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SELF-MANAGEMENT IN YOUTH WITH SPINA BIFIDA: ASSOCIATIONS WITH

PARENT FACTORS IN THE CONTEXT OF A SUMMER CAMP INTERVENTION

A DISSERTATION SUBMITTED TO

THE FACULTY OF THE GRADUATE SCHOOL

IN CANDIDACY FOR THE DEGREE OF

DOCTOR OF PHILOSOPHY

PROGRAM IN CLINICAL PSYCHOLOGY

BY

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CHICAGO, IL

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CHAPTER ONE

INTRODUCTION

Spina bifida (SB) is the most common congenital birth defect of the central nervous system, affecting three of every 10,000 live births in the United States (Centers for Disease Control and Prevention, 2011). Individuals with SB face a unique and complex set of physical, psychosocial, and cognitive challenges. SB is associated with a multitude of medical and orthopedic complications; as such, individuals with SB are required to follow a complex medical regimen throughout their lives. This medical regimen can be challenging to manage independently. The transfer of condition-related responsibilities from caregiver to individual with SB is a major developmental milestone for these individuals.

Although many youth with SB desire autonomy, including independence in managing condition-related responsibilities (e.g., bladder and bowel programs, skin checks; Holmbeck & Devine, 2010), these youth can be slow to acquire this independence (Andren & Grimby, 2004; Holmbeck & Devine, 2010). Few studies have been conducted to identify modifiable risk factors associated with medical autonomy in youth with SB (O'Hara & Holmbeck, 2013; Psihogios, Kolbuck, & Holmbeck, 2015). Parents play an essential role in shaping the home environment for all youth, but especially for youth with SB who are dependent on their parents for both typical *and* medical caregiving. Parent adjustment factors, behaviors, perceptions, and attitudes are identifiable, modifiable factors that have potential to greatly affect the ability of youth with SB to gain and maintain condition-related independence. Therefore, the current study sought to

increase understanding of the relationships among parent personal factors, parental perceptions and attitudes, and youth with SB's condition self-management.

Further, few interventions specific to promoting self-management for individuals with SB exist (Stubberud, Langenbahn, Levine, Stanghelle, & Schanke, 2015). Previous studies have found that a summer camp-based intervention is effective at increasing independence in completing self-management activities for youth and adolescents with SB (Holbein et al., 2013; O'Mahar, Holmbeck, Jandasek, & Zukerman, 2010). Both parents and campers have reported improvements in specific health-related self-care goals up to one month following camp. Qualitatively, parents have reported that their campers' abilities to complete tasks independently and advocate for their own independence improve during this time period following camp. Holbein et al. (2013) found that the effects of the camp intervention were moderated by youth cognitive and health factors as well as SES. Specifically, youth IQ, number of shunt revisions, and family income moderated the changes in health-related self-care tasks experienced by campers. While it is important to understand the contributions of moderating factors to campers' response to the intervention, these specific moderators (youth IQ, number of shunt revisions, SES) are not modifiable. It is imperative to understand modifiable factors that could influence youths' response to the intervention, such as parent factors.

While camp presents a prime opportunity for campers to learn and practice new skills, it is possible that when campers return to their regular home routines, they may regress to their previous level of independence, rather than continuing to move forward. Parents, therefore, play a critical role in ensuring that the gains made at camp are maintained over time. Therefore, an additional purpose of the proposed study was to better understand the potential associations among parent adjustment factors (personal distress, parenting stress, and SB-specific parenting

stress), parental perceptions and attitudes (parental perception of child vulnerability, parental illness uncertainty, parental overprotection, parental optimism, and parental future expectations), and youth with SB's response to a camp-based intervention targeting health-related independence.

The present study had three specific objectives. The *first* objective was to identify cross-sectional associations between the parent variables (including parent personal factors and parent perceptions/attitudes) and youth self-management (both youth responsibility for and youth mastery over condition-related tasks). The *second* objective was to determine the relationships between these parent variables and youth responses over time to the psychosocial camp-based intervention targeting self-management. Specifically, the parent variables were used to predict changes in youth condition self-management goal attainment and youth independence in performing and mastery of medical tasks from Time 1 (pre-intervention) to Time 3 (post-intervention follow-up). The *third* objective was to examine the roles of parent factors in explaining differences in youth self-management trajectories (e.g., changes in self-management over time) for youth who participated in the intervention over two summers. The current study increases understanding of how parent factors can influence both youth self-management behaviors and youth with SB's response to the camp-based intervention. These results can influence future beneficial adaptations of the presented intervention and identify potential targets for future parent-focused interventions for families of youth with SB.

CHAPTER TWO

REVIEW OF THE RELEVANT LITERATURE

Overview of Spina Bifida

Spina bifida (SB) is a congenital birth defect that occurs in the early weeks of gestation, when the neural tube fails to close completely (Copp et al., 2015). SB myelomeningocele is the most common and severe form of SB. With this type of SB, the spinal cord and meningeal membranes protrude through the unfused portion of the spinal column enclosed by a sac. These lesions are closed surgically, either pre- or postnatally (Copp et al., 2015). However, individuals with SB experience a wide range of subsequent difficulties, including deficits in motor, orthopedic, sensory, cognitive, self-care, and social functioning (Zukerman, Devine, & Holmbeck, 2011) as well as the co-occurrence of neurological conditions (e.g., hydrocephalus and Chiari II malformation; Yeates, Fletcher, & Dennis, 2008). The clinical symptoms of SB place considerable physical, psychological, and social demands on both individuals with SB and their families (Greenley, Holmbeck, & Rose, 2006; Holmbeck et al., 2003; Kelly, Zebracki, Holmbeck, & Gershenson, 2008; Singh, 2003). These medical and associated psychosocial issues necessitate lifelong care by a multidisciplinary medical team.

Self-Management for Individuals with SB

SB is associated with a number of complications, including lower-body paralysis, urinary and bowel dysfunction, and hydrocephalus (Copp et al., 2015). These medical issues often require lifelong use of assistive devices for mobility and adherence to medical regimens such as

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bowel and bladder programs. It is not uncommon for the extensive medical regimen of children with SB to include medications, clean intermittent catheterization, bowel programs, skin checks, monitoring of shunt functioning, and use of orthopedic devices (Copp et al., 2015; Sandler, 2010; Zukerman et al., 2011). Adherence to this complex medical regimen is necessary to prevent complications such as urinary tract infections and skin breakdowns (e.g., pressure injuries) that are potentially life-threatening (Copp et al., 2015).

