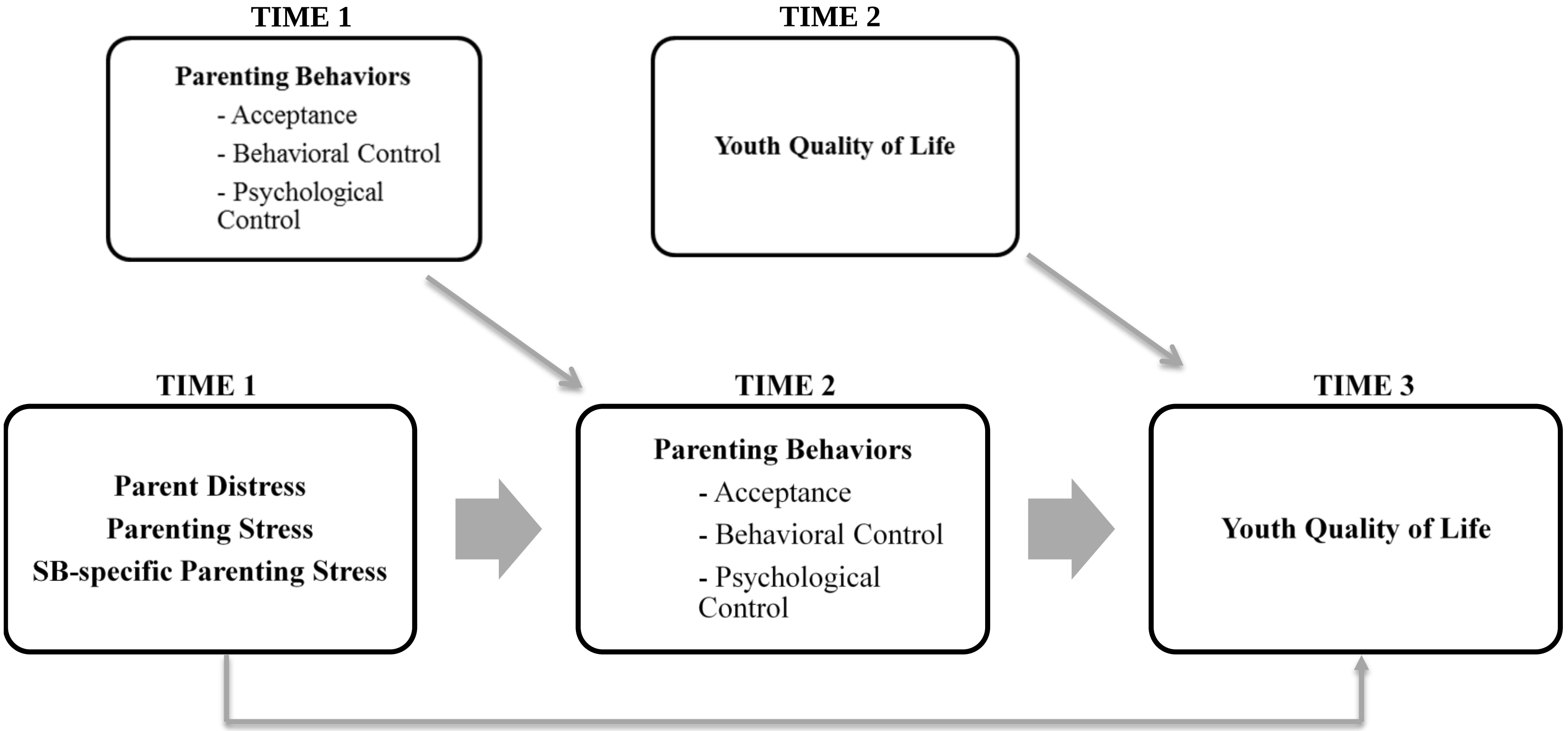


Figure 2. Model for Objective 3: Parenting Behaviors as Mediators of the Association Between Parent Distress, Parenting Stress, and SB-specific Parenting Stress and Youth HRQOL in Youth with Spina Bifida.



CHAPTER THREE

METHODS

Participants

Participants were recruited from an ongoing longitudinal study examining family and peer relationships, neuropsychological functioning, and psychological adjustment (e.g., Devine, et al., 2012). The current study focuses on the psychosocial and family functioning of families of youth with SB, with time points spaced two years apart. (The present study's analyses focus on the first three time points: Time 1(baseline), Time 2 (2 years), and Time 3 (4 years)). Families of youth with SB were recruited from four Midwestern hospitals and a statewide SB association. Families were recruited in person at regularly scheduled clinic visits and through recruitment letters. Interested families were screened in person or by phone by a member of the research team. Families were invited to participate if the child met the following criteria at Time 1: (a) diagnosis of SB (including myelomeningocele, lipomeningocele, and myelocystocele); (b) age 8-15 years; (c) ability to speak and read English or Spanish; (d) involvement of at least one primary caregiver; and (e) residence within 300 miles of Loyola University Chicago (to allow for data collection at participants' homes).

A total of 246 families were approached during recruitment, and 163 families agreed to participate. Of these 163 families, 21 families could not be contacted or declined to participate after their initial consent and two families did not meet inclusion criteria. The final sample of participants included 140 families of children with SB (at Time 1, 53.6% female, M_{age} = 11.40).

Procedure

The current study was approved by university and hospital Institutional Review Boards and utilized a multi-method, multi-informant longitudinal research design. Data were collected by trained undergraduate and graduate student research assistants during home visits that lasted approximately three hours. At Time 1, two 3-hour home visits were conducted, and two and four years later, at Time 2 and Time 3 respectively, only one 3-hour home visit was conducted. For home visits with families who primarily spoke Spanish in the home, at least one research assistant was bilingual. Informed consent from parents and assent from youth were obtained at the beginning of the first visit. Parents completed releases of information to allow for data collection from medical charts, health professionals, and school teachers. The larger protocol involved youth, parent, teacher, health professional, and peer questionnaires; youth, parent, and peer interviews; youth neuropsychological testing; videotaped family interaction tasks of the child and his/her parent(s); and videotaped peer interaction tasks of the youth and his/her friend. Parents completed identical questionnaires separately. Questionnaires that were only available in English were adapted for Spanish speakers using forward and back translation by a trained translation team from the University of Houston. The current study used youth- and parent-reported questionnaire data and observational data of family interaction tasks. Families received $150, a t-shirt, and a pen as compensation for participation at each time point. At Time 3, a sub-sample of participants was over 18 years old. For these participants, only the target young adult completed questionnaires, interviews, and neuropsychological assessments. No family or peer interaction tasks were completed during these “over 18” visits. Young adults received $100 as compensation for participation at this time point.

Measures

Covariates

Demographics. At Time 1 parents reported on youth and family demographic information through questionnaires. Parents reported on child age, gender, and race/ethnicity. Parents also reported on their own gender, ethnicity, education, employment, income, and relationship to child. The Hollingshead Index of socioeconomic status (SES) was computed using parents’ education and occupation, with higher scores indicating higher SES (Hollingshead, 1975).

Youth Illness Severity. Parents completed the Medical History Questionnaire (MHQ; Holmbeck et al., 2003). This survey contains questions about youth’s disease-specific medical information, including bowel and bladder functioning, ambulation, medications, providers and frequency of medical care, and surgery history. In addition to the MHQ, data were collected from participants’ medical charts to assess the following information: type of SB (i.e., lipomeningocele, meningocele, or myelomeningocele), shunt status, lesion level, (i.e., sacral, lumbar, or thoracic) and ambulation method (i.e., ankle-foot orthoses (AFOs), knee-ankle-foot orthoses (KAFOs), or hip-knee-ankle-foot orthoses (HKAFOs), wheelchair, or no assistance). These variables were used to compute an illness severity index based on inclusion in a specific group: shunt status (no = 1, yes = 2), myelomeningocele (no = 1, yes = 2), lesion level (sacral = 1, lumbar = 2, thoracic = 3), and ambulation status (no assistance/AFOs = 1, KAFOs/HKAFOs = 2, wheelchair = 3). Scores ranged from four to ten, with higher scores indicating higher levels of severity (see Hommeyer, Holmbeck, Willis, & Coers, 1999).

Youth IQ. Youth were administered the Vocabulary and Matrix Reasoning subtests of the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) at Time 1. Scores on these two subtests can be used to compute an estimated Full Scale IQ (FSIQ), which functions as a proxy for general intellectual functioning. The WASI is a well-validated measure of child intelligence with a normative mean of 100 and a standard deviation of 15. The Vocabulary subtest consists of 42 items used to assess a child’s expressive vocabulary, verbal knowledge, and fund of knowledge. The Matrix Reasoning subtest consists of 35 items designed to assess nonverbal fluid reasoning and general intellectual ability. These subtests have demonstrated high levels of internal consistency for individuals aged 6 to 16 years ($\alpha = .89$ for Vocabulary, $\alpha = .92$ for Matrix Reasoning; Wechsler, 1999).

Youth Measures

Youth’s HRQOL was assessed using youth report on the Pediatric Quality of Life Scale (PedsQL™ 4.0 Generic Core Scales; Varni, Seid, & Kurtin, 2001). The PedsQL assesses both physical and psychosocial aspects of quality of life. Due to the physical limitations associated with SB, the 8-item physical scale of the PedsQL will not be used in this study. The 15-item psychosocial scale is comprised of three subscales: emotional

(5 items), social (5 items), and school/work functioning (5 items). Each item asks how much of a problem a given task has been over the last month (for example, “I hurt or ache”) and is rated on a 5-point Likert scale (0 = never a problem, 4 = almost always a problem). Raw scores are converted to standard scores that range from 0 to 100, with higher scores indicating better HRQOL.

Parent Measures

Parent Distress. Mothers and fathers separately completed the Symptom Checklist-Revised (SCL-90-R; Derogatis, Rickels, & Rock, 1976). This measure assesses psychological symptoms in parents. Each item is rated on a 4-point scale ranging from 0 (not at all distressed) to 4 (extremely distressed) for symptoms experienced over the past week. The SCL-90-R is made up of nine symptom subscales and three larger indices, but only the Global Severity Index (GSI) will be used in this study. The GSI is the average of all items from all subscales, with higher scores indicating higher global distress. Previous studies using the GSI with this sample have demonstrated high internal consistency (α = .95 - .98; Devine et al., 2012).

Parenting Stress. The Parenting Stress Index (PSI, Abidin, 1990) was used to assess parenting stress (e.g., stress an individual experiences as a direct result of being a parent). Of the 24 items on this scale, 22 items consist of a statement about the parent-child relationship that is rated on a 4-point scale ranging from 1 (strongly disagree) to 4 (strongly agree). The final two items are statements about how parents view themselves as parents and are rated on 5-point scales. Previous research supports the validity of using single subscales (Abidin, 1990), and three subscales of the PSI – restriction of role, perceived parental competence, and social isolation – will be used. In creating total scores, raw scores will be converted to z-scores so that 4- and 5-point scale items can be totaled together. Higher scores on this measure indicate higher reported parenting stress.

SB-specific Parenting Stress. Parents completed the Family Stress Scale (FSS; Quittner, Glueckauf, & Jackson, 1990), a 19-item scale assessing common stressors in families of a child with SB. This scale assesses the stress an individual experiences as a direct result of parenting a child with SB. Of the 19 items, 13 are non-disease specific (e.g., "mealtimes and bedtimes") and 6 are disease-specific (e.g., "medical care/appointments"). Items are rated on a 5-point Likert scale (1= "not at all stressful" and 5 = "extremely stressful"). The current study will use a total score comprised of the disease-specific items with higher total scores indicating higher levels of SB-specific parenting stress. Research using the FSS has shown adequate internal consistency in chronically ill populations (α = .81 - .84; Quittner et al., 1998).

Observed Parenting Behaviors: Acceptance, Behavioral Control, and Psychological Control. To reduce common-method variance between the independent parent variables (that are being assessed via parent self-report) and the mediating parenting variables, observations of parenting variables (rather than self-report) will be used. Families (mother, father, and youth) completed a set of video-taped interaction tasks designed to generate family interaction and discussion. These structured tasks were counter-balanced and included a warm-up game, a discussion of two age-appropriate vignettes, a discussion of transferring disease-specific responsibilities to the child, and a discussion of identified family conflicts. In the vignettes task, families were given two age-appropriate vignettes of situations (one specific to youth with SB) that adolescents might typically encounter, and were asked to discuss possible resolutions to these situations. In the transferring of responsibilities task, families were asked to discuss one to two responsibilities that could eventually be transferred from the parent to the child (e.g., independent catheterization). In the conflict task, each family member was first asked to complete the Parent-Adolescent Conflict Scale (PAC; Prinz, Foster, Kent, & O'Leary, 1979), which is a 15-item measure that assesses the occurrence of common potentially conflictive issues (e.g., "Whether he/she does chores around the house) between the parent and child. Ten items specific to SB (e.g., "How he/she does his/her skin checks") were added for the current study. All 25 items will be used for the proposed analyses. In completing the PAC, respondents first indicate whether an issue was discussed within the past two weeks ("yes/no"). If an issue was discussed in the last two weeks, the respondent then indicates how often the discussion has occurred on a 4-point scale (1 = "not often," 4 = "very often) and how "hot" the discussions were on a 5-point scale (1 = "calm" and 5 = "very angry). Families were then presented with the five issues that they rated as most common and intense, and were asked to discuss and attempt to resolve three or more of these issues. Families were given 10 minutes to complete each of the observational tasks.

These videotaped interactions, with the exception of the warm-up game task, were coded using a global-coding method called the Family Interaction Macro-coding System (FIMS; Holmbeck, Zebracki, Johnson, Belvedere, & Hommeyer, 2007; Kaugars et al., 2011). FIMS includes codes that assess interaction style, conflict, affect, control, problem solving, and family systems using 5-point Likert-type ratings. For example, the item assessing ‘‘Warmth’’ captures signs of a positive connection in the relationship as shown through verbal or nonverbal behaviors (1 = “very cold” and 5 = “very warm”). The subscales examined in this study, Parental Acceptance, Behavioral Control, and Psychological Control, were composite codes; they were composed of multiple items (see Table 1). Internal consistency alphas for these subscales in this study were found to range from .67 to .91 and interrater reliability coefficients (ICCs) ranging from .61 to .89.

Table 1: FIMS Composite Codes with Individual Items

FIMS Composite Codes	**FIMS Items**
Parental Acceptance	Listens to others
	Humor and laughter
	Warmth
	Anger[a]
	Supportiveness
Parental Behavioral Control	Confidence in stating opinions
	Parental structuring of task
	Parental dominance
Parental Psychological Control	Pressures others to agree
	Tolerates differences and disagreements[a]
	Receptive to statements made by others[a]
	Parent promotes autonomy in child [a]

[a] Reverse coded

Statistical Treatment

Preliminary Analyses

Prior to hypothesis testing, the psychometric properties (e.g., alphas) of all measures were evaluated. This included determining whether variables contained outliers or were skewed. Descriptive statistics were computed for all outcome measures to determine basic distributional properties. Data transformation and reduction techniques were used when appropriate. It was anticipated that there would be families that would not participate at all three time points. Therefore, prior to hypothesis testing, attrition analyses were performed to evaluate differences between families who discontinued participation and those who did not.

Primary Analyses

All analyses included youth IQ, SB disease severity, child age, and SES as covariates, as all four of these may contribute to parent distress, parenting stress, SB-specific parenting stress, and youth HRQOL. In order to have a broad understanding of the dependence among the independent and dependent variables, Pearson correlations will be performed and a correlation matrix will be created prior to hypothesis testing.

Analytic Plan for Objective 1. A series of hierarchical multiple regression analyses were conducted to examine associations between parent distress, parenting stress, and SB-specific parenting stress at Time 1 with youth HRQOL at Times 1, 2, and 3. When running cross-sectional regression analyses, independent variables were entered in the following order: (Step 1) covariates – IQ, illness severity, SES; (Step 2) individual predictor (parent distress, parenting stress, or SB-specific parenting stress). When running longitudinal regression analyses, independent variables were entered in the following order: (Step 1) HRQOL at Time 1(for Time 2 outcome) or HRQOL at Time 2 (for Time 3 outcome); (Step 2) covariates – IQ, illness, severity, age, SES; (Step 3) individual predictor. Separate regressions were run for each predictor variable, and separate sets of regression analyses were run for maternal and paternal variables.

Analytic Plan for Objective 2. Additional hierarchical multiple regression analyses were run to determine which parent variables (parent distress, parenting stress, or SB-specific parenting stress) are the best predictors of youth HRQOL at Time 2 and Time 3. Independent variables were entered in the following order: (Step 1) HRQOL at Time 1 (for Time 2 outcomes) or HRQOL at Time 2 (for Time 3 outcome); (Step 2) covariates – IQ, illness severity, age, SES at Time 1; (Step 3) parent distress, parenting stress, and SB-specific parenting stress at Time 1. Variables entered at Step 3 were entered in a forward selection fashion, such that the variable that significantly improves the model most will be entered first and the process will be repeated until none of the independent variables significantly improves the model.

Analytic Plan for Objective 3. Preacher and Hayes' (2008) bootstrapping methods were employed to determine the impact of parent distress, parenting stress, and SB-specific parenting stress at Time 1 on youth HRQOL at Time 3, as mediated by parenting behaviors (acceptance, behavioral control, psychological control) at Time 2. Bootstrapping has been validated in the literature and is preferred over other methods, such as the Sobel Test (Sobel, 1982), as bootstrapping is less conservative and reduces the possibility of Type II errors (Preacher & Hayes, 2008). This procedure produces an empirical approximation of the product of the estimated coefficients' sampling distribution constituting the direct path and percentile-based bootstrap confidence intervals (CIs and bootstrap measures of standard errors using 5000 resamples, with replacement, from the dataset (Preacher & Hayes, 2008)). When zero is not between the upper and lower bounds of the confidence interval, it can be claimed, with 95%

confidence, that the indirect effect is not zero, indicating a significant indirect effect. A total of thirty models were run with either maternal or paternal parent distress, subscales of parenting stress (role restriction, perceived competence, social isolation), or SB-specific parenting stress at Time 1 predicting youth HRQOL at Time 3, mediated by parenting behaviors (acceptance, behavioral control, or psychological control) at Time 2, while controlling for youth IQ, illness severity, age, SES, and parenting behaviors at Time 1 and youth HRQOL at Time 2 (2 parents X 5 predictors X 3 mediators = 30 models).

For mediation models analyzed using percentile bootstrapping methods, assuming a power of .80, and an alpha of .05, a sample size of 36 is required to detect large effect sizes, a sample size of 78 is required to detect medium effect sizes, and a sample of 558 is required to detect small effect sizes (Fritz & MacKinnon, 2007). Thus, the current study had enough power to detect medium or large effect sizes.

CHAPTER FOUR

RESULTS

Preliminary Analyses

All variables were examined for outliers, but none were identified. Additionally, all independent and dependent variables were tested for skewness. A conservative approach to identifying skewness was used; variables were considered skewed and transformed if skewness values were greater than 1.0 (Tabachnick & Fidell, 2013). The results indicated that four variables were positively skewed: mother-report on the SCL-90 (skewness value = 2.90), father-report on the SCL-90 (skewness value = 1.39), mother report on the Family Stress Scale (skewness value = 1.05), and father report on the Family Stress Scale (skewness value = 1.44). Each of these variables were first transformed using the square root transformation. However, these variables continued to be skewed significantly, and the log transformation was used for all four variables.

Attrition Analyses

As was anticipated, though a majority of families did participate at all three time points ($N = 94$; 67%), not all families who participated at Time 1 participated at each subsequent time points ($N_{\text{Time 1 only}} = 18$, 12.9%; $N_{\text{Time 1\& Time 2}} = 18$, 12.9%; $N_{\text{Time 2 \& Time3}} = 10$, 7.1%). Univariate analyses of variance (ANOVAs) were performed to compare these four groups at Time 1 on SES, youth IQ, youth age, youth illness severity, and youth-reported HRQOL. No significant differences were found on these factors among those who participated at all three time points, those who participated only at Time 1, those who participated at Time 1 and Time 2 and those who participated at Time 1 and Time 3 (SES: $F(3, 128) = 1.37$, $p = .26$; IQ: $F(3, 128) = 1.50$, $p = .22$; age: $F(3, 126) = 1.87$, $p = .14$; illness severity: $F(3, 105) = .60$, $p = .62$; HRQOL: $F(3, 120) = 1.98$, $p = .12$).

Additionally, a second set of attrition analyses were performed. The three groups of partial-completers were combined and t-test analyses were performed comparing only two groups: those families that completed all three time points (full-completers) and those who did not complete all three time points (partial-completers) on the factors of SES, youth IQ, youth age, youth illness severity, and youth-reported HRQOL at Time 1. These t-tests revealed no significant differences between full-completers and partial-completers on any of these factors (SES: $t(128) = 1.02$, $p = .31$; IQ: $t(130) = 1.63$, $p = .11$; age: $t(136) = -1.32$, $p = .19$; illness severity: $t(107) = -.57$, $p = .57$; HRQOL: $t(121) = -.46$, $p = .65$).

Correlation Matrix

Prior to hypothesis testing, a series of Pearson correlations were performed, and a correlation matrix was created (Table 2). This matrix shows the correlation between the independent variables and the dependent variable (HRQOL) at T1, T2, and T3. Results indicated that HRQOL was positively correlated across time points ($\text{HRQOL}_{\text{T1-T2}}$: $r=.33$; $\text{HRQOL}_{\text{T1-T3}}$: $r = .33$; $\text{HRQOL}_{\text{T2-T3}}$: $r = .42$; all p’s < .01). Additionally, significant correlations were found among many of the covariates and independent variables. Participant age was positively correlated with father SB-specific parenting stress ($r = .26$, $p < .05$). Youth IQ was positively correlated with SES ($r = .48$, $p < .01$) and negatively correlated with paternal role restriction (*r* = -.26, *p* < .05). SES was also positively correlated with paternal social isolation (*r* = .21, *p* < .05), and negatively correlated with maternal role restriction (*r* = -.19, *p* < .05) and maternal social isolation (*r* = -.18, *p* < .05). Youth illness severity was positively associated with maternal distress (*r* = .22 *p* < .05).

A number of significant associations existed among the maternal and paternal distress and stress variables (Table 2). Despite significant correlations among the independent variables, because the constructs of distress, parenting stress, and SB-specific parenting stress were conceptualized as separate entities, composite scores were not created. Results indicated no significant correlations between the covariates or independent variables and HRQOL at any time point.

Table 2. Correlation Matrix of Covariates and Independent Variables and Health-Related Quality of Life at Each Time Point

		Youth							Mothers				
		HRQOL T1	HRQOL T2	HRQOL T3	Age	IQ	SES	Illness Severity	Distress	RR	PC	SI	SB Stress
Youth	HRQOL T1	*r* = 1											
	HRQOL T2	*r* = .33**	*r* = 1										
	HRQOL T3	*r* = .33**	*r* = .42**	*r* = 1									
	Age	*r* = -.05	*r* = -.03	*r* = -.20	*r* = 1								
	IQ	*r* = .18	*r* = .03	*r* = -.04	*r* = -.13	*r* = 1							
	SES	*r* = .14	*r* = .02	*r* = .01	*r* = .03	*r* =.48**	*r* = 1						
	Illness Severity	*r* = .03	*r* =- .09	*r* = -.19	*r* = .18	*r* =-.10	*r* =.13	*r* = 1					
Mothers	Distress	*r* = -.07	*r* = .05	*r* = -.08	*r* = .06	*r* =.05	*r* =-.12	*r* =.22*	*r* = 1				
	Role Restriction	*r* = .05	*r* = .14	*r* = .18	*r* = .04	*r* =-.05	*r* =-.19*	*r* =.06	*r* =.46**	*r* = 1			
	Perceived Competence	*r* = .06	*r* =- .12	*r* = -.05	*r* = -.12	*r* =.01	*r* =.12	*r* =-.01	*r* =-.40**	*r* = -.58**	*r* = 1		
	Social Isolation	*r* = .01	*r* = .09	*r* = .13	*r* =-.01	*r* =-.05	*r* =-.18*	*r* =.16	*r* =.52**	*r* =.63**	*r* =-.46**	*r* = 1	
	SB-Specific Parenting Stress	*r* = ..04	*r* = -.06	*r* = .01	*r* =-.01	*r* =.08	*r* =-.05	*r* =.11	*r* =.40**	*r* =.36**	*r* =-.26*	*r* =.38**	*r* = 1
Fathers	Distress	*r* = -.14	*r* = -.28	*r* = -.06	*r* =.10	*r* =-.01	*r* =.07	*r* =.18	*r* =.28**	*r* =.17	*r* =-.22*	*r* =.17	*r* =.13
	Role Restriction	*r* = -.03	*r* = -.07	*r* = -.02	*r* =.09	*r* =-.26*	*r* =.11	*r* =-.02	*r* =.18	*r* =.22*	*r* =-.14	*r* =.26*	*r* =.06
	Perceived Competence	*r* = -.02	*r* = -.02	*r* = -.02	*r* =-.05	*r* =.18	*r* =.18	*r* =-.13	*r* =-.18	*r* =-.13	*r* =.22*	*r* =-.07	*r* =-.16
	Social Isolation	*r* = .01	*r* = -.01	*r* = -.11	*r* =.10	*r* =-.06	*r* =-.06	*r* =.17	*r* =.35	*r* =.25*	*r* =-.16	*r* =.42	*r* =.12
	SB-Specific Parenting Stress	*r* = -.09	*r* = -.20	*r* = -.24	*r* =.26*	*r* =-.08	*r* =-.08	*r* =.14	*r* =.28*	*r* =.23*	*r* =-.15	*r* =.12	*r* =.52**

Key:
RR = Role Restriction
PC = Perceived Competence
SI = Social Isolation
SB stress = SB-specific parenting stress

* Correlation is significant at .05 level
** Correlation is significant at .01 level

Table 2. Correlation Matrix of Covariates and Independent Variables and Health-Related Quality of Life at Each Time Point (continued)

		Fathers				
		Distress	RR	PC	SI	SB Stress
Youth	HRQOL T1					
	HRQOL T2					
	HRQOL T3					
	Age					
	IQ					
	SES					
	Illness Severity					
Mothers	Distress					
	Role Restriction					
	Perceived Competence					
	Social Isolation					
	SB-Specific Parenting Stress					
Fathers	Distress	*r* = 1				
	Role Restriction	*r* =.24*	*r* = 1			
	Perceived Competence	*r* =-.33**	*r* =-.50**	*r* = 1		
	Social Isolation	*r* =.35**	*r* =.56**	*r* =-.44**	*r* = 1	
	SB-Specific Parenting Stress	*r* =.35**	*r* =.31**	*r* =-.30**	*r* =.23*	*r* = 1

Key:

RR = Role Restriction
PC = Perceived Competence
SI = Social Isolation
SB stress = SB-specific parenting stress

* Correlation is significant at .05 level
** Correlation is significant at .01 level

Hypothesis Testing

Objective 1

A series of hierarchical multiple regression analyses were conducted to examine the associations between parent distress, parenting stress, and SB-specific parenting stress at Time 1 with youth HRQOL at Times 1, 2, and 3 (Table 3).

Time 1. Hierarchical multiple regression analyses were conducted to evaluate how well each mother factor (parent distress, parenting stress – role restriction, perceived competence, social isolation – and SB-specific parenting stress) predicted youth-reported HRQOL at Time 1 (Table 3). For each analysis, the covariates of age, IQ, SES, and illness severity were entered simultaneously in the first step. Predictors were entered in step 2. Each predictor was entered in a separate regression. Cross-sectionally at Time 1, maternal distress (β=-.03, p=.80), role restriction (β=.15, p=.17), perceived competence (β=.003, p=.98), social isolation (β=.07, p=.55), and SB-specific parenting stress (β=-.01, p=.91) were not significant predictors of youth-reported HRQOL.

Parallel multiple regression analyses were performed for father factors (Table 3). Cross-sectionally at Time 1, paternal distress (β=-.17 p=.17), role restriction (β=-.05, p=.72), perceived competence (β=.04, p=.74), social isolation (β=.02, p=.87), and SB-specific parenting stress (β=-.11, p=.43) were not significant predictors of youth-reported HRQOL.

Time 2. Hierarchical multiple regression analyses were conducted to evaluate how well each parent factor at Time 1 predicted youth-reported HRQOL at Time 2 (Table 3). For each analysis, HRQOL at Time 1 was entered in Step 1, the covariates of age, IQ, SES, and illness severity were entered simultaneously at Step 2, and the individual predictors (at Time 1) were entered at Step 3. Each predictor was entered in a separate regression. In these analyses, maternal distress (β=-.04, p=.76), role restriction (β=.08, p=.46), perceived competence (β=.04, p=.70), social isolation (β=.07, p=.51), and SB-specific parenting stress (β=-.21, p=.10) were not significant predictors of youth-reported HRQOL at Time 2. Additionally, paternal distress (β=-.12, p=.34), role restriction (β=.001, p=.99), perceived competence (β=-.11, p=.34), social isolation (β=.04, p=.77), and SB-specific parenting stress (β=-.23, p=.07) were not significant predictors of youth-reported HRQOL at Time 2.