Advances in medical care have increased the life expectancy of individuals with SB, with many living well into adulthood (Oakeshott, Hunt, Poulton, & Reid, 2010). Although parents manage condition-related responsibilities for infants and young children with SB, school-aged children are expected to begin to manage their condition independently (Beacham & Deatrick, 2013; Yun & Kim, 2017). This shift from family management of the condition to self-management has been defined as "an active daily and flexible process in which youth and their parents share responsibility and decision making for achieving control of their condition, health, and well-being through a wide range of activities and skills. The goal of this increasing responsibility is to develop skills needed for transition to adulthood and independent living" (Sawin, Bellin, Roux, Buran, & Brei, 2009, p. 34). The process of transferring primary responsibility for completing medical tasks from caregiver to youth with SB is a typical part of development for these families. Accordingly, research has shown that children with SB assume increasing responsibility for self-management behaviors with increasing age (Psihogios et al., 2015; Stepansky, Roache, Holmbeck, & Schultz, 2010).

Despite this trajectory of increased responsibility, many children with SB are slow to develop self-management independence, with one study finding that 60% of children with SB require moderate or high levels of caregiver assistance to carry out daily condition-related tasks

(Dahl et al., 2000) and another reporting that ~50% of adolescents with SB were unable to complete one common condition-related task (clean intermittent catheterization) independently (Donlau, Mattsson, & Glad-Mattsson, 2012). It is possible that the transfer of responsibility from parent to child with SB occurs slowly due to concerns about adherence, as increased youth condition-related responsibility as been associated with poorer medical adherence (Psihogios et al., 2015). Ongoing parental involvement in medical care may be necessary to achieve optimal condition-related adherence and, subsequently, prevent medical complications. It is also possible that youth with SB lack the executive, cognitive, or fine motor functioning required to complete condition-related tasks independently (Copp et al., 2015). Therefore, parents often remained involved in SB management well into late adolescence and early adulthood. Still, the ultimate goal of the transfer of medical responsibilities from parents to youth with SB is for the affected youth to achieve control over their condition, health, and well-being (Sawin et al., 2009) as well as to grant individuals greater independence in everyday life (e.g., more time spent away from parents outside the home).

A number of factors, including condition severity, personal, and interpersonal factors, have been identified as affecting condition-related behaviors for children and adolescents with SB specifically. Condition severity factors, such as lesion level, motor impairment, and bladder and bowel functioning, have been found to be associated with self-management (Greenley, Coakley, Holmbeck, Jandasek, & Wills, 2006; Hommeyer, Holmbeck, Wills, & Coers, 1999; Yun & Kim, 2017). Individuals with greater condition severity are less likely to independently complete condition-related tasks. Further, a literature review focused on adolescents with heterogenous chronic illnesses identified both personal and interpersonal factors as sources of risk and resilience to condition self-management (Lansing & Berg, 2014). Personal factors in SB,

such as condition-related knowledge (Ievers et al., 1999; Stepansky et al., 2010) and self-efficacy (van Achterberg, Holleman, Cobussen-Boekhorst, Arts, & Heesakkers, 2008) have been found to be associated with adherence to prescribed condition-related tasks. There are also associations between social factors, including family functioning and peer relations, and positive self-management behaviors (Holmbeck & Devine, 2010). Still, more research is necessary to identify *modifiable* factors that affect self-management in this population.

Theoretical Models of Self-Management

In line with Ecological Systems Theory (Bronfenbrenner, 1977), Modi et al. (2012) proposed a comprehensive Pediatric Self-management Model. This model posits that self-management behaviors operate within individual, family, community, and health care system contexts and that all four of these key domains include both modifiable and non-modifiable factors that can influence self-management behaviors. For example, within the individual domain, nonmodifiable influences could include age and gender while modifiable factors could include depressive symptoms, condition-related knowledge, self-efficacy, and confidence (Modi et al., 2012). The Pediatric Self-Management Model is applicable to many childhood chronic health conditions, including SB; research with this population provides evidence for the relationships between individual factors on condition-specific self-management. For example, executive functioning and depressive symptoms have been identified as individual factors that influence self-management for youth with SB (Stern, Driscoll, Ohanian, & Holmbeck, 2018).

The family, community, and health care system domains of the Pediatric Self-management Model also contain modifiable and non-modifiable influences on self-management behaviors. Given the aims of the current study, this discussion will focus primarily on the *family* domain. Family, in this theoretical framework, can implicate caregivers, siblings, and other

extended family members that are identified as part of a child's health-related support system. Family make-up (e.g., single- versus two-parent households), family socioeconomic status, and family race and ethnicity are all non-modifiable family factors that can affect self-management (Modi et al., 2012). Modifiable family factors identified in this theoretical framework include caregiver involvement, family disease and treatment knowledge, family internalizing/ externalizing symptoms, family health beliefs and perceptions, and family interaction variables that can support or hinder illness management. These factors can be attributed to the entire family or to any individual within the family (e.g., a parent) and are proposed to influence self-management behaviors.

A significant strength of the Pediatric Self-Management Model is its generalizability. This model can be and has been applied to a range of pediatric health conditions (e.g., asthma [Klok, Kaptein, Duiverman, & Brand, 2013], diabetes [Spencer, Cooper, & Milton, 2013], cystic fibrosis [Barker & Quittner, 2016], sickle cell disease [Crosby et al., 2016], including SB [Psihogios et al., 2016; Stern et al. 2018]). Further, the four contributing domains (e.g., individual, family, community, healthcare system) are, by definition within the model, broad which allows for a variety of different types of factors to be included within each domain. Additionally, the outcome of "self-management behaviors" included in the model is also given a broad definition in this model as any behavior "conducted by a child or family member… performed in the context of care for the chronic condition" (Modi et al., 2012). This general outcome allows for the Pediatric Self-Management Model to be applied in different contexts to different behaviors and different specific outcomes. It also accounts for *processes* by which modifiable and non-modifiable influences may affect self-management behaviors. While the generalizability of the Pediatric Self-Management Model allows for it to be used widely, this

breadth also introduces limitations. For example, in the context of the current study, specific SB-related influences or outcomes are not included in detail in the model. Additionally, individual parent factors are not mentioned explicitly in this model. Therefore, it is helpful to draw from a theoretical model specific to individuals with SB.

In pediatric nursing research, an alternate but complementary theory of self-management has been described – the Individual and Family Self-Management Theory (IFSMT; Ryan & Sawin, 2009). This theory, specific to individuals with SB, conceptualizes self-management as both an individual and family process consisting of multiple dimensions – context, process, and outcomes. Within the context dimension are condition specific (e.g., complexity of condition and treatment), environmental (e.g., access to care, medical setting, culture, socioeconomic status), and individual and family (e.g., developmental stages, cognitive ability, perspectives) factors. The process dimension of self-management includes knowledge and beliefs (e.g., self-efficacy, outcome expectancy, congruency of individual and family goals), self-regulation skills and abilities (e.g., goal setting, self-monitoring, decision making, planning, emotional control), and social facilitation (e.g., influence, support, collaboration). Lastly, the outcomes dimension contains both proximal (e.g., daily self-management behaviors) and distal (e.g., health status, quality of life) outcomes.