Time 3. Hierarchical multiple regression analyses were conducted to evaluate how well each parent factor at Time 1 predicted youth HRQOL at Time 3 (Table 3). For each analysis, HRQOL at Time 2 was entered in Step 1, the covariates of age, IQ, SES, and illness severity were entered simultaneously at Step 2, and the individual predictors (at Time 1) were entered in Step 3. Each predictor was entered in a separate regression. In these analyses maternal distress (β=.09, p=.45), role restriction (β=.20, p=.08), perceived competence (β=-.11, p=.33), social isolation (β=.11, p=.36), and SB-specific parenting stress (β=.11, p=.42) were not significant predictors of youth-reported HRQOL at Time 3. Parallel analyses were run for paternal predictors. Paternal distress (β=.21, p=.13), role restriction (β=-.05, p=.72), perceived competence (β=-.10, p=.41), social isolation (β=.03, p=.82), and SB-specific parenting stress (β=-.10, p=.52) were not significant predictors of youth-reported HRQOL at Time 3.

Table 3. Summary of Regression Analyses for Parent Variables Predicting Youth-Reported Health-Related Quality of Life (Objective 1).

		Time 1				Time 2				Time 3			
	Variable	Step	*b*	β	ΔR^2	Step	*b*	β	ΔR^2	Step	*b*	β	ΔR^2
Mothers	Distress	2	-1.79	-.03	.00	3	-2.28	-.04	.00	3	5.12	.09	.01
	Role Restriction	2	3.44	.15	.02	3	2.13	.08	.01	3	4.52	.20	.04
	Perceived Competence	2	.12	.01	.00	3	1.67	.04	.00	3	-3.59	-.11	.01
	Social Isolation	2	1.58	.07	.01	3	1.82	.07	.01	3	2.23	.11	.01
	SB-specific Parenting Stress	2	-.38	-.01	.00	3	-6.45	-.21	.04	3	2.76	.11	.01
Fathers	Distress	2	-11.27	-.17	.03	3	-7.77	-.12	.01	3	13.84	.21	.03
	Role Restriction	2	-.20	-.05	.00	3	.01	.00	.00	3	-.17	-.05	.00
	Perceived Competence	2	.14	.04	.00	3	-.40	-.11	.01	3	-.31	-.10	.01
	Social Isolation	2	.10	.02	.00	3	.18	.04	.00	3	.12	.03	.00
	SB-specific Parenting Stress	2	-2.53	-.11	.02	3	-6.92	-.23	.04	3	-2.23	-.10	.01

Note: All predictor variables are measured at Time 1, and **separate regressions were run for each predictor for each parent**. For cross-sectional analyses, the covariates of age, IQ, SES, and illness severity were entered at Step 1. For longitudinal analyses, youth HRQOL at the previous time point was entered at Step 1 and the covariates of age, IQ, SES, and illness severity were entered at Step 2.

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

Objective 2.

To address Objective 2, to determine which parent variables (parent distress, parenting stress (role restriction, perceived competence, social isolation), and SB-specific parenting stress) best predicted youth HRQOL, additional hierarchical multiple regression analyses were performed (Tables 4-7).

Time 2. Hierarchical multiple regression analyses were conducted to evaluate which parent factor at T1 best predicted youth-reported HRQOL at Time 2. For each analysis, HRQOL at Time 1 was entered in Step 1, the covariates of age, IQ, SES, and illness severity were entered simultaneously at Step 2, and the parent predictors (at Time 1) were entered at Step 3 in a forward selection fashion (Table 4). None of the maternal variables were found to be significant predictors of youth HRQOL at Time 2, but were entered into the model in the following order: SB-specific parenting stress (β=-.24, p=.06), social isolation (β=.17, p=.15), role restriction (β=.11, p=.47), perceived competence (β=.07, p=.62), distress (β=-.04, p=.79). Parallel analyses with paternal variables were performed (Table 5). None of the paternal variables were found to be significant predictors of youth HRQOL at Time 2, but were entered into the model in the following order: SB-specific parenting stress (β=-.23, p=.07), perceived competence (β=-.15, p=.22), distress (β=-.05, p=.76), social isolation (β=.03, p=.88), role restriction (β=.001, p=.99).

Table 4. Summary of Hierarchical Multiple Regression Analyses for Maternal Variables Predicting Youth-Reported Health-Related Quality of Life at Time 2 (Objective 2).

Variable	Step	*b*	*SE (b)*	β	ΔR^2
T1 HRQOL	1	.38	.12	.37	.138**
IQ	2	.02	.13	.02	.026
SES		.06	.18	.05	
Illness Severity		-.73	1.40	-.07	
Child Age		-.89	.95	-.13	
SB-specific Parenting Stress	3	-7.55	3.90	-.24	.05
Social Isolation	4	4.52	3.53	.17	.02
Role Restriction	5	2.99	4.15	.11	.01
Perceived Competence	6	2.94	5.96	.07	.00
Distress	7	-2.89	10.56	-.04	.00

Note: All predictor variables are measured at Time 1. The covariates of age, IQ, SES, and illness severity (Step 2) were entered in a simultaneous fashion. The predictors (Steps 3-7) were entered in a forward selection fashion.
$*p < .05$, $**p < .01$

Table 5. Summary of Hierarchical Multiple Regression Analyses for Paternal Variables Predicting Youth-Reported Health-Related Quality of Life at Time 2 (Objective 2).

Variable	Step	*b*	*SE (b)*	β	ΔR^2
T1 HRQOL	1	.57	.13	.52	.27**
IQ	2	.01	.13	.01	.04
SES		.16	.17	.14	
Illness Severity		.56	1.39	.05	
Child Age		-1.17	.87	-.17	
SB-specific Parenting Stress	3	-6.92	3.73	-.23	.04
Perceived Competence	4	-.53	.43	-.15	.02
Distress	5	-3.23	10.71	-.05	.00
Social Isolation	6	.12	.80	.03	.00
Role Restriction	7	.01	.68	.00	.00

Note: All predictor variables are measured at Time 1. The covariates of age, IQ, SES, and illness severity (Step 2) were entered in a simultaneous fashion. The predictors (Steps 3-7) were entered in a forward selection fashion.
$*p < .05$, $**p < .01$

Time 3. Hierarchical multiple regression analyses were conducted to evaluate which parent factor at Time 1 best predicted youth-reported HRQOL at Time 3. For each analysis, HRQOL at Time 2 was entered in Step 1, the covariates of age, IQ, SES, and illness severity were entered simultaneously at Step 2, and the parent predictors (at Time 1) were entered at Step 3 in a forward selection fashion (Table 6). Again, none of the maternal variables were found to be significant predictors of youth HRQOL at Time 3, but were entered into the model in the following order: role restriction (β=.23, p=.08), social isolation (β=-.07, p=.66), SB-specific parenting stress (β=.07, p=.64), distress (β=04, p=.82), perceived competence (β=.01, p=.94). Parallel analyses were performed with paternal variables (Table 7). None of the paternal variables were found to be significant predictors of youth HRQOL at Time 3, but were entered into the model in the following order: distress (β=.22, p=.14), SB-specific parenting stress (β=-.15, p=.31), perceived competence (β=-.14, p=.32), role restriction (β=-.22, p=.17), social isolation (β=-.04, p=.85).

Table 6. Summary of Hierarchical Multiple Regression Analyses for Maternal Variables Predicting Youth-Reported Health-Related Quality of Life at Time 3 (Objective 2).

Variable	Step	*b*	*SE (b)*	β	ΔR^2
T2 HRQOL	1	.43	.11	.50	.25**
IQ	2	.08	.12	.12	.05
SES		.02	.15	.02	
Illness Severity		-1.01	1.31	-.11	
Child Age		-.54	.83	-.10	
Role Restriction	3	5.48	.81	-.08	.05
Social Isolation	4	-1.40	3.13	-.07	.00
SB-specific Parenting Stress	5	1.87	3.97	.07	.00
Distress	6	2.07	8.97	.04	.00
Perceived Competence	7	.423	5.35	-.01	.00

Note: All predictor variables are measured at Time 1. The covariates of age, IQ, SES, and illness severity (Step 2) were entered in a simultaneous fashion. The predictors (Steps 3-7) were entered in a forward selection fashion.

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

Table 7. Summary of Hierarchical Multiple Regression Analyses for Paternal Variables Predicting Youth-Reported Health-Related Quality of Life at Time 3 (Objective 2).

Variable	Step	*b*	*SE (b)*	β	ΔR^2
T2 HRQOL	1	.49	.11	.56	.31**
IQ	2	-.01	.12	-.01	.07
SES		.10	.15	.10	
Illness Severity		-2.29	1.23	-.25	
Child Age		-.47	.81	-.09	
Distress	3	13.77	9.11	.22	.03
SB-specific Parenting Stress	4	-3.5	3.42	-.15	.02
Perceived Competence	5	-.39	.39	-.14	.02
Role Restriction	6	-.74	.53	-.22	.03
Social Isolation	7	-.14	.73	-.04	.00

Note: All predictor variables are measured at Time 1. The covariates of age, IQ, SES, and illness severity (Step 2) were entered in a simultaneous fashion. The predictors (Steps 3-7) were entered in a forward selection fashion.

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

Objective 3.

The third objective was to examine the longitudinal effects of parenting behaviors as mediators of the relationships between parent factors and youth HRQOL (Figure 2). Based on the findings above, no significant direct effects were expected to be found (as the parent factors included were not found to be significant predictors of youth HRQOL). However, Hayes' bootstrapping methods were used to test for indirect effects.

Results indicated no significant direct or indirect mediation effects (p's $> .05$). Two significant relationships were found between specific parent factors and parenting behaviors. Maternal isolation at Time 1 significantly predicted maternal acceptance at Time 2 (β=-.11, p<.05), with higher isolation predicting lower observed maternal acceptance. Paternal SB-specific parenting stress at Time 1 significantly predicted paternal psychological control at T2 (β=-.19, p=.05), with higher levels of SB-specific parenting stress predicting lower observed paternal psychological control. Despite these significant effects, these parenting behaviors were not found to significantly predict youth HRQOL, thus did not significantly mediate the relationship between maternal isolation or paternal SB-specific parenting stress and youth HRQOL (p's>.05).

Exploratory Analyses

Discrepancies have been found between youth-report and parent proxy report of HRQOL within the families of youth with SB. Specifically, parents have been found to report lower HRQOL than youth self-report (Murray et al., 2015). In this study, mothers and fathers were asked to report on their child's HRQOL. Given the previously found discrepancy between self- and parent proxy-report of HRQOL, exploratory analyses were performed examining study Objectives 1 and 2, using parent proxy-report of youth HRQOL.

Assessment of proxy-report of HRQOL

At each study time point parent-proxy report of HRQOL was assessed using the PedsQL, which has well-established reliability and validity in children with both acute and chronic health conditions. The parent proxy-report version of the PedsQL asks parents how much of a problem each item has been over the past month using a 5-point Likert scale rating (0 = never a problem to 4 = almost always a problem). Raw scores are then transformed into standard scores ranging form 0 to 100, with higher scores indicating better HRQOL. Similar to the self-report PedsQL (described previously), the parent proxy-report of the PedsQL yields a 15-item psychosocial total score, as well as four subscale scores to assess a child's physical, emotional, social, and school functioning. Given the physical limitations associated with SB, the physical subscale of the proxy-report PedsQL was deemed inappropriate and only the psychosocial total score was used in analyses. In the current study, internal consistency was adequate (α's = 0.83 – 0.90).

In the current study, small, statistically significant correlations were found between self-report and parent-proxy report of HRQOL. At T1, both mother proxy-report ($r = .22$, $p < .05$) and father proxy-report ($r = .28$, $p < .01$) were positively associated with youth self-report. Correlations were significant at T2 (mother proxy-report: $r = .41$, father proxy-report: $r = .43$; *p's* < .01). However at T3, while mother proxy-report remained significantly correlated ($r = .37$, $p < .01$), father proxy-report was no longer significantly correlated with youth self-report ($r = .22$, $p=.15$). Additionally, at each time point, mother proxy-report and father proxy-report were moderately correlated (T1: $r = .51$, T2: $r = .59$, T3: $r = .45$, all p's $< .01$).

Objective 1 – Exploratory Analyses.

Hierarchical multiple regression analyses were conducted to evaluate how well each mother factor (parent distress, parenting stress – role restriction, perceived competence, social isolation – and SB-specific parenting stress) predicted mother-reported youth HRQOL (Table 8). For each analysis, the covariates of age, IQ, SES, and illness severity were entered simultaneously in the first step. Cross-sectionally at Time 1, maternal distress ($\beta=-.21$, $p<.05$) and maternal SB-specific parenting stress ($\beta=-.41$, $p<.05$) were significantly associated with mother-report of youth HRQOL, such that increased distress and SB-specific parenting stress were associated with lower mother-report of youth HRQOL. The parenting stress factors of role restriction, perceived competence, and social isolation were not significantly associated with mother report of youth HRQOL (p's$>.05$). Longitudinal analyses (predicting mother report of youth HRQOL at Time 2 and Time 3) did not reveal any significant predictors.

Parallel multiple regression analyses were performed for father factors prediction father-report of youth HRQOL (Table 8). Similar to analyses of mother-reports, cross-sectionally at Time 1, paternal distress ($\beta=-.26$, $p<.05$) and paternal SB-specific parenting stress ($\beta=-.43$, $p<.05$) were significantly associated with father-report of youth HRQOL, such that increased distress and SB-specific parenting stress were associated with lower father-report of youth HRQOL. The parenting stress factors of role restriction, perceived competence, and social isolation were not significantly associated with father report of youth HRQOL (*p*'s>.05). Longitudinal analyses (predicting father report of youth HRQOL at Time 2 and Time 3) did not reveal any significant predictors. Because of these null longitudinal results, further longitudinal exploratory analyses (addressing Objectives 2 and 3) were not performed.

Table 8. Summary of Regression Analyses for Parent Variables Predicting Parent-Reported Health-Related Quality of Life (Exploratory Analyses – Objective 1).

		Time 1				Time 2				Time 3			
	Variable	Step	*b*	β	ΔR^2	Step	*b*	β	ΔR^2	Step	*b*	β	ΔR^2
Mothers	Distress	2	-10.45	-.21	.04*	3	-8.84	-.14	.02	3	3.21	.05	.01
	Role Restriction	2	-2.80	-.16	.03	3	-.13	-.01	.00	3	-1.42	-.06	.01
	Perceived Competence	2	4.55	.16	.03	3	.72	.02	.00	3	4.96	.14	.02
	Social Isolation	2	-2.44	-.13	.02	3	.92	.04	.01	3	-.77	-.03	.01
	SB-specific Parenting Stress	2	-8.74	-.41	.17**	3	.61	.02	.00	3	-5.43	-.18	.03
Fathers	Distress	2	-14.06	-.26	.06*	3	3.28	.05	.07	3	1.90	.02	.00
	Role Restriction	2	-.43	-.12	.01	3	.43	.12	.01	3	.57	.13	.01
	Perceived Competence	2	.56	.20	.04	3	-.67	-.22	.04	3	.48	.13	.01
	Social Isolation	2	-.91	-.22	.04	3	.49	.59	.01	3	-1.16	-.22	.01
	SB-specific Parenting Stress	2	-8.68	-.43	.17**	3	.75	.03	.00	3	1.56	.05	.01

Note: All predictor variables are measured at Time 1, and **separate regressions were run for each predictor for each parent**. For cross-sectional analyses, the covariates of age, IQ, SES, and illness severity were entered at Step 1. For longitudinal analyses, youth HRQOL at the previous time point was entered at Step 1 and the covariates of age, IQ, SES, and illness severity were entered at Step 2.

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

Objective 2 – Exploratory Analyses.

This study's second objective was to determine which parent variables (parent distress, parenting stress (role restriction, perceived competence, social isolation), and SB-specific parenting stress) best predicted youth HRQOL. Based on the significant results described above, cross-sectional hierarchical multiple regression analyses were performed to determine which of the mother and father factors best predicted that parent's report of youth HRQOL at Time 1. For both mothers (Table 9) and fathers (Table 10), with all of the parent factors entered into the model, only SB-specific parenting stress significantly predicted parent-report of youth HRQOL (mothers: β=-.40, p<.05; fathers: β=-.43, p<.05).

Table 9. Summary of Hierarchical Multiple Regression Analyses for Maternal Variables Predicting Mother-Reported Health-Related Quality of Life at Time 1.

Variable	Step	*b*	β	ΔR^2
IQ	1	.03	.05	.09
SES		.09	.11	
Illness Severity		-.70	-.09	
Child Age		-1.18	-.24	
SB-specific Parenting stress	2	-8.37	-.40	.15**
Distress	3	-8.23	-.17	.02
Perceived Competence	4	1.52	.06	.01
Role Restriction	5	-.44	-.03	.00
Social Isolation	6	.30	.02	.00

Note: All predictor variables are measured at Time 1. The covariates of age, IQ, SES, and illness severity (Step 1) were entered in a simultaneous fashion. The predictors (Steps 2-6) were entered in a forward selection fashion.
*p < .05, **p < .01

Table 10. Summary of Hierarchical Multiple Regression Analyses for Paternal Variables Predicting Father-Reported Health-Related Quality of Life at Time 1.

Variable	Step	*b*	β	ΔR^2
IQ	1	.22	.32	.10
SES		-.14	-.15	
Illness Severity		.48	.05	
Child Age		-.56	-.10	
SB-specific Parenting stress	2	-8.68	-.43	.17**
Distress	3	-10.80	-.19	.03
Social Isolation	4	-.37	-.09	.01
Role Restriction	5	.56	.17	.02
Perceived Competence	6	-.07	-.16	.00

Note: All predictor variables are measured at Time 1. The covariates of age, IQ, SES, and illness severity (Step 1) were entered in a simultaneous fashion. The predictors (Steps 2-6) were entered in a forward selection fashion.

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

CHAPTER FIVE

DISCUSSION

Past research has examined HRQOL in youth with SB, and this research indicates that these youth have poorer HRQOL compared to TD youth and youth with other chronic health conditions (e.g., Murray et al., 2015). While some factors (SES, IQ, illness severity) have been found to impact HRQOL in this population, these are all unchangeable demographic factors or difficult-to-modify illness-related factors. Therefore, understanding modifiable factors that may influence HRQOL in youth with SB is necessary to inform intervention development targeting improvement in this important construct. The current study sought to identify modifiable targets for intervention, specifically parent factors that may influence HRQOL. Research in other illness groups has found that parent factors, such as parent distress and parenting stress, can influence youth quality of life above and beyond the severity of the youth's illness (Bolghan-Abadi, Kimiaee, & Amir, 2011; Aran, Shaley, Biran, & Gross-Tsur, 2007). Therefore, the current study examined the impact of three distinct factors – parent distress, parenting stress, and SB-specific parenting stress – on HRQOL in youth with SB.

Despite previous research indicating that parent factors may influence HRQOL in youth with SB, the current study found that parent distress, parenting stress, and SB specific parenting stress did not significantly predict youth HRQOL either cross-sectionally or longitudinally. Additionally, the parenting behaviors of acceptance, behavioral control, and psychological control were not found to mediate the relationships between parent distress and stress and youth HRQOL directly or indirectly. Given the considerable influence of the family on psychosocial adjustment in youth with chronic illnesses, the finding that none of the parent variables predicted youth HRQOL was surprising. Previous studies have found associations between parent variables and youth HRQOL in this population (e.g., parental hope, parental overprotection, maternal psychological distress; Sawin et al., 2002; Abad, 2007). However, previous research with the same sample (using data from Time 1 and Time 2; Murray, 2013) found that family-environment factors did not significantly impact youth-reported HRQOL. Despite testing a comprehensive theoretical model of factors impacting HRQOL, Murray's (2013) study identified very few social-environmental factors that were predictive of decreased future HRQOL. Specifically, only one illness variable (pain intensity) and three social variables (parent-reported social competence, parent-reported community support, and a composite score of mother-, father-, and teacher-report of social skills) significantly predicted youth-reported HRQOL. This study found no other demographic, illness-related, or social-environmental factors to be related to youth-report of HRQOL (Murray, 2013). This study sought to expand on Murray's (2013) study by examining specific parent factors and parenting behaviors that may impact HRQOL for youth with SB. However the results of these studies suggest that family- and parent-specific factors may not significantly impact HRQOL in youth with SB.

The current study did find that some parent factors did significantly predict some parenting behaviors. Specifically, increased maternal social isolation was found to predict lower observed maternal acceptance, and increased paternal SB-specific parenting stress was found to predict lower observed paternal psychological control. While the latter finding is contrary to the hypothesized relationship, it is possible that fathers experiencing increased SB-specific parenting stress are more focused on maintaining their child's medical routine than they are on controlling their child's behavior, and, thus, exhibited lower levels of psychological control. This interpretation suggests that parenting behaviors centering around medical domains may be different than general parenting behaviors, and it may be important for future research to separate medically- and non-medically-centered parenting behaviors. For example, a parent may exhibit higher levels of psychological control and lower levels of warmth if a child is resistant, for example, to maintaining their catheterization schedule during their scheduled catheterization time. During other times, this parent may exhibit lower levels of psychological control and higher levels of warmth. Assessments of parenting behaviors specific to adherence to medical regimen may be helpful in improving the understanding of the complex, transactional relationships between youth with SB and their caregivers.

Given the null results when using the independent parent variables to predict youth HRQOL and the more surprising results that, when using youth report of HRQOL, none of the covariates (age, SES, IQ, and illness severity) were significantly related to (cross-sectionally) or significantly predictive of (longitudinally) youth-reported HRQOL, the validity of the measure used to assess youth HRQOL is called into question. These null results may have been attributable to lack of variability, lack of stability, or a ceiling effect for the HRQOL variable. However, closer analysis of its psychometric properties found that this variable was normally distributed and showed adequate variability between time points. Though the psychometrics of this variable are acceptable, the PedsQL has not been validated in this population. It is possible that this assessment of HRQOL may not be the “best” assessment of HRQOL for youth with SB. In fact, items from the full PedsQL (specifically, the 8-item physical subscale) were not included in this study due to the physical limitations imposed by SB. Therefore, an instrument tailored specifically to individuals with SB would likely be a better assessment of this construct in this population.

Though in the last decade there has been a dramatic increase in the number of measures of HRQOL in pediatric populations (Drotar, 2004), the measurement of HRQOL has also presented a number of methodological challenges. HRQOL is a multidimensional, abstract, and complex construct. These qualities make it difficult to describe and, therefore, difficult to assess. There are clear benefits to using a general measure of HRQOL (such as the PedsQL). Specifically, using a general measure of the construct allows for comparison of HRQOL across illness groups. Though many chronic illnesses share common features (such as family conflict, fatigue, pain, stigmatization by peers, and financial burden), specific illnesses also have unique characteristics that may not be adequately assessed by a generic measure. SB is one such illness that has effects that may not be captured by a general assessment of HRQOL. SB is a congenital disorder with a chronic course. Youth with SB experience a chronic type of stress due to the daily struggles of a complex medical regimen involving multiple domains, including managing limited mobility and bowel and bladder routines.

Therefore, the current approach to assessing HRQOL in youth with SB may be problematic. HRQOL instruments developed for healthy children or children with other chronic illnesses (e.g., diabetes) may not the capture small but clinically important differences in this population because they are not designed to measure the impact of SB on HRQOL. Further, there are no validated SB-specific instruments assessing HRQOL that include bladder and bowel domains and mobility assessments, which have been found to greatly impact constructs such as parenting stress in this population, and perhaps also impact youth-reported HRQOL. The need for a SB-specific HRQOL questionnaire has been recognized, and recently (September 2015, January 2016), two new assessments of HRQOL in this population were developed, validated, and published (Szymanski et al., 2015; Velde et al., 2016). The use of these instruments will likely improve the assessment of HRQOL in this population.

Given the impact that decreased mobility and bowel and bladder management have on parenting stress (Kanaheswari, Razak, & Ong, 2011), it is possible that assessments of HRQOL including these domains (such as the Spina Bifida Pediatric Questionnaire (SBPG; Velde et al., 2016), or Quality of Life Assessment in Spina Bifida for Children (QUALAS-C; Szymanski et al., 2015)) may better allow for the detection of a relationship between parenting constructs and youth HRQOL. In fact, the exploratory analyses using parent-proxy reports of HRQOL highlight the potential importance of assessing these SB-specific factors. For both mothers and fathers, higher levels of SB-specific parenting stress was the best predictor of proxy-report of decreased youth HRQOL. Though questions on the psychosocial subscale of the PedsQL did not specifically mention mobility or bowel/bladder management, it is possible that parents (but not youth) considered these daily struggles when responding to these questions. It is also possible that parents may have been better able to understand the impact that SB has on their child's overall functioning, and, therefore, more successfully translated daily stressors these youth experience (that TD youth do not) into their report of HRQOL. It was surprising that illness-severity was not significantly related to youth or parent-proxy reports of youth HRQOL. However, this study's assessment of illness-severity did not include questions concerning bowel and bladder functioning. It is possible that bowel and bladder dysfunction is the illness-related factor most impactful on HRQOL. The significant association between SB-specific parenting stress (but not illness-severity) and parent proxy-report of youth HRQOL highlights the importance of including the bowel/bladder domain when assessing HRQOL in youth with SB.

In addition to issues with the actual domains on the measure used to assess HRQOL, youth with SB may have had difficulty understanding and interpreting items on an HRQOL questionnaire. Individuals with SB, specifically those with hydrocephalus, often display cognitive deficits, including difficulties with language, attention, executive functions, and memory (Yeates, Fletcher, & Dennis, 2016). These cognitive limitations may have impaired youth's ability to complete study questionnaires, including the PedsQL. In addition to impaired cognitive abilities, youth with SB often exhibit difficulties with social functioning, including poor social competence (Lennon et al., 2015). Completion of the PedsQL requires the skill of perspective taking, as many of the items require youth to compare themselves to same-age peers (e.g., “I cannot do things that other kids my age can do.”). Youth with SB, given their deficits in both cognitive and social functioning, may be unable to make the social comparisons necessary to complete these items validly. It is also important to note the ages of participants in this study (ranging from 8 to 15 years of age) when considering the validity of and ability to interpret an assessment of HRQOL. Though the PedsQL has been normed and validated in youth this young (Varni, Seid, and Kurtin, 2001), it still may be difficult for children to engage in appropriate perspective taking. It is possible that the young age of participants and cognitive and social limitations of youth with SB may account for the discrepancies in self- and parent proxy-report of HRQOL. It is also possible that the proxy-report of HRQOL for youth, which has been found to be consistently lower than self-report, may be a more accurate assessment of youth HRQOL in this population. The questionable validity of this assessment may have undermined the possibility of finding significant associations between parent factors and youth-report of HRQOL in the current study.

Another explanation of the null results of this study is that parent factors may not be the most important factors to consider when assessing HRQOL in youth with SB. It is possible that social functioning may have been a better predictor of HRQOL for these youth. Previous research has found that youth with SB have significant social difficulties. Researchers have found that youth with SB are at risk for social immaturity and having fewer, poorer quality friendships (Blum, Resnick, Nelson, & St. Germaine, 1991; Ellerson, Stewart, Ritchie, & Hirth, 1996; Devine, Holmbeck, Gayes, & Purnell, 2012).

Peer relationships and friendships are important to healthy development. However, disease management (i.e., doctor’s visits) may interfere with school attendance and the development of stable peer relationships (Olsson, Boyce, & Toumbourou, 2005). Results from Murray’s (2013) study suggest that social adjustment may significantly affect HRQOL in youth with SB. Despite increased time spent with and reliance on family members, it is possible that the impact of social relationships/friendships is greater than the impact of family relationships on youth HRQOL, specifically in the domains of quality of life that are assessed with the psychosocial subscale of the PedsQL (emotional, social, and school functioning).

Beyond conceptual, theoretical, and measurement issues influencing the findings of the current study, statistical factors may also account for study findings. The analyses conducted in this study were fairly conservative. First, HRQOL and parenting behaviors were controlled at earlier time points, thus eliminating some of the variance in the dependent and mediating variables. The change in HRQOL over time may not have been large enough to yield significant variability in the residuals that remained after controlling for previous levels of HRQOL. Analyses were even more conservative given the utilization of different reporters across dependent and independent measures (excluding the exploratory analyses), as well as the use of observational methods, which eliminated the possibility of common method variance in findings. Taken together, this conservative study design limited the possibility of significant findings.

Strengths, Limitations, and Future Research

This study had several strengths. First, the current study sought to expand the limited knowledge on modifiable factors affecting HRQOL in youth with SB. Second, the current study used multiple methods and reporters, which has been encouraged within research in general, and the field of SB research specifically (Holmbeck et al., 2006). The exploratory analyses using parent proxy-report of youth HRQOL demonstrated the variability that can exist among different reporters. Third, longitudinal data was used to examine relationships over time, which allows for consideration of developmental changes in childhood and adolescence as well as the ability to support causal conclusions. Fourth, the study included father-report. It cannot be assumed that all caregivers (mothers and fathers) experience their role as caretakers identically, and it is important to include fathers in research studies so that these potential differences can be better understood.