IFSMT proposes that the constructs encompassed by each of these dimensions are relevant to both the affected individual and the individuals' family members (or non-related members of this individual's support team) and that the interactions of these constructs can be especially important in understanding long-term self-management-related outcomes. For example, if an adolescent with SB expects to live independently, including independence in medical care after high school (outcome expectancy, an individual process construct), but they

are unable to acquire a full-time job with benefits (context), they may remain dependent on their parents for medical insurance. Another example: if parents of a child with SB expect that their child should be able to complete clean intermittent catherization independently (parental outcome expectancy, a family process construct), but the child does not believe he or she can complete this process independently (congruency of goals, a family process construct) because of poor fine motor dexterity (context), there may be conflict within the family and the child may become non-adherent to medical recommendations. These hypothetical examples illustrate interactions among the individual and family variables in the IFSMT model which are important in understanding self-management for individuals with SB.

Utilizing the frameworks provided by each of these theories and applying them to SB specifically, a number of *modifiable* parent factors that can influence youth self-management will be explored further in the current study. These parent factors include both parent personal adjustment factors (personal distress, general parenting stress, and SB-specific parenting stress) and parental perceptions/attitudes (parental perception of child vulnerability, parental illness uncertainty, overprotection, optimism, and expectations for the future).

Parents of Youth with Spina Bifida

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). Family relationships are particularly important for youth with SB, since these youth tend to be more socially isolated from their peers than typically developing (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 families (e.g., parents), and, therefore, may be more affected by parent factors than are TD youth. Two types of parent factors will be examined in this study due to their potential influence how youth with SB develop autonomy and self-management skills –parent personal adjustment and parental perceptions/attitudes.

While, theoretically, these individual parent factors may be related to youth self-management behaviors, there is very little research that explores these relationships. This lack of research exists not only in the literature on families of youth with SB, but also in the larger literature focused on families of youth with pediatric health conditions more generally. Below, the available empirical support for inclusion of each parent variable will be presented, including the results of studies using different, potentially-related outcomes, evidence from research with other pediatric medical populations, and research examining related relationships with adult populations.

Parent Personal Adjustment Factors

Personal distress. *Parent personal distress* is operationalized as maladaptive psychological functioning that encompasses several domains of psychological distress (e.g., anxiety, depression, hostility) exhibited by an individual (who is also a parent). 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), and poorer psychological adjustment can influence parents' perceptions of their child with SB. For example, experiences of increased personal distress can affect how mothers of youth with SB perceive and report on their child's well-being (Driscoll, Buscemi, & Holmbeck, 2018).

There is a body of research linking parent psychosocial functioning to child psychosocial functioning, both in general child and in pediatric populations (e.g., Brooks et al., 2016; Weissman et al., 2006). Additionally, findings from research conducted with adult populations can be applied cautiously to children with pediatric conditions. For example, among adults with type 1 and type 2 diabetes, greater depressive symptoms were found to be associated with poorer physical functioning and less adherence to medical regimen (Ciechanowski, Katon, Russo, & Hirsch, 2003; Renn, Feliciano, & Segal, 2011). Similar relationships between increased depressive symptoms and poorer self-management behaviors have been found in adults with coronary artery disease, HIV, cancer, and Parkinson's disease (Katon, 2003). Research also suggests that anxiety in adults with chronic health conditions may actually improve self-management behaviors (Dowson, Kuijer, & Mulder, 2004). However, parental anxiety may hinder self-management behaviors in youth with chronic health conditions, as parental anxiety leads to overprotection (Spada et al., 2012). Therefore, it could be hypothesized that increased parent personal distress would be associated with lower self-management in the families of youth with SB.

General parenting stress. Parents are often faced with balancing many responsibilities, and, therefore, may experience an increased amount of stress (when compared to non-parents). *General parenting stress* is conceptualized as the mental or emotional strain or pressure an individual experiences as a direct result of the demands of being a parent (Deater-Deckard, Chen, & El Mallah, 2015). 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, Chandra, & Ong, 2011).

Increased parenting 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 separately below), there are a number of non-disease-related factors that are associated with increased experiences of stress for parents. For example, ethnic minority parents report significantly higher levels of parenting stress due to societal disadvantages such as lower education levels, lower income, higher rates of unemployment, single parenthood, and assimilation/acculturation (Nomaguchi & House, 2013; Ong, Norshireen, & Chandran, 2010). In one study, these life stressors as well as maternal mental health status and mother-report of child's adaptive skills were proposed to moderate the relationship between having a child with SB and parenting stress in these individuals (Ong et al., 2010).

There is substantial empirical support for the damaging effects of sociological stress on physical health and health behaviors in adults (Thoits, 2010). While this research focuses on individual stress and health behaviors, there are also links between parenting stress and poorer child health outcomes. One study found parenting stress to be associated with childhood obesity as well as difficulty making lifestyle changes to treat childhood obesity (Epstein, Paluch, Gordy, Saelens, & Ernst, 2000; Koch, Sepa, & Ludvigsson, 2008). Therefore, it is possible that increased general parenting stress could interfere with self-management for youth with SB.

SB-specific parenting stress. *SB-specific parenting stress*, the mental or emotional strain an individual experiences as a direct result of parenting a child with SB, is considered to be distinct from general parenting stress (Driscoll et al., 2018). Parenting a child with a longstanding or life-threatening illness or medical condition is uniquely challenging and can negatively affect many aspects of the parent's life, including 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, Pitt, & Mellins, 1990). One qualitative study found that adhering to daily medical regimen and balancing the independence-dependence needs of youth with SB were repeatedly reported to be major challenges and sources of stress for parents of youth with SB (Sawin et al., 2003).

Mobility and bladder and bowel dysfunction in individuals with SB are ongoing stressors for these individuals and their caregivers, and related condition management activities have been found to significantly contribute to SB-specific parenting stress. For example, maintaining clean intermittent catheterization schedules or reminding a child to maintain this schedule was found to significantly increase the level of parenting stress experienced by mothers of youth with SB (Kanaheswari et al., 2011). Another SB-specific factor found to affect parenting stress is the 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 ambulate via wheelchair (Antiel et al., 2016). 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.

While research has demonstrated the relationship between condition management and SB-specific parenting stress, few studies have examined the effects of SB-specific parenting stress on either parent or youth outcomes, including self-management. In fact, studies of condition-specific stress in other chronic health populations indicate that condition-related stress may not affect self-management. For example, one study of adults with type 1 or type 2 diabetes found that perceived condition burden was not related to frequency of self-management behaviors (Weijman et al., 2005). It is possible that individuals experience condition-related stress *because* they are concerned about remaining adherent to prescribed medical regimen. However, it seems likely that parents with a high level of condition-related stress (in this case,

SB-specific parenting stress), may be less likely to transfer condition-related responsibilities to their child with SB. More research is necessary to fully understand the relation between SB-specific parenting stress and youth self-management of condition-related tasks.