However, there are several limitations of the current study that should be addressed in future work. First, the current study used the PedsQL to asses HRQOL in youth with SB. This measure has not been normed in this population specifically. Due to the limited mobility of many youth with SB, the physical subscale of this measure was not used as the items were deemed inappropriate for these youth to complete. It is possible that a SB-specific measure of youth HRQOL would be more appropriate for assessment of this construct in this population. Second, attrition at Times 2 and 3 in this study should be considered. Though attrition analyses revealed no significant differences in demographic factors or in youth-reported HRQOL among full- and partial-participants, it is possible that the families with parents experiencing the most distress and stress did not participate at future time points. Third, the time between study time points (2 years between each time point; 4 years total) may have been too long to be predictive in this case. Many of the youth included in the study may have gone through significant transitions or developmental changes between visits. Additionally, SB is an illness with many life-threatening illness-related complications that could have a quick or sudden onset (UTIs, shunt malfunctions). Therefore, parent factors at Time 1 may not be predictive of youth factors at subsequent time points with a two-year interval.

Conclusions and Clinical Implications

The results of the current study have important implications for work with families of youth with SB. First, though youth with SB are at-risk for poor HRQOL, it appears that parent factors or demographic factors may not significantly impact HRQOL in this population. Given the consistently lower HRQOL of youth with SB and the potentially important role HRQOL plays in adherence and disease management, it is of the utmost importance that factors that *do* affect HRQOL (i.e., social factors) in this population be identified. While the mediation models were not significant, parent factors were found to affect parenting behaviors (maternal social isolation and maternal acceptance; paternal SB-specific parenting stress and paternal psychological control). Interventions targeted to alleviate parenting stress and distress in this population could have clinically significant effects for not only parents, but youth with SB as well. Finally, special consideration should be given when choosing an instrument to assess HRQOL in this population. When possible, SB-specific instruments that include bowel/bladder domains (such as the Spina Bifida Pediatric Questionnaire (SBPG; Velde et al., 2016), or Quality of Life Assessment in Spina Bifida for Children (QUALAS-C; Szymanski et al., 2015) should be used. These instruments may more adequately and comprehensively assess HRQOL in youth with SB than a general measure (e.g., PedsQL).

APPENDIX A

MEASURES

FSS-MM

The following is a list of things that may be stressful when raising a child with spina bifida. We would like you to think of stress as meaning something that taxes your resources, or as something that is more than you can handle comfortably. Please rate the stressfulness of each item on the scale below:

1 = not at all stressful
2 = a bit stressful
3 = fairly stressful
4 = quite stressful
5 = extremely stressful

	Not at all stressful	A bit stressful	Fairly stressful	Quite stressful	Extremely stressful
1. Outings in the community	1	2	3	4	5
2. Relationships with our friends or extended family.	1	2	3	4	5
3. Discipline.	1	2	3	4	5
4. My marital/intimate relationship.	1	2	3	4	5
5. Mealtimes and bedtimes.	1	2	3	4	5
6. Educational concerns.	1	2	3	4	5
7. Safety.	1	2	3	4	5
8. Communication with my child.	1	2	3	4	5
9. My child's relations with other children.	1	2	3	4	5

	Not at all stressful	A bit stressful	Fairly stressful	Quite stressful	Extremely stressful
10. My child's behavior problems.	1	2	3	4	5
11. My child's emotional problems.	1	2	3	4	5
12. My child's relationships with his/her brother(s) and sister(s).	1	2	3	4	5
13. Financial responsibilities.	1	2	3	4	5
14. Medical care/appointments.	1	2	3	4	5
15. Catheterization.	1	2	3	4	5
16. Medications.	1	2	3	4	5
17. Bowel program.	1	2	3	4	5
18. Food/diet.	1	2	3	4	5
19. Braces/wheelchair/ambulation.	1	2	3	4	5

Medical History Questionnaire

1. What type of spina bifida does your child have?

    _____ Lipomeningocele (lipo)
    _____ Myelomeningocele (MM)
    _____ Other Please specify: ______________________
    _____ Not sure

2. What is the level of your child's lesion?

    _____ Sacral
    _____ Lumbar
    _____ Thoracic
    _____ Not sure

3. Does your child have a shunt? yes _____ no _____

    a. IF YES, has your child's shunt been infected? yes _____ no _____
    b. IF YES, have you child had a shunt revision? yes _____ no _____
    c. IF your child's SHUNT HAS BEEN INFECTED, how many times? _____
    d. IF your child has had a SHUNT REVISION, how many times? _____

4. Does your child have seizures or take medication to prevent seizures?
    yes _____ no _____

5. Is your child able to do independent toileting?
    yes _____ no _____

6. Is your child on a catheterization schedule? yes _____ no _____

    a. If YES, does your child do the catheterization (check one)?

        _____ independently without reminding
        _____ independently with reminding
        _____ with partial assistance
        _____ with complete assistance

    b. Has your child ever had a bladder or urinary tract infection? yes _____ no _____

    c. How many times has your child had a bladder or urinary tract infection? _______

    d. Has your child had bladder stimulation? yes _____ no_____

7. Is your child on a bowel program?
yes _____ no _____

a. If YES, what type of bowel program (suppositories, diet, enemas, digital manipulation, etc.)?
____
____
____

b. If YOUR CHILD IS ON A BOWEL PROGRAM, do your child do this program (check one)?
_____ independently without reminding
_____ independentl y with reminding
_____ with partial assistance
_____ with complete assistance

c. Has your child had bowel stimulation? yes ____ no ____

8. Does your child use diapers? yes _____ no _____

a. If YES, where does your child use diapers (please check all that apply)?
_____ school
_____ home
_____ on outings
_____ all the time
_____ other? ______________________________

9. Does your child use braces? yes _____ no _____

a. If YES, what type (please check all that apply)?
_____ ankle-foot
_____ knee-ankle-foot
_____ hip-knee-ankle-foot
_____ reciprocating brace
_____ full control brace
_____ swivel walker
_____ parapodium
_____ twister cables
_____ night splint
_____ back brace

10. Does your child use crutches? yes _____ no _____

11. Does your child use a walker? yes _____ no _____

a. If YES, where does your child use a walker (please check all that apply)?
_____ school
_____ home
_____ for long distance walking
_____ on outings
_____ all the time
_____ other? ____

12. Does your child use a wheelchair? yes _____ no _____

a. If YES, where does your child use a wheelchair (please check all that apply)?
_____ school
_____ home
_____ for long distance travel
_____ on outings
_____ all the time
_____ other? ____

13. If your child uses more than one mobility device, please write down the percentage of time that you use each device (please **make sure** that the percentages add up to 100%):

_____ % unassisted walkin g (no braces)
_____ % braces alone (no crutches or walker)
_____ % braces with crutches or walker
_____ % wheelchair
= 100 %

14. Please list your child's medications (include NAME OF MEDICATION, AMOUNT, HOW OFTEN TAKEN):

	Name of Medication	Amount	How Often Taken?
1.			
2.			
3.			
4.			
5.			
6.			
7.			
8.			
9.			

10. ____________________ __________ ______________________

15. Please list your child's surgeries over the **past two years** (include year of surgery, reason for surgery; examples include: shunt revision, shunt replacement, leg surgery, back surgery, tethered cord, etc.):

	Year of Surgery	Reason for Surgery
1.		
2.		
3.		
4.		
5.		
6.		
7.		
8.		
9.		
10.		
11.		
12.		
13.		
14.		
15.		

16. What changes have occurred in your child's health **OVER THE PAST TWO YEARS**?

17. What types of health insurance does your child have?

____

18. In the past year, how many visits has your child had with a primary care physician (regular family doctor)? ____________________

Please describe the reason(s) for these visits: ______________________________

____

____

19. In the past year, how many visits has your child had with a urologist? _______________

Please describe the reason(s) for these visits: ______________________________

____

____

20. In the past year, how many visits has your child had with an orthopedist? _____________

Please describe the reason(s) for these visits: ______________________________

____

____

21. In the past year, how many visits has your child had with a neurologist? _____________

Please describe the reason(s) for these visits: ______________________________

____

____

22. In the past year, how many visits has your child had with a physical or occupational therapist? (please specify which one)

____

Please describe the reason(s) for these visits: ______________________________

____

____

23. In the past year, on how many occasions has your child visited the emergency room? ______

Please describe the reason(s) for these visits: ______________________________

____

____

24. In the past year, how many visits has your child had with any other type of health care professional? ____

Type of health professional seen: ______________________________

____

Please describe the reason(s) for these visits: ______________________________

____

____

25. In the past year, how many times has your child been hospitalized? ______________

Length of stay ____

Please describe the reason(s) for these hospitalizations: ______________

____

____

26. In the past 6 months, has your child had any injuries that required a visit to a doctor, other medical professional, clinic, or emergency room? (Circle one) YES NO

If yes, how many injuries? ______

27. In the past month, has your child had any minor injuries or accidents that left a visible mark, but did not require a trip to the doctor? (Circle one) YES NO

If yes, approximately how many accidents or injuries? ______

28. In the last month, did your child have any close calls in which he/she might have been injured, but was not? (Circle one) YES NO

If yes, approximately how many times did this occur? ______

PSI

These questions ask you to record an answer which best describes your feelings about being a parent to the child discussed in this questionnaire. While you may not find an answer which exactly states your feelings, please record the answer which comes closest to describing how you feel. **YOUR FIRST REACTION TO EACH QUESTION SHOULD BE YOUR ANSWER.**

Please record the degree to which you agree or disagree with the following statements by filling in the number which best describes how you feel.

Strongly Disagree	Disagree	Agree	Strongly Agree
1	2	3	4

_____ 1. When my child came home from the hospital, I had doubtful feelings about my ability to handle being a parent.

_____ 2. Being a parent is harder than I thought it would be.

_____ 3. I feel capable and on top of things when I am caring for my child.

_____ 4. I can't make decisions without help.

_____ 5. I have had many more problems raising children than I expected.

_____ 6. I enjoy being a parent.

_____ 7. I feel that I am successful most of the time when I try to get my child to do or not do something.

_____ 8. Since I brought my child home from the hospital, I find that I am not able to take care of this child as well as I thought I could. I need help.

_____ 9. I often have the feeling that I cannot handle things very well.

_____ 10. Most of my life is spent doing things for my child.

_____ 11. I find myself giving up more of my life to meet my children's needs than I ever expected.

_____ 12. I feel trapped by my responsibilities as a parent.

_____ 13. I often feel that my child's needs control my life.

_____ 14. Since having this child, I have been unable to do new and different things.

19

PSI

Strongly Disagree	Disagree	Agree	Strongly Agree
1	2	3	4

______ 15. Since having this child, I feel that I am almost never able to do things that I like to do.

______ 16. It is hard to find a place in our home where I can go to be by myself.

______ 17. I feel alone and without friends.

______ 18. When I go to a party, I usually expect not to enjoy myself.

______ 19. I am not as interested in people as I used to be.

______ 20. I often have the feeling that other people my own age don't particularly like my company.

______ 21. When I run into a problem taking care of my children, I have a lot of people I can talk to for help or advice.

______ 22. Since having children, I have a lot fewer chances to see my friends and to make new friends.

23. When I think about myself as a parent, I believe (please circle one):

A) I can handle anything that happens.
B) I can handle most things pretty well.
C) sometimes I have doubts, but I find that I handle most things without any problems.
D) I have some doubts about being able to handle things.
E) I don't think I handle things very well at all.

24. I feel that I am (please circle one):

A) a very good parent.
B) a better than average parent.
C) an average parent.
D) a person who has some trouble being a parent.
E) not very good at being a parent.

Peds QL – Child

In the past **ONE month**, how much of a **problem** has this been for you . . .

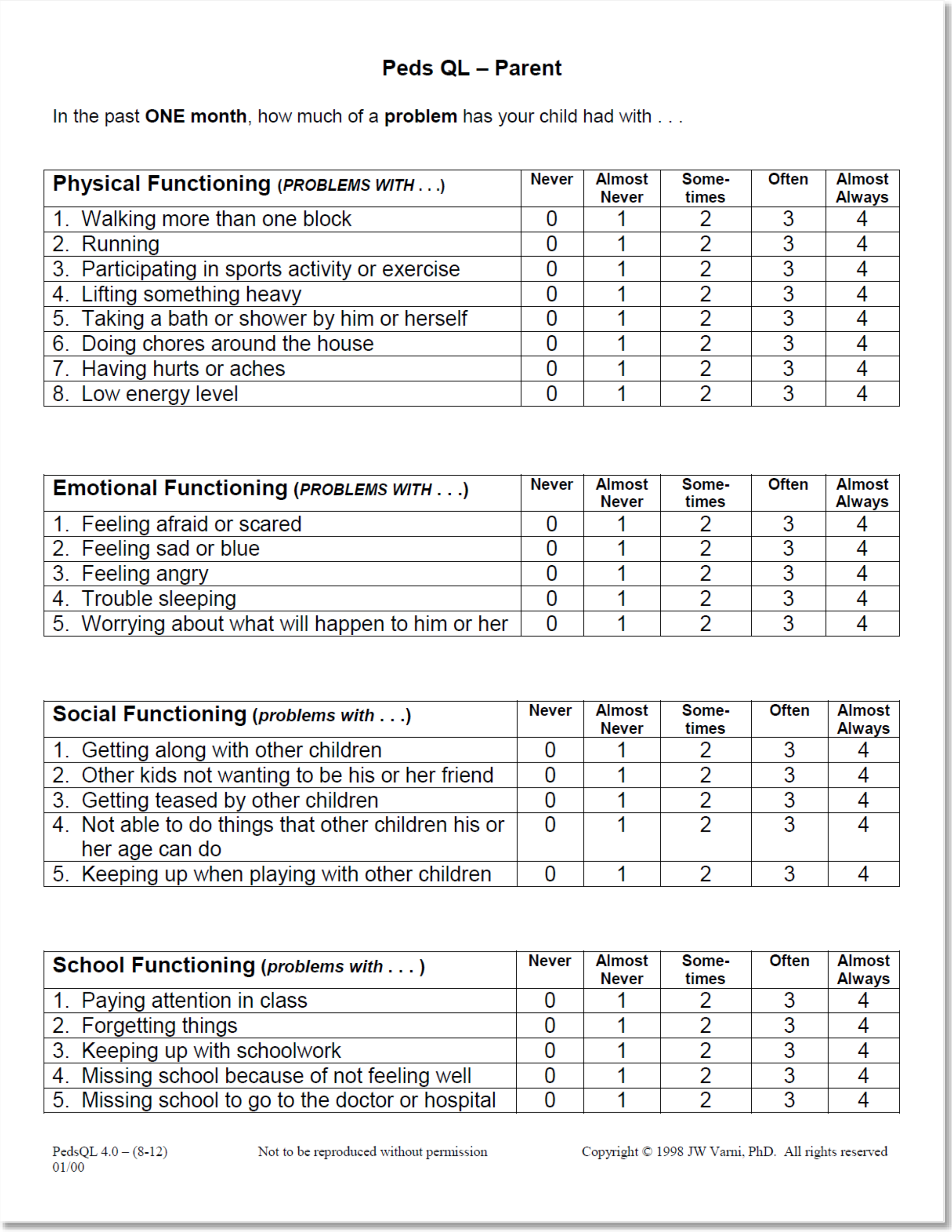
About My Health and Activities (***PROBLEMS WITH . . .***)	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. It is hard for me to walk more than one block	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activity or exercise	0	1	2	3	4
4. It is hard for me to lift something heavy	0	1	2	3	4
5. It is hard for me to take a bath or shower by myself	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I hurt or ache	0	1	2	3	4
8. I have low energy	0	1	2	3	4

About My Feelings (***PROBLEMS WITH . . .***)	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad or blue	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

How I Get Along with Others (***problems with . . .***)	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. I have trouble getting along with other kids	0	1	2	3	4
2. Other kids do not want to be my friend	0	1	2	3	4
3. Other kids tease me	0	1	2	3	4
4. I cannot do things that other kids my age can do	0	1	2	3	4
5. It is hard to keep up when I play with other kids	0	1	2	3	4

About School (***problems with . . .***)	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. It is hard to pay attention in class	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my schoolwork	0	1	2	3	4
4. I miss school because of not feeling well	0	1	2	3	4
5. I miss school to go to the doctor or hospital	0	1	2	3	4

PedsQL 4.0 – (8-12)
01/00



Peds QL – Parent

In the past **ONE month**, how much of a **problem** has your child had with . . .