Parent Perceptions, Attitudes, and Behaviors

Both the Pediatric Self-Management Model and IFSMT highlight family and parent perceptions, attitudes, and health-related beliefs, theoretically, as factors that can influence the way self-management behaviors occur. Therefore, this study aims to investigate the associations between a number of parent cognitions (including perceptions, beliefs, and attitudes) and self-management in youth with SB.

Parental perception of child vulnerability. *Parental perception of child vulnerability* (PPCV) reflects parents' attitudes or beliefs that their child is especially at risk for or more susceptible to serious illness, injury, or harm (Green & Solnit, 1964; Thomasgard & Metz, 1997). PPCV is especially relevant to pediatric populations, as children with chronic medical conditions are more likely to be perceived as vulnerable by parents than their TD peers (Haverman et al., 2014; Houtzager, Möller, Maurice-Stam, Last, & Grootenhuis, 2015). For parents of youth with SB, factors unrelated to youth's condition (e.g., parent adjustment) have been found to influence PPCV more significantly than condition-related factors (e.g., IQ, severity of condition; Driscoll et al., 2018). In terms of demographic factors related to PPCV, this study (that included both mothers and fathers of youth with SB) found that mothers and fathers who reported lower SES and fathers of children who had greater condition severity perceived their child as being more vulnerable (Driscoll et al., 2018).

Some studies have examined the relationship between PPCV and illness management (though not self-management specifically) in other pediatric illness groups. For example,

Spurrier et al. (2000) found that parents of children with asthma who perceived their child as more vulnerable were more likely to restrict their children's activities, keep their children home from school, and take their child to the doctor for acute care. Similarly, for families of children with chronic pain, increased PPCV was found to be associated with poorer child functioning and more child pain-related health care utilization (Connelly, Anthony, & Schanberg, 2012). In summary, the results of these studies indicate that when parents perceive their children as more highly vulnerable, they tend to be more reliant on health care providers. Though not explored explicitly, it is possible that parents with these perceptions are more hesitant to allow their child with a chronic health condition to have responsibility over their medical regimen.

Illness uncertainty. *Illness uncertainty* is defined as a cognitive experience that occurs when an individual cannot determine the meaning of illness-related events (Mishel, 1983). Illness uncertainty is thought to arise in response to illness-related events that are appraised (in this case, by parents) as unclear or unpredictable, either due to insufficient information or from unpredictable symptoms or illness events. To date, condition-related uncertainty has not been studied in SB populations. However, illness uncertainty has been found to be reliably associated with both parent adjustment (e.g., general psychological distress) and child adjustment outcomes (e.g., depressive symptoms) across multiple pediatric medical conditions (Carpentier, Mullins, Chaney & Wagner, 2006; Chaney et al., 2016; Pai et al., 2007; Stewart, 2000; Tackett et al., 2016; White et al., 2005).

Research in pediatric illness populations supports the relevance of this parental perception to the study of self-management. For example, a study of families of youth with juvenile rheumatic diseases found that increased parental illness uncertainty lead to increased caregiver demand (Chaney et al., 2016). Specifically, regardless of their child's current health

status, in the presence of illness uncertainty, parents tended to intensify their efforts to establish

stability and respond with increased vigilance for both illness-related and illness-unrelated

events. Although not tested directly, it could be hypothesized that illness uncertainty could lead

to increases in miscarried helping by parents, a behavior that would likely hinder growth in self-

management for youth (Anderson & Coyne, 1991; Fales, Essner, Harris, & Palermo, 2014).

However, further research is needed to support this proposed association between parental illness

uncertainty and self-management in youth with SB.

Overprotection. While the instinct to protect and care for one's child is normative,

overprotection is defined as a set of parenting behaviors that go beyond what is developmentally

appropriate and most parents would do in similar circumstances (Thomasgard, 1998). Parental

overprotective behaviors have been characterized as being more restrictive, intrusive, and

controlling. These behaviors may be problematic in youth development of self-management

skills, as overprotective parents may be hesitant to allow their child to accept responsibility for

important, condition-related tasks. For example, adolescents with type 1 diabetes who indicated

that they perceived their parents as *collaborators* in care (rather than as *controllers* of care), were

more autonomous in care management and more adherent to a prescribed medical regimen

(Wiebe et al., 2005).

Research has shown that both mothers and fathers of children with SB are more

overprotective than parents of typically developing youth (Holmbeck et al., 2002). This

difference in behaviors has been found when utilizing both self-report and observational

measures. Further, parental overprotection was associated with lower levels of preadolescent

decision-making autonomy and behavioral autonomy (Holmbeck et al., 2002). Still, as youth

with SB age, parental protectiveness decreases (Malm-Buatsi et al., 2015), though potentially not

to the same lower level of protectiveness that is displayed by parents of typically developing

youth. Therefore, overprotection displayed by parents of youth with SB may contribute to the

delay in transfer of condition-related responsibilities from parent to youth throughout

adolescence and young adulthood. More research is necessary to better understand the

relationship between parental overprotection and self-management in youth with SB.

Optimism. *Optimism* is broadly conceptualized as a person's inclination to anticipate the

best possible outcome (Fayed, Klassen, Dix, Klaassen, & Sung, 2011; Kurtz-Nelson & McIntyre,

2017). Optimism is an empowering, constructive attitude that involves anticipation and hopeful

expectation towards a desired outcome. A large longitudinal study of optimism followed

participants from childhood to adulthood and found that optimism was related to higher levels of

income and education and lower rates of divorce (Ellen, Remes, & Sovia, 2004). Parental

optimism has been found to predict positive parenting behavior in at-risk samples (Taylor et al.,

2012) and to serve as a protective factor for parents experiencing a variety of risk factors (Kurtz-

Nelson & McIntyre, 2017). In pediatric illness populations, parental optimism has been studied

most thoroughly in the context of childhood cancer diagnoses. Although optimism among

parents and caregivers has not been conclusively shown to affect cancer prognosis, optimism has

been shown to improve parental coping when their children are hospitalized and to reduce the

negative effects of severe illness on the family (LaMontagne, Hepworth, Salisbury, & Riley,

2003; Wrosch & Scheir, 2003).