Physical Functioning (*PROBLEMS WITH . . .*)	Never	Almost Never	Some-times	Often	Almost Always
1. Walking more than one block	0	1	2	3	4
2. Running	0	1	2	3	4
3. Participating in sports activity or exercise	0	1	2	3	4
4. Lifting something heavy	0	1	2	3	4
5. Taking a bath or shower by him or herself	0	1	2	3	4
6. Doing chores around the house	0	1	2	3	4
7. Having hurts or aches	0	1	2	3	4
8. Low energy level	0	1	2	3	4

Emotional Functioning (*PROBLEMS WITH . . .*)	Never	Almost Never	Some-times	Often	Almost Always
1. Feeling afraid or scared	0	1	2	3	4
2. Feeling sad or blue	0	1	2	3	4
3. Feeling angry	0	1	2	3	4
4. Trouble sleeping	0	1	2	3	4
5. Worrying about what will happen to him or her	0	1	2	3	4

Social Functioning (*problems with . . .*)	Never	Almost Never	Some-times	Often	Almost Always
1. Getting along with other children	0	1	2	3	4
2. Other kids not wanting to be his or her friend	0	1	2	3	4
3. Getting teased by other children	0	1	2	3	4
4. Not able to do things that other children his or her age can do	0	1	2	3	4
5. Keeping up when playing with other children	0	1	2	3	4

School Functioning (*problems with . . .*)	Never	Almost Never	Some-times	Often	Almost Always
1. Paying attention in class	0	1	2	3	4
2. Forgetting things	0	1	2	3	4
3. Keeping up with schoolwork	0	1	2	3	4
4. Missing school because of not feeling well	0	1	2	3	4
5. Missing school to go to the doctor or hospital	0	1	2	3	4

PedsQL 4.0 – (8-12)
01/00

SCL-90-R

Below is a list of problems people sometimes have. Please read each one carefully, and circle the number to the right that best describes HOW MUCH THAT PROBLEM HAS DISTRESSED OR BOTHERED YOU DURING THE PAST 7 DAYS INCLUDING TODAY.

Circle only one number for each problem and do not skip any items.

DURING THE PAST 7 DAYS, HOW MUCH WERE YOU DISTRESSED BY:	NOT AT ALL	A LITTLE BIT	MODERATELY	QUITE A BIT	EXTREMELY
1. Headaches	0	1	2	3	4
2. Nervousness or shaking inside	0	1	2	3	4
3. Repeated unpleasant thoughts that won't leave your mind	0	1	2	3	4
4. Faintness or dizziness	0	1	2	3	4
5. Loss of sexual interest or pleasure	0	1	2	3	4
6. Feeling critical of others	0	1	2	3	4
7. The idea that someone else can control your thoughts	0	1	2	3	4
8. Feeling others are to blame for most of your troubles	0	1	2	3	4
9. Trouble remembering things	0	1	2	3	4
10. Worried about sloppiness or carelessness	0	1	2	3	4
11. Feeling easily annoyed or irritated	0	1	2	3	4
12. Pains in heart or chest	0	1	2	3	4
13. Feeling afraid in open spaces or on the streets	0	1	2	3	4

DURING THE PAST 7 DAYS, HOW MUCH WERE YOU DISTRESSED BY:	NOT AT ALL	A LITTLE BIT	MODERATELY	QUITE A BIT	EXTREMELY
14. Feeling low in energy or slowed down	0	1	2	3	4
15. Thoughts of ending your life	0	1	2	3	4
16. Hearing voices that other people do not hear	0	1	2	3	4
17. Trembling	0	1	2	3	4
18. Feeling that most people cannot be trusted	0	1	2	3	4
19. Poor appetite	0	1	2	3	4
20. Crying easily	0	1	2	3	4
21. Feeling shy or uneasy with the opposite sex	0	1	2	3	4
22. Feelings of being trapped or caught	0	1	2	3	4
23. Suddenly scared for no reason	0	1	2	3	4
24. Temper outbursts that you could not control	0	1	2	3	4
25. Feeling afraid to go out of your house alone	0	1	2	3	4
26. Blaming yourself for things	0	1	2	3	4
27. Pains in lower back	0	1	2	3	4
28. Feeling blocked in getting things done	0	1	2	3	4
29. Feeling lonely	0	1	2	3	4
30. Feeling blue	0	1	2	3	4

DURING THE PAST 7 DAYS, HOW MUCH WERE YOU DISTRESSED BY:	NOT AT ALL	A LITTLE BIT	MODERATELY	QUITE A BIT	EXTREMELY
31. Worrying too much about things	0	1	2	3	4
32. Feeling no interest in things	0	1	2	3	4
33. Feeling fearful	0	1	2	3	4
34. Your feelings being easily hurt	0	1	2	3	4
35. Other people being aware of your private thoughts	0	1	2	3	4
36. Feeling others do not understand you or are unsympathetic	0	1	2	3	4
37. Feeling that people are unfriendly or dislike you	0	1	2	3	4
38. Having to do things very slowly to insure correctness	0	1	2	3	4
39. Heart pounding or racing	0	1	2	3	4
40. Nausea or upset stomach	0	1	2	3	4
41. Feeling inferior to others	0	1	2	3	4
42. Soreness of your muscles	0	1	2	3	4
43. Feeling that you are watched or talked about by others	0	1	2	3	4
44. Trouble falling asleep	0	1	2	3	4
45. Having to check and double-check what you do	0	1	2	3	4
46. Difficulty making decisions	0	1	2	3	4
47. Feeling afraid to travel on buses, subways, or trains	0	1	2	3	4

DURING THE PAST 7 DAYS, HOW MUCH WERE YOU DISTRESSED BY:	NOT AT ALL	A LITTLE BIT	MODERATELY	QUITE A BIT	EXTREMELY
64. Awakening in the early morning	0	1	2	3	4
65. Having to repeat the same actions such as touching, counting, or washing	0	1	2	3	4
66. Sleep that is restless or disturbed	0	1	2	3	4
67. Having urges to break or smash things	0	1	2	3	4
68. Having ideas or beliefs that others do not share	0	1	2	3	4
69. Feeling very self-conscious with others	0	1	2	3	4
70. Feeling uneasy in crowds, such as shopping or at a movie	0	1	2	3	4
71. Feeling everything is an effort	0	1	2	3	4
72. Spells of terror or panic	0	1	2	3	4
73. Feeling uncomfortable about eating or drinking in public	0	1	2	3	4
74. Getting into frequent arguments	0	1	2	3	4
75. Feeling nervous when you are left alone	0	1	2	3	4
76. Others not giving you proper credit for your achievements	0	1	2	3	4
77. Feeling lonely even when you are with people	0	1	2	3	4
78. Feeling so restless you couldn't sit still	0	1	2	3	4
79. Feelings of worthlessness	0	1	2	3	4

DURING THE PAST 7 DAYS, HOW MUCH WERE YOU DISTRESSED BY:	NOT AT ALL	A LITTLE BIT	MODERATELY	QUITE A BIT	EXTREMELY
80. The feeling that something bad is going to happen to you	0	1	2	3	4
81. Shouting or throwing things	0	1	2	3	4
82. Feeling afraid you will faint in public	0	1	2	3	4
83. Feeling that people will take advantage of you if you let them	0	1	2	3	4
84. Having thoughts about sex that bother you a lot	0	1	2	3	4
85. The idea that you should be punished for your sins	0	1	2	3	4
86. Thoughts and images of a frightening nature	0	1	2	3	4
87. The idea that something serious is wrong with your body	0	1	2	3	4
88. Never feeling close to another person	0	1	2	3	4
89. Feelings of guilt	0	1	2	3	4
90. The idea that something is wrong with your mind	0	1	2	3	4

ID# ________

1. Vocabulary

START

Start Point

Ages 6–8: Item 5
Ages 9–89: Item 9

Reverse Rule

All Ages: Administer Items 1–4 in forward sequence if score of 0 or 1 on Item 5 or 6.

Ages 9–89: Administer Items 5–8 in reverse sequence if score of 0 or 1 on Item 9 or 10.

Discontinue Rule

After 5 consecutive scores of 0

STOP

Stop Point

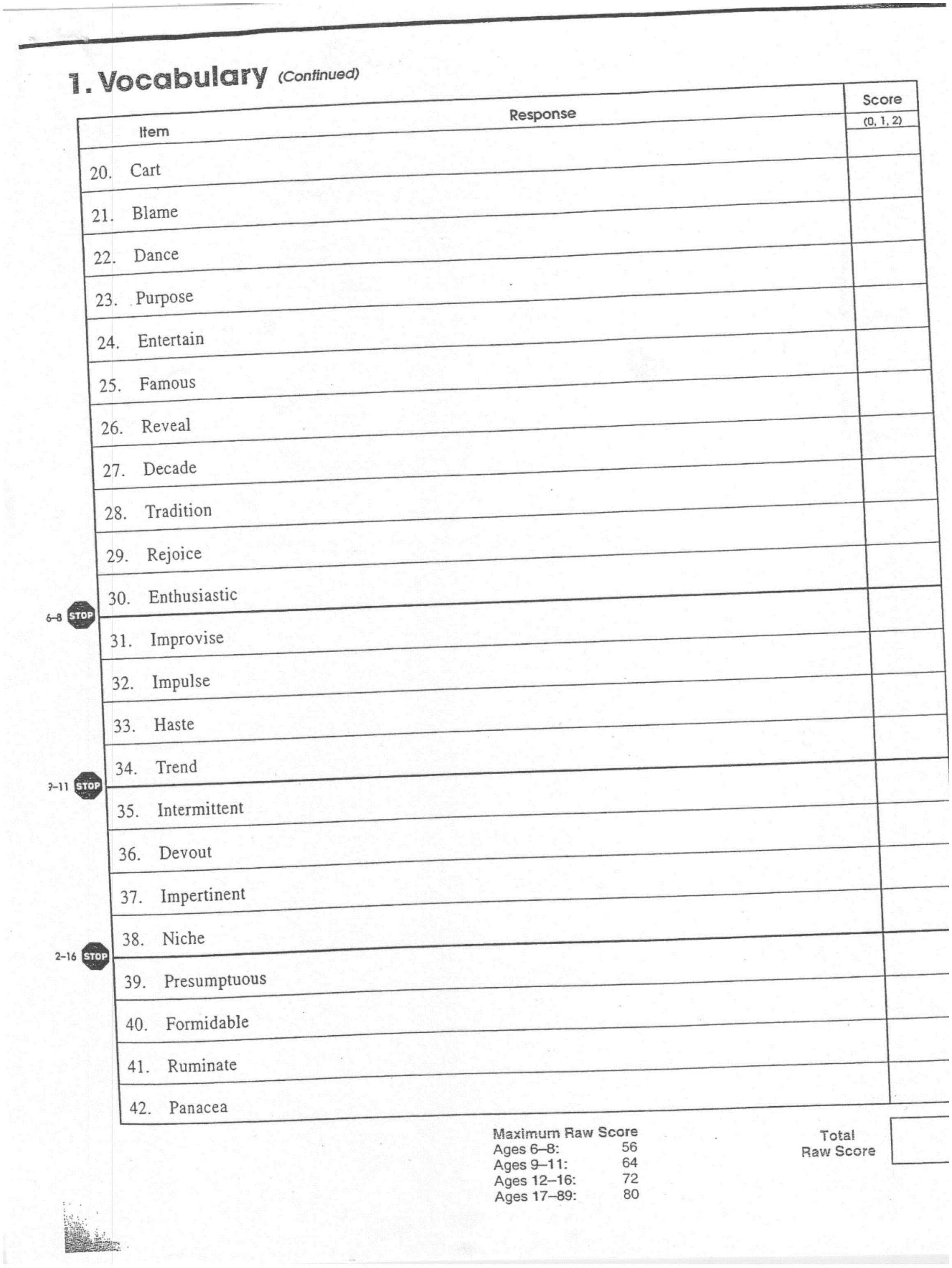
Ages 6–8:	After Item 30
Ages 9–11:	After Item 34
Ages 12–16:	After Item 38
Ages 17–89:	No stop point

Scoring Rule

Items 1–4:	0 or 1
Items 5–42:	0, 1, or 2

Item	Response	Score
		(0 or 1)
1. Fish		
2. Shovel		
3. Map		
4. Shell		
		(0, 1, 2)
6–8 5. Shirt		
6. Shoe		
7. Flashlight		
8. Car		
9–89 9. Bird		
10. Calendar		
11. Number		
12. Bell		
13. Lunch		
14. Police		
15. Vacation		
16. Pet		
17. Balloon		
18. Transform		
19. Alligator		

Continue

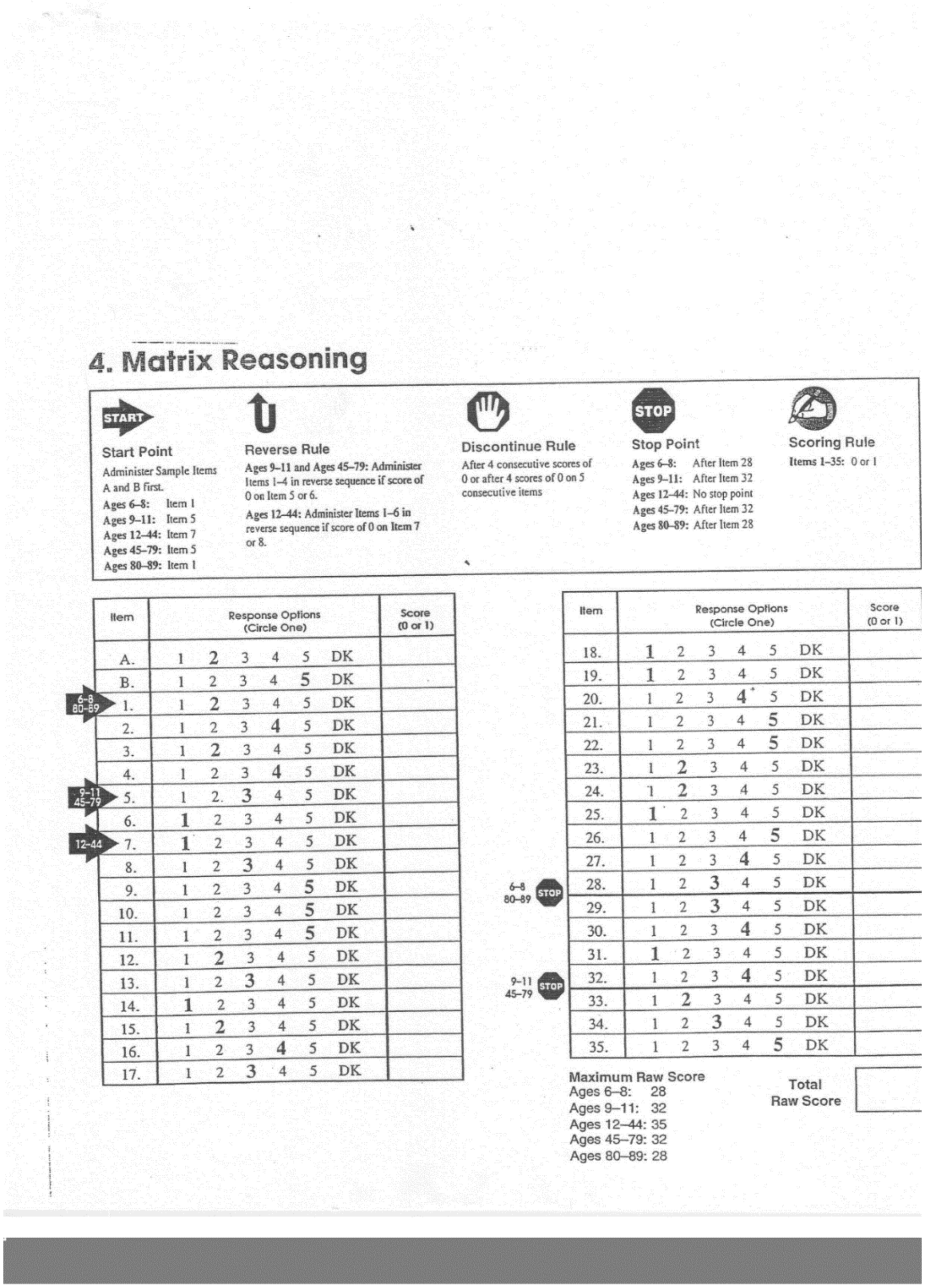


1. Vocabulary *(Continued)*

	Item	Response	Score (0, 1, 2)
	20. Cart		
	21. Blame		
	22. Dance		
	23. Purpose		
	24. Entertain		
	25. Famous		
	26. Reveal		
	27. Decade		
	28. Tradition		
	29. Rejoice		
6–8 STOP	30. Enthusiastic		
	31. Improvise		
	32. Impulse		
	33. Haste		
9–11 STOP	34. Trend		
	35. Intermittent		
	36. Devout		
	37. Impertinent		
2–16 STOP	38. Niche		
	39. Presumptuous		
	40. Formidable		
	41. Ruminate		
	42. Panacea		

Maximum Raw Score	
Ages 6–8:	56
Ages 9–11:	64
Ages 12–16:	72
Ages 17–89:	80

Total Raw Score ☐



4. Matrix Reasoning

Start Point

Administer Sample Items A and B first.

Ages 6–8: Item 1
Ages 9–11: Item 5
Ages 12–44: Item 7
Ages 45–79: Item 5
Ages 80–89: Item 1

Reverse Rule

Ages 9–11 and Ages 45–79: Administer Items 1–4 in reverse sequence if score of 0 on Item 5 or 6.

Ages 12–44: Administer Items 1–6 in reverse sequence if score of 0 on Item 7 or 8.

Discontinue Rule

After 4 consecutive scores of 0 or after 4 scores of 0 on 5 consecutive items

Stop Point

Ages 6–8: After Item 28
Ages 9–11: After Item 32
Ages 12–44: No stop point
Ages 45–79: After Item 32
Ages 80–89: After Item 28

Scoring Rule

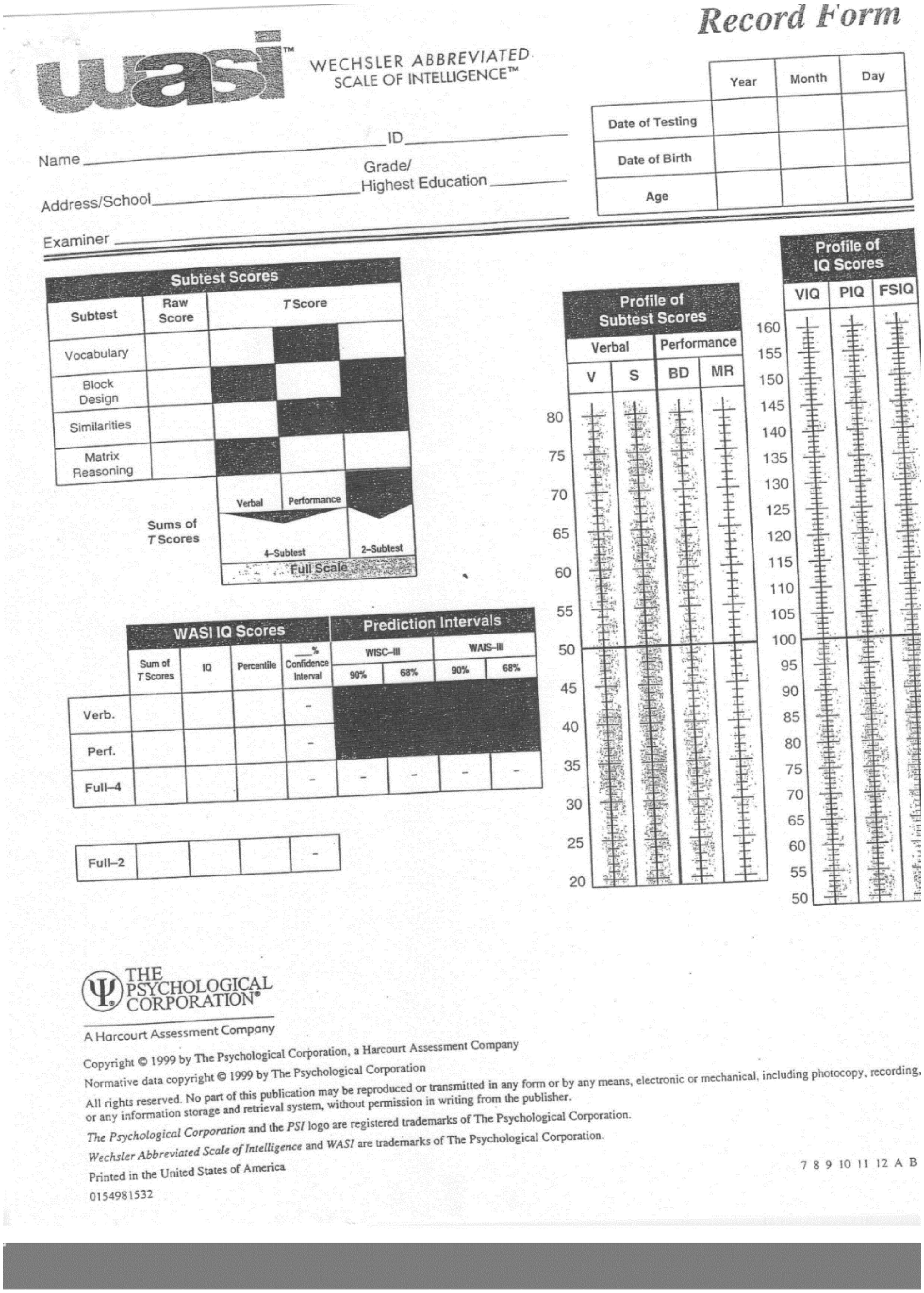
Items 1–35: 0 or 1

	Item	Response Options (Circle One)	Score (0 or 1)
	A.	1 **2** 3 4 5 DK	
	B.	1 2 3 4 **5** DK	
6–8 80–89	1.	1 **2** 3 4 5 DK	
	2.	1 2 3 **4** 5 DK	
	3.	1 **2** 3 4 5 DK	
	4.	1 2 3 **4** 5 DK	
9–11 45–79	5.	1 2. **3** 4 5 DK	
	6.	**1** 2 3 4 5 DK	
12–44	7.	**1** 2 3 4 5 DK	
	8.	1 2 **3** 4 5 DK	
	9.	1 2 3 4 **5** DK	
	10.	1 2 3 4 **5** DK	
	11.	1 2 3 4 **5** DK	
	12.	1 **2** 3 4 5 DK	
	13.	1 2 **3** 4 5 DK	
	14.	**1** 2 3 4 5 DK	
	15.	1 **2** 3 4 5 DK	
	16.	1 2 3 **4** 5 DK	
	17.	1 2 **3** 4 5 DK	

	Item	Response Options (Circle One)	Score (0 or 1)
	18.	**1** 2 3 4 5 DK	
	19.	**1** 2 3 4 5 DK	
	20.	1 2 3 **4** 5 DK	
	21.	1 2 3 4 **5** DK	
	22.	1 2 3 4 **5** DK	
	23.	1 **2** 3 4 5 DK	
	24.	1 **2** 3 4 5 DK	
	25.	**1** 2 3 4 5 DK	
	26.	1 2 3 4 **5** DK	
	27.	1 2 3 **4** 5 DK	
6–8 80–89 STOP	28.	1 2 **3** 4 5 DK	
	29.	1 2 **3** 4 5 DK	
	30.	1 2 3 **4** 5 DK	
	31.	**1** 2 3 4 5 DK	
9–11 45–79 STOP	32.	1 2 3 **4** 5 DK	
	33.	1 **2** 3 4 5 DK	
	34.	1 2 **3** 4 5 DK	
	35.	1 2 3 4 **5** DK	

Maximum Raw Score
Ages 6–8: 28
Ages 9–11: 32
Ages 12–44: 35
Ages 45–79: 32
Ages 80–89: 28

Total Raw Score ☐



Record Form

WASI™ WECHSLER ABBREVIATED SCALE OF INTELLIGENCE™

Name ____________ ID ____________

Address/School ____________ Grade/ Highest Education ____________

Examiner ____________

	Year	Month	Day
Date of Testing			
Date of Birth			
Age			

Subtest Scores

Subtest	Raw Score	*T* Score		
Vocabulary				
Block Design				
Similarities				
Matrix Reasoning				
Sums of *T* Scores		Verbal	Performance	
		4-Subtest		2-Subtest
		Full Scale		

Profile of Subtest Scores

Verbal		Performance	
V	S	BD	MR

80 75 70 65 60 55 50 45 40 35 30 25 20

Profile of IQ Scores

VIQ	PIQ	FSIQ

160 155 150 145 140 135 130 125 120 115 110 105 100 95 90 85 80 75 70 65 60 55 50

	WASI IQ Scores				Prediction Intervals			
					WISC–III		WAIS–III	
	Sum of *T* Scores	IQ	Percentile	___% Confidence Interval	90%	68%	90%	68%
Verb.				–				
Perf.				–				
Full–4				–	–	–	–	–

Full–2				–

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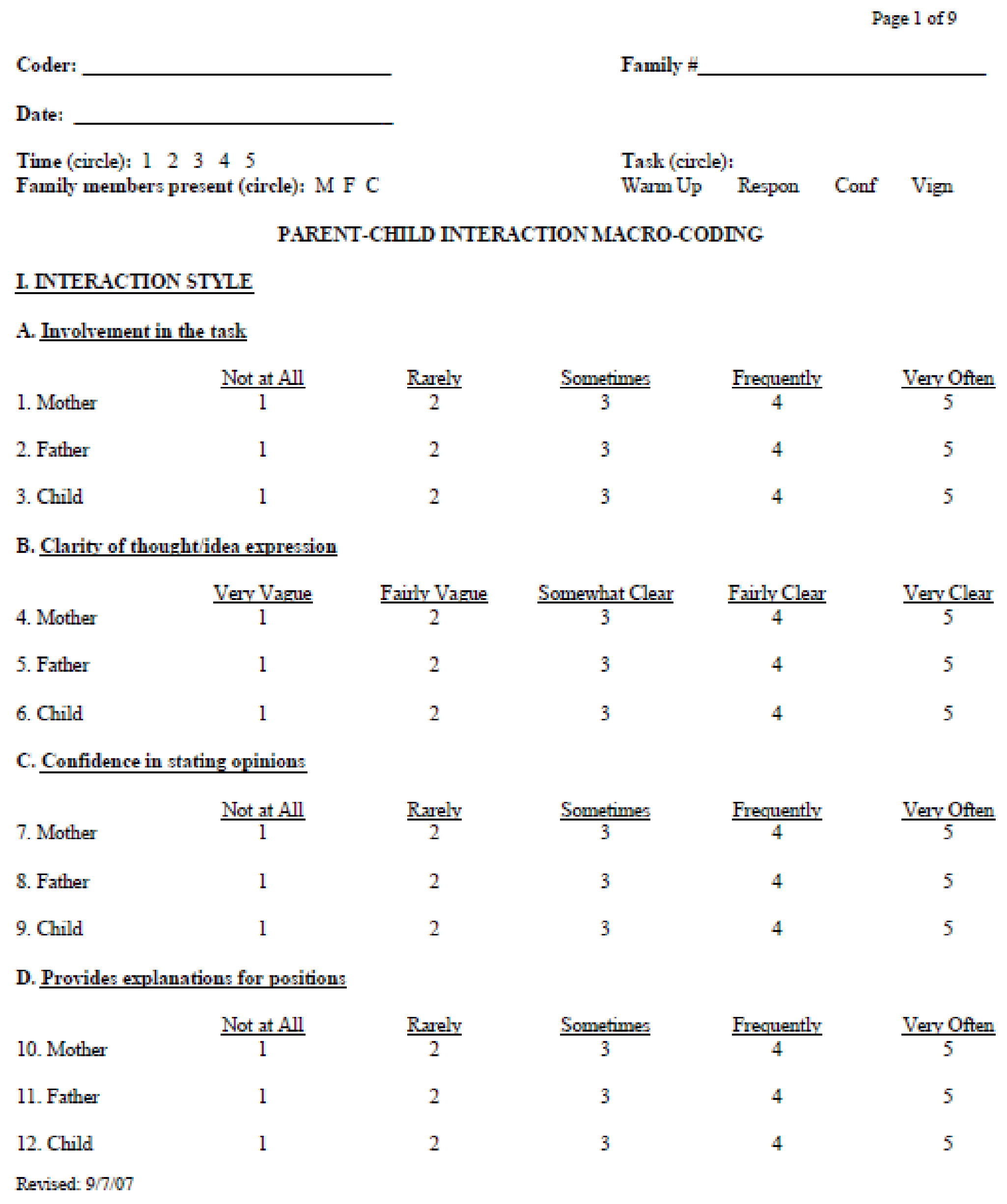