There results from studies with adult chronic illness populations is also applicable to the

current study. In adults with chronic pain, optimism has been identified as one of the most

important factors predicting positive pain outcomes (Goodin, Bier, & McGuire, 2009; Hanssen,

Peters, Vlaeyen, Meevissen, & Vancleef, 2013). For example, one study of adults with chronic

musculoskeletal pain found that higher levels of optimism in these adult patients were related to persistence, flexible treatment goal management, and a commitment to new treatment goals (Esteve et al., 2018). Optimists are more inclined than pessimists to pursue goals intensely, demonstrate flexibility in goal adjustment, and are more likely to pursue new goals when current goals are unattainable. Additionally, optimism is associated with improvement in immunological markers and slowing disease progression in adults living with HIV (Brown, Hanson, Schmotzer, & Webel, 2014). A systematic review was conducted on studies examining the relationship between optimism and health status (Avvenuti, Baiardini, & Giardini, 2016). Based on this review, the authors propose two possible explicative hypotheses: (1) optimism may have a direct effect on the neuroendocrine system and immune responses, and (2) optimism may have an indirect effect on health outcomes by promoting protective health behaviors, adaptive coping strategies, and positive mood. Applying these findings to parents of youth with SB, optimism may aid parents in facilitating the process of transferring condition-related responsibilities from parent to child by promoting positive mood, flexible problem solving, and adaptive coping strategies. However, to date, this specific relationship remains unexplored in research.

Expectations for the future. Future orientation is broadly defined as the extent to which an individual thinks about the future, anticipates future consequences, and plans ahead before acting (Steinberg et al., 2009). Research on future orientation typically focuses on this construct in adolescence and adulthood. However, research on parents' expectations for their children's future have been deemed very influential for children's educational, occupational, and personal outcomes (Irwin & Elley, 2013). The construct of *parental expectations for the future* is operationalized in this study as a parents' expectations for the attainment of typical developmental milestones (e.g., graduating high school, employment). Research suggests that

optimistic expectations for future functioning are positively associated with outcomes; these parental expectations for the future may serve as a source of motivation (Goldenberg, Gallimore, Reese, & Garnier, 2001; Ivey, 2004). It could be hypothesized, then, that parents of youth with SB who have optimistic expectations for their child's future will also be more likely to promote independence in completing condition-related tasks.

Summary

Theoretical models highlight parent factors – including adjustment, stress, perceptions, attitudes, behaviors, and expectations – as having the potential to influence condition self-management for youth with SB. However, there is a clear gap in the literature in this area. There is little-to-no research investigating the associations among parent factors and self-management for individuals with SB. Therefore, the proposed study aims to increase understanding of the relationships among parent personal factors, perceptions, attitudes, behaviors, and expectations, and youth with SB's condition self-management.

Interventions Targeting Self-Management for Individuals with Spina Bifida

Self-management interventions are traditionally designed by healthcare professionals with the intent of preparing individuals to assume responsibility for managing their chronic health condition or engaging in health promotion activities. These interventions can be individual- or family-centered and can address self-management skills directly or can target factors thought to influence self-management skills (Ryan & Sawin, 2009). A review of the literature revealed that there have been few interventions addressing the unique self-management needs of individuals with SB of any age, let alone, interventions designed specifically for children and adolescents.

Dicianno and colleagues (2016) described the feasibility of using a mobile health intervention to promote self-management in young adults with SB. While the mobile health intervention (iMHere) was found to be feasible, self-management improvements were only reported for high users of the intervention. Currently, an additional mobile-health intervention promoting adherence to bladder management programs (e.g., clean intermittent catherization) for adolescents and young adults with SB is being tested in a randomized controlled trial (Amaral, 2016). Only one intervention promoting self-management, a summer camp-based program, has included children with SB in the evaluation process.

Two studies on this camp-based independence intervention for SB have been conducted. The intervention consists of three components: (1) collaborative (i.e., parent and camper) goal identification, (2) 1-hour daily group workshops that include psychoeducation and teaching of cognitive tools, and (3) goal monitoring by camp counselors and daily problem solving with campers on how to successfully accomplish goals (O'Mahar et al., 2010). This intervention has been found to improve both medically-related and social goals (O'Mahar et al., 2010). This intervention was also associated with improved youth management of SB responsibilities and independence in completion of SB-related tasks. These improvements were maintained one-month post-intervention (O'Mahar et al., 2010). These results were later replicated with a larger sample and a modified intervention (Holbein et al., 2013). Although Holbein et al. found that the intervention benefitted most campers, cognitive functioning and family income (a proxy for socioeconomic status) moderated the outcomes. Specifically, youth IQ, number of shunt revisions, and family income moderated the intervention effects for health-related self-care tasks experienced by campers.

The results of these studies are promising. This camp-based intervention is effective in improving self-management skills for children and adolescents with SB, at least in the short-term (Holbein et al., 2013; O'Mahar et al., 2010). Further, significant condition-related (number of shunt revisions, youth IQ) and demographic (SES) moderators have been identified. While it is important to understand the influence of these moderating factors, these specific factors (youth IQ, number of shunt revisions, SES) are not modifiable. It is imperative to understand *modifiable* factors that could influence youths' responses to the intervention.

While camp presents a unique opportunity for campers to learn and practice new skills, it is possible that when campers return to their regular home routines, they may regress to their previous level of independence, rather than continuing to move forward. Parents, therefore, play a critical role in ensuring that the gains made at camp are maintained over time. Parent factors represent *modifiable* contributors to the development of self-management skills. An understanding of how these factors can influence youth with SB's response to the intervention can lead to future beneficial adaptations of the intervention (for campers) and could identify potential targets for future parent-focused interventions, as self-management in SB is a family-centric process (Ryan & Sawin, 2009). Therefore, an additional purpose of the proposed study is to better understand the potential relations among parent personal factors, parental perceptions and attitudes, and youth with SB's response to a camp-based intervention targeting health-related independence.

The Current Study

The current study sought to expand our limited understanding of how parent personal adjustment factors and parental perceptions and attitudes are related to condition-related independence in youth with SB. Three distinct parent adjustment variables – parent distress,

general parenting stress, and SB-specific parenting stress – were examined. Additionally, the following parent perceptions/attitudes/behaviors were examined: parental perceptions of child vulnerability (PPCV), parental perception of youth condition uncertainty, parental overprotection, parental optimism, and parental expectations for the future. This study aimed to differentiate the contributions of these adjustment factors, perceptions, attitudes, and behaviors to youth responsibility for and mastery over self-management tasks. This study also proposed to investigate the relationships between these parent factors have and youth's responses to a camp-based psychosocial intervention that targets condition-related independence in individuals with SB (described above; Holbein et al., 2013, O'Mahar et al., 2010). Such responses were assessed both in terms of changes in self-management behaviors over the course of one summer and, for a subsample of participants, trajectories of self-management behaviors over two summers. Lastly, both youth self-management behaviors and parental perceptions/behaviors are expected to change as youth age (e.g., youth responsibility and mastery increasing; PPCV and overprotection decreasing). Because the age range of youth in this study's sample spanned multiple developmental periods (childhood and adolescence) and due to the expected changes in many study variables with changes in child age, this study will also examine age as a moderator of the relationships between parent factors and youth self-management.

It has been recommended that the research examining the functioning of individuals with SB include a developmental emphasis, a focus on both illness-specific and general family processes, and models considering family-related variables that serve as potential buffers for associations between risk factors and negative outcomes (Holmbeck, Zebracki, Papadakis, & Driscoll, 2017). The proposed study was in line with these recommendations, including a broad

age range (7-19 years), accounting for age, condition-severity, and intellectual functioning in analyses, and utilizing a family systems theory in selecting parent predictor variables.

Study Hypotheses

The present study had three objectives. The first objective was to identify relationships between the parent variables (including parent personal factors and parent perceptions/ attitudes/behaviors) and youth self-management (both youth responsibility for and youth mastery over condition-related tasks). It was hypothesized that higher levels of (a) general parenting stress, (b) SB-specific parenting stress, (c) parental distress, (d) parental perceptions of child vulnerability, (e) parental illness uncertainty, and (f) parental overprotection and lower levels of (g) parental optimism and (h) expectations for the future would be associated with lower levels of youth responsibility and mastery over condition-related tasks (*Hypotheses 1a-h;* see Figure 1), and that these relationships would exist specifically for older campers (when examining age as a moderator). These hypotheses were tested cross-sectionally, with both parent variables and youth self-management assessed at Time 1 (pre-intervention).

The second objective was to determine the relationships between these parent variables and youth response to the psychosocial camp-based intervention targeting self-management. Specifically, the parent variables were used to predict changes in youth condition self-management goal attainment and youth independence in performing and mastery of medical tasks from Time 1 (pre-intervention) to Time 3 (post-intervention follow-up; see Figure 2). It was hypothesized that lower levels of (a) general parenting stress, (b) SB-specific parenting stress, (c) parental distress, (d) parental perceptions of child vulnerability, (e) parental illness uncertainty, and (f) parental overprotection and higher levels of (g) parental optimism and (h) expectations for the future would be associated with greater increases in goal attainment and

Parental Adjustment Factors

- Personal Distress
- General Parenting Stress
- SB-specific Parenting Stress

Parent Perceptions/Attitudes/Behaviors

- Perception of Child Vulnerability
- Illness Uncertainty
- Overprotection
- Optimism
- Expectations for the Future

Youth Self-Management

- Responsibility for Condition-related Tasks
- Mastery over Condition-related Tasks

Figure 1. Proposed associations between parent factors and youth self-management

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┌─────────────┐     ╱──────────────╲  ╱──────────────╲  ╱──────────────╲
│             │     │    Time 1     │  │    Time 2     │  │    Time 3     │
│             │     │   Pre-camp    │  │  During Camp  │  │ 1-month post- │
│             │     ╲──────────────╱  ╲──────────────╱  │     camp      │
└─────────────┘                                          ╲──────────────╱
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Time 1 — Pre-camp

Time 2 — During Camp

Time 3 — 1-month post-camp

Camper & Parent — Collaborative goal-setting

Camper Questionnaires
- Goals assessment
- Independence in completing SB-related tasks

Parent Questionnaires
- Demographic & medical variables
- Goals assessment
- Independence in completing SB-related tasks
- Mastery over SB-related tasks
- Parent personal distress
- General parenting stress
- SB-specific parenting stress
- Parental perceptions of child vulnerability
- Parental illness uncertainty
- Parental overprotection
- Parental optimism
- Parent expectations for the future

Camper Questionnaires
- Goals assessment

Counselor Questionnaire
- Goals assessment

Assessment of Cognitive Functioning

Camper Questionnaires
- Goals assessment
- Independence in completing SB-related tasks