Page 1 of 9

Coder: ____________________ **Family #**____________________

Date: ____________________

Time (circle): 1 2 3 4 5 **Task** (circle):
Family members present (circle): M F C Warm Up Respon Conf Vign

PARENT-CHILD INTERACTION MACRO-CODING

I. INTERACTION STYLE

A. Involvement in the task

	Not at All	Rarely	Sometimes	Frequently	Very Often
1. Mother	1	2	3	4	5
2. Father	1	2	3	4	5
3. Child	1	2	3	4	5

B. Clarity of thought/idea expression

	Very Vague	Fairly Vague	Somewhat Clear	Fairly Clear	Very Clear
4. Mother	1	2	3	4	5
5. Father	1	2	3	4	5
6. Child	1	2	3	4	5

C. Confidence in stating opinions

	Not at All	Rarely	Sometimes	Frequently	Very Often
7. Mother	1	2	3	4	5
8. Father	1	2	3	4	5
9. Child	1	2	3	4	5

D. Provides explanations for positions

	Not at All	Rarely	Sometimes	Frequently	Very Often
10. Mother	1	2	3	4	5
11. Father	1	2	3	4	5
12. Child	1	2	3	4	5

Revised: 9/7/07

E. Requests input from other family members

	Not at All	Rarely	Sometimes	Frequently	Very Often
13. Mother requests input from child	1	2	3	4	5
14. Father requests input from child	1	2	3	4	5
15. Child requests input from mother	1	2	3	4	5
16.Child requests input from father	1	2	3	4	5
17. Mother requests input from father	1	2	3	4	5
18. Father requests input from mother	1	2	3	4	5

F. Listens to others

	Not at All	Rarely	Sometimes	Frequently	Very Often
19. Mother	1	2	3	4	5
20. Father	1	2	3	4	5
21. Child	1	2	3	4	5

G. Off-task behavior

	Not at All	Rarely	Sometimes	Frequently	Very Often
22. Mother	1	2	3	4	5
23. Father	1	2	3	4	5
24. Child	1	2	3	4	5

H. Receptive to statements made by others

	Very Unreceptive	Fairly Unreceptive	Somewhat Receptive	Fairly Receptive	Very Receptive
25. Mother receptive to Child	1	2	3	4	5
26. Father receptive to Child	1	2	3	4	5
27. Child receptive to Mother	1	2	3	4	5
28. Child receptive to Father	1	2	3	4	5
29. Mother receptive to Father	1	2	3	4	5
30. Father receptive to Mother	1	2	3	4	5

I. Attunement

	None	Little	Some	Frequent	Very Often
31. Mother-Child	1	2	3	4	5
32. Father- Child	1	2	3	4	5
33. Mother-Father	1	2	3	4	5

J. Mutuality

	None	Little	Some	Frequent	Very Often
34. Mother-Child	1	2	3	4	5
35. Father- Child	1	2	3	4	5
36. Mother-Father	1	2	3	4	5

K. Positive Escalation

	Not at All	Rarely	Sometimes	Frequently	Very Often
37. Mother-Child	1	2	3	4	5
38. Father- Child	1	2	3	4	5
39. Mother-Father	1	2	3	4	5

L. Maturity

	Not at All	Rarely	Sometimes	Frequently	Very Often
40. Child	1	2	3	4	5

M. Child is Needy

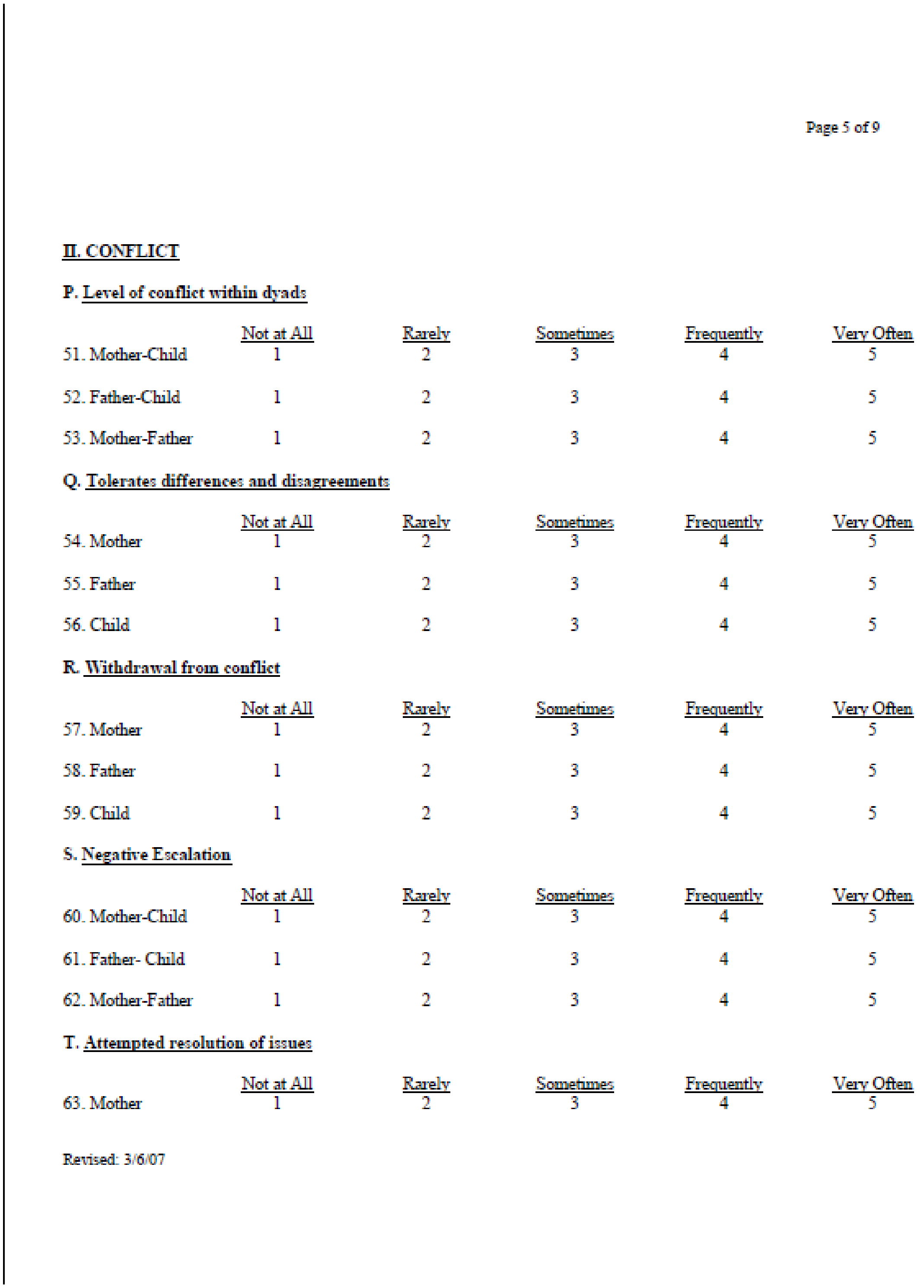
	Not at All	Rarely	Sometimes	Frequently	Very Often
41. Child	1	2	3	4	5

N. Eye Contact

	Not at All	Rarely	Sometimes	Frequently	Very Often
42. Mother	1	2	3	4	5
43. Father	1	2	3	4	5
44. Child	1	2	3	4	5

O. Physical Contact

	Not at All	Rarely	Sometimes	Frequently	Very Often
45. Mother to Child	1	2	3	4	5
46. Father to Child	1	2	3	4	5
47. Child to Mother	1	2	3	4	5
48. Child to Father	1	2	3	4	5
49. Mother to Father	1	2	3	4	5
50. Father to Mother	1	2	3	4	5



II. CONFLICT

P. Level of conflict within dyads

	Not at All	Rarely	Sometimes	Frequently	Very Often
51. Mother-Child	1	2	3	4	5
52. Father-Child	1	2	3	4	5
53. Mother-Father	1	2	3	4	5

Q. Tolerates differences and disagreements

	Not at All	Rarely	Sometimes	Frequently	Very Often
54. Mother	1	2	3	4	5
55. Father	1	2	3	4	5
56. Child	1	2	3	4	5

R. Withdrawal from conflict

	Not at All	Rarely	Sometimes	Frequently	Very Often
57. Mother	1	2	3	4	5
58. Father	1	2	3	4	5
59. Child	1	2	3	4	5

S. Negative Escalation

	Not at All	Rarely	Sometimes	Frequently	Very Often
60. Mother-Child	1	2	3	4	5
61. Father- Child	1	2	3	4	5
62. Mother-Father	1	2	3	4	5

T. Attempted resolution of issues

	Not at All	Rarely	Sometimes	Frequently	Very Often
63. Mother	1	2	3	4	5

64. Father	1	2	3	4	5
65. Child	1	2	3	4	5

III. AFFECT

U. Intensity of positive affect expression/emotionality

	Not at All	Rarely	Sometimes	Frequently	Very Often
66. Mother	1	2	3	4	5
67. Father	1	2	3	4	5
68. Child	1	2	3	4	5

V. Frequency of positive affect

	Not at All	Rarely	Sometimes	Frequently	Very Often
69. Mother	1	2	3	4	5
70. Father	1	2	3	4	5
71. Child	1	2	3	4	5

W. Intensity of negative affect expression/emotionality

	Not at All	Rarely	Sometimes	Frequently	Very Often
72. Mother	1	2	3	4	5
73. Father	1	2	3	4	5
74. Child	1	2	3	4	5

X. Frequency of negative affect

	Not at All	Rarely	Sometimes	Frequently	Very Often
75. Mother	1	2	3	4	5
76. Father	1	2	3	4	5
77. Child	1	2	3	4	5

Y. Warmth

	Not at All	Rarely	Sometimes	Frequently	Very Often
78. Mother	1	2	3	4	5

79. Father	1	2	3	4	5
80. Child	1	2	3	4	5

Z. Supportiveness

	Not at All	Rarely	Sometimes	Frequently	Very Often
81. Mother	1	2	3	4	5
82. Father	1	2	3	4	5
83. Child	1	2	3	4	5

AA. Humor and laughter

	Not at All	Rarely	Sometimes	Frequently	Very Often
84.. Mother	1	2	3	4	5
85. Father	1	2	3	4	5
86. Child	1	2	3	4	5

AB. Anger

	Not at All	Rarely	Sometimes	Frequently	Very Often
87. Mother	1	2	3	4	5
88. Father	1	2	3	4	5
89. Child	1	2	3	4	5

IV. CONTROL

AC. Dominance

	Not at All	Rarely	Sometimes	Frequently	Very Often
90. Mother	1	2	3	4	5
91. Father	1	2	3	4	5
92. Child	1	2	3	4	5

AD. Pressures others to agree

	Not at All	Rarely	Sometimes	Frequently	Very Often
93. Mother	1	2	3	4	5

94. Father	1	2	3	4	5
95. Child	1	2	3	4	5

AE. **Parents present a united front**

	Not at All	Rarely	Sometimes	Frequently	Very Often
96. United Front?	1	2	3	4	5

V. PARENTAL BEHAVIORS AND COLLABORATIVE PROBLEM SOLVING

AF. **Parental promotion of dialogue and collaboration**

	Not at All	Rarely	Sometimes	Frequently	Very Often
97. Mother	1	2	3	4	5
98. Father	1	2	3	4	5

AG. **Parental structuring of task**

	Not at All	Rarely	Sometimes	Frequently	Very Often
99. Mother	1	2	3	4	5
100. Father	1	2	3	4	5

AH. **Promotes Autonomy**

	Not at All	Rarely	Sometimes	Frequently	Very Often
101. Mother	1	2	3	4	5
102. Father	1	2	3	4	5

AI. **Active Catering to the Child**

	Not at All	Rarely	Sometimes	Frequently	Very Often
103. Mother	1	2	3	4	5
104. Father	1	2	3	4	5

AJ. **Parental Behavior that Infantilizes the Child**

	Not at All	Rarely	Sometimes	Frequently	Very Often
105. Mother	1	2	3	4	5

106. Father	1	2	3	4	5

VI. SUMMARY FAMILY MEASURES

AK. Degree of family impairment

	None	Slight	Some	Moderate	Severe
107. Impairment	1	2	3	4	5

AL. General family atmosphere

	Not at All	Rarely	Sometimes	Frequently	Very Often
108. Overly close, stuck, over concerned with each other (enmeshed)	1	2	3	4	5
109. Isolated, disconnected, apathetic towards each other (disengaged)	1	2	3	4	5
110. Depression, sadness, hopelessness	1	2	3	4	5
111. Family is engaged in off-task behavior	1	2	3	4	5
112.Openness, comfortableness, optimism, and warmth	1	2	3	4	5
113. The family is able to reach agreement or resolution	1	2	3	4	5

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VITA

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