Parent Questionnaires
- Goals assessment
- Independence in completing SB-related tasks
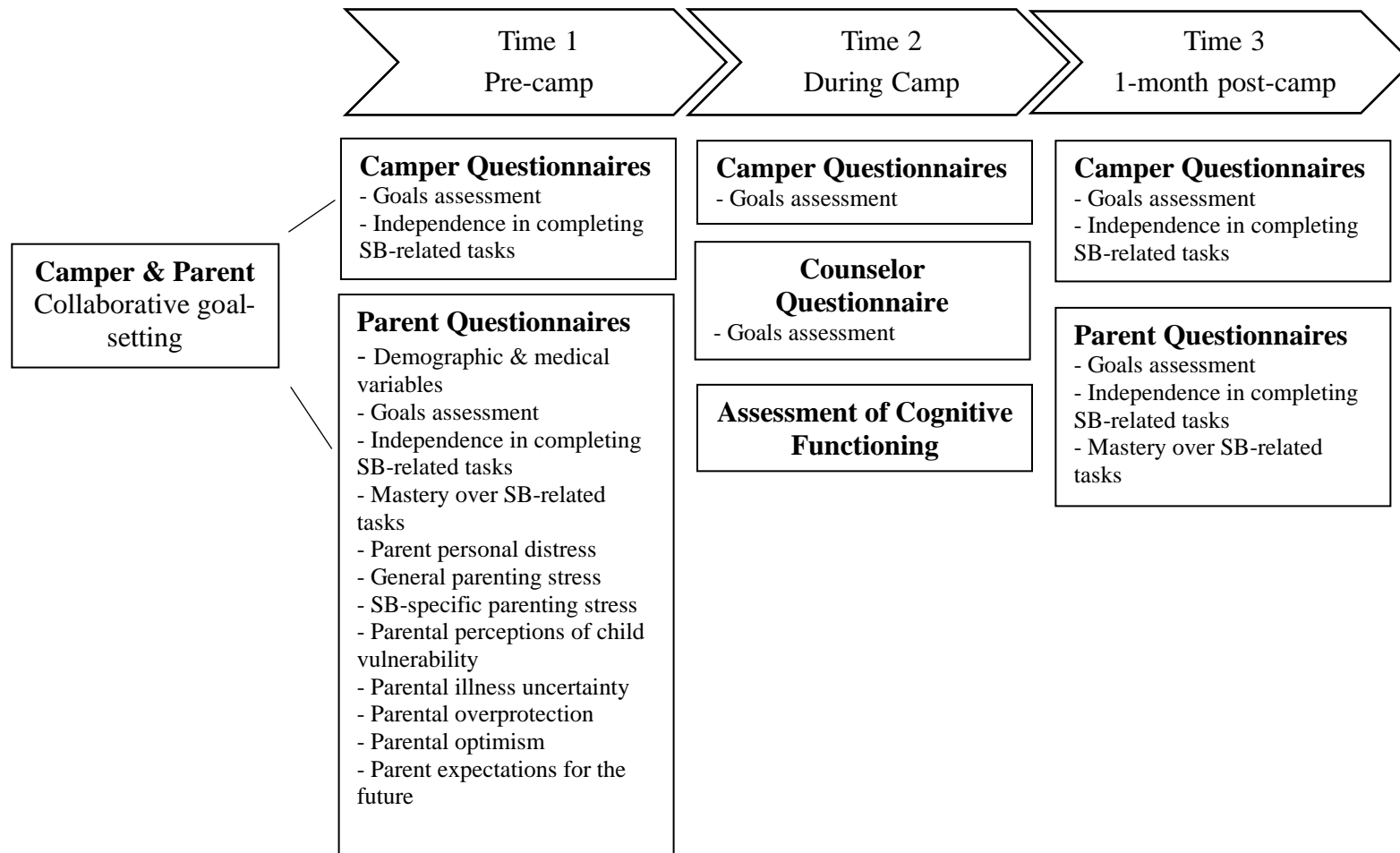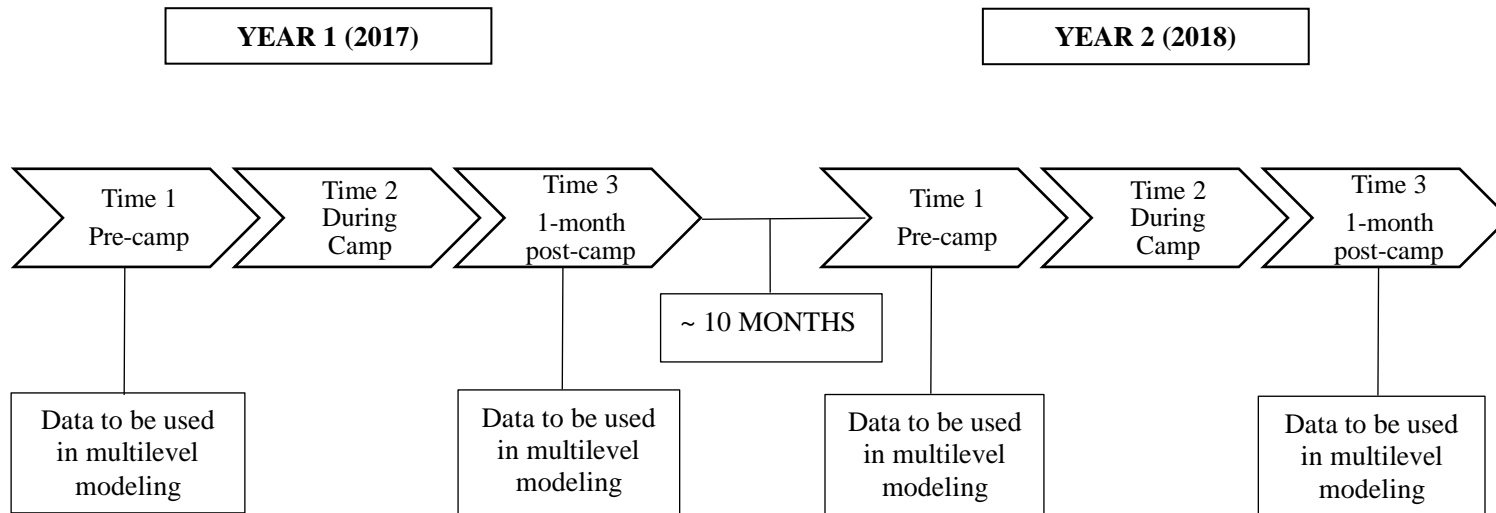- Mastery over SB-related tasks

Figure 2. Summary of data collection assessing summer-camp based intervention

greater improvements in medical self-management (i.e., independence in performing and mastery or health-related tasks; *Hypotheses 2a-h*). Again, it was hypothesized that these relationships would be increasingly significant with increased camper age.

The third objective was to examine the roles of parent factors in explaining differences in youth self-management trajectories (e.g., changes in self-management over time) for youth who participate in the intervention over two summers. These analyses included data from four assessment time points over the course of two summers (see Figure 3). It was hypothesized that lower levels of (a) general parenting stress, (b) SB-specific parenting stress, (c) parental distress, (d) parental perceptions of child vulnerability, € parental illness uncertainty, and (f) parental overprotection and higher levels of (g) parental optimism and (h) expectations for the future (Year 1, Time 1) would be associated with greater improvements (e.g., steeper trajectories) in medical self-management (growth from Year 1, Time 1 to Year 2, Time 3; *Hypotheses 3a-h*).

YEAR 1 (2017)

YEAR 2 (2018)

| Time 1 Pre-camp | Time 2 During Camp | Time 3 1-month post-camp | Time 1 Pre-camp | Time 2 During Camp | Time 3 1-month post-camp |

~ 10 MONTHS

Data to be used in multilevel modeling

Data to be used in multilevel modeling

Data to be used in multilevel modeling

Data to be used in multilevel modeling

Note: See Figure 2 for detail of constructs assessed at each data collection point.

Figure 3. Summary of data collection across two summers

CHAPTER THREE

METHODS

Participants

Participants were recruited from an ongoing longitudinal study examining the effects of a camp-based intervention targeting social and medically-based independence for youth and young adults with SB (Holbein et al., 2013; O'Mahar et al., 2010). Although this intervention study has been ongoing since 2009, the current study used data from summer 2017 and 2018 as these were the first two summers that included assessment of parent factors.

Participants were youth and one participating parent/caregiver attending an overnight summer camp in northern Illinois. Camp Independence exclusively serves individuals with SB and is conducted across eight week-long consecutive sessions, with each session serving one of three age groups (children: ages 7-13 years, adolescents: ages 14-19 years, young adults: ages 20+ years). Individuals with SB aged 7 years and older were eligible to apply to Camp Independence, although those with severe allergies or unpredictable health conditions (e.g., uncontrolled seizures) were ineligible. Placements in camp sessions were reserved on a first come first serve basis. Financial assistance was available to all families as needed. Every camper was approached to participate in the research study. Because parental involvement was not included in study procedures for young adult campers (those >20 years), only data from child and adolescent participants (e.g., <20 years of age) were included in this study's analyses. Additionally, there were adolescent campers (n = 5; 3 in 2017, 2 in 2018) who completed study

procedures without a parent participant. Given that the aims of the current study required parent

participation, these campers were excluded from this study's analyses.

The final sample included 89 camper-parent dyads, with 48 of these families (53.9% of

total sample) participating across both years. Youth and family demographic information and

youth condition-related information is included in Table 1. Demographic information (e.g.,

race/ethnicity, age) was not collected from parent participants so is not available to report.

Table 1. Youth and Parent Demographic and Condition-Severity Information

	Youth (N=89) M (SD) or N (%)	Parent Year 1 (N=89) N (%)	Parent Year 2 (N=48) N (%)
Gender: female	54 (60.7%)	78 (87.6%)	41 (93.2%) �
Age	12.20 (3.00)	--	--
Race			
Caucasian	64 (71.9%)	--	--
African-American/Black	8 (9.0%)	--	--
Hispanic	11 (12.4%)	--	--
Asian	3 (3.4%)	--	--
Multi-racial	3 (3.4%)	--	--
Previous years attended camp	2.67 (2.66)	--	--
Family Income	9.23 (5.82) ᵮ	--	--
IQ	89.52 (16.10)	--	--
SB type			
Myelomeningocele	73 (82.0%)	--	--
Other	7 (7.9%)	--	--
Unknown/not reported	9 (10.1%)	--	--
Lesion Level			
Thoracic	1 (1.1%)	--	--
Lumbar	53 (59.6%)	--	--
Sacral	18 (20.2%)	--	--
Unknown/not reported	17 (19.1%)	--	--
Shunt: present	66 (74.2%)	--	--
Unknown/not reported	1 (1.1%)	--	--
Ambulation			
No Assistance	5 (5.6%)	--	--
Braces, crutches, or combination (does not include wheelchair)	28 (31.4%)	--	--
Includes Wheelchair	56 (62.9%)	--	--

Note. Yearly family income was reported on a 21-point scale, from <$10,000 per year to >$20,000 per year, with each point on the scale representing increments of $10,000. For this sample, family income ranged from <$10,000 - $200,000+ per year with a mean of ~$65,000 and a standard deviation of ~$50,200. Youth and Year 1 N includes families that participated in 2017 only, in 2018 only, or in 2017 and in 2018.
ᵮ Sample size is reduced for these characteristics due to missing data/assessments.

Procedure

The current study was approved by the university Institutional Review Board and utilized a multi-method, multi-informant longitudinal research design. Figure 2 depicts the flow of the study. Families received information about Camp Independence from their health care professionals and through various print and online information provided by a local SB organization. Financial assistance and sliding scale fees were available for all families who could not afford to pay. A letter explaining the study, camper consent/assent, caregiver consent (for campers <20 years), and Time 1 questionnaires were sent to families in the mail ~1 month prior to the first day of camp. Assents, consents, and Time 1 (baseline) questionnaires were collected from families on the first day of camp. Families who had not completed questionnaires prior to coming to camp were given the opportunity to do so on the first day of camp. Families were paid $10.00 for completing Time 1 questionnaire packets. Questionnaires for Time 2 were completed throughout the week by campers and counselors. Time 3 questionnaires were sent to parent and child participants approximately one month post-intervention via mail with prepaid envelopes for return. In summer 2018, families were given a choice to receive their Time 3 questionnaires in hard copy via mail or electronically, through Opinio, via email. Of the 68 families who participated in 2018, 36 (52.9%) chose to receive Time 3 questionnaires electronically, and 24 of these families (66.7%) returned completed electronic questionnaires. The remaining 32 families opted to receive hard copies of questionnaires, and 25 (78%) returned completed Time 3 questionnaires. Families who opted to receive questionnaires electronically had significantly younger campers ($t(66)=2.27$, $p < .05$) and significantly higher family income ($t(66) = -2.47$, $p < .05$) than those who chose to receive hard copies of questionnaires (M_{age}: 11.33 years [electronic], 12.97 [hard copies]; M_{income}: 11.69 [electronic], 8.13 [hard copies]). There were no

differences in camper IQ, lesion level, or number of previous years participating in the camp

intervention between families who opted for electronic or hard copies of questionnaires. The

overall return rate for Time 3 questionnaires for child and adolescent participants was 63.5% in

2017 and 72.1% in 2018. Upon receipt of Time 3 questionnaires, families were paid $40.00 (via

a check sent in the mail). Follow-up phone calls were made at Time 1 and Time 3 to ensure

receipt of all study materials and to answer any questions. All materials were available in in

either English or Spanish. Questionnaires that were only available in English were adapted for

Spanish speakers using forward and back translation. The current study uses both youth- and

parent-reported questionnaire data.

At Time 1, campers and their parents completed measures assessing camper functioning,

medical information, and demographic information. Participating parents also completed

questionnaires assessing their own adjustment, perceptions, attitudes, and behaviors at Time 1.

At Time 2, campers and counselors completed measures assessing campers' social and medical

goal progress and social functioning. Neurocognitive tests were administered by trained research

assistants during camp. At Time 3, both campers and parents completed measures assessing

camper functioning as well as feedback about the camp program (generally) and about the

intervention, specifically.

Intervention

The intervention has been described previously (Holbein et al., 2013; O'Mahar et al.,

2010). Generally, the intervention was designed to address previously identified social and

independence difficulties specific to youth and young adults with SB. The intervention is

embedded within a typical camp program designed for individuals with spina bifida that includes

swimming, arts and crafts, and other outdoor/physical activities (high ropes, rock climbing,

boating, etc.). Camp Independence is located in northeastern Illinois on the larger 300-acre

YMCA day and summer camp, Camp Duncan.

The independence intervention has three main components: (1) goal-setting, (2)

counselor monitoring of goals, and (3) one-hour daily workshops. Before the start of camp,

parents and campers collaborate to identify two specific goals – one health-related self-care goal

and one social goal – for the campers to work on throughout the camp session. Lists of potential

health-related self-care and social goals are provided to families to choose from. At camp,

counselors monitor camper goals and collaborate with campers daily to review campers' specific

goals, identify daily steps to achieve goals, and discuss barriers to goal achievement. This

problem-solving process is completed every day during the camp week. This problem-solving

process is guided by daily questionnaires that campers and counselors complete collaboratively.

Counselors receive training prior to camp on how to facilitate this problem-solving process with

campers. Additionally, a trained post-baccalaureate research assistant lives at camp each week

and is available to answer questions and aid counselors in tracking goal progress during the

week.

In addition to goal-setting and monitoring, campers participate in hour-long groups,

called "Independence Workshop," that occur on four days of the camp week for ~one hour each

day. Workshops include psychoeducation and strategies (e.g., problem-solving and

communication) aimed at promoting health-related and social independence. Workshops are led

by trained post-baccalaureate research assistants, follow a manualized curriculum for each

session, and include multiple interactive activities, such as group and partner discussions, art

projects, games, workbook exercises, and role plays. Each day, a different topic is addressed: (1)

communication and taking care of relationships, (2) self-esteem and emotional wellness, (3)

living with SB (e.g., personal reactions and dealing with others' reactions to SB), and (4) health-related self-care and planning for the future. Campers are provided workbooks to use throughout the week and take home with them. Three versions of the intervention manual are used to ensure developmentally appropriate content and activities for each age group (7-13 year old campers, 14-19 year old campers, 20+ year old campers). Minor revisions (changes to the warm-up games and activities) were made to the intervention prior to the start of camp in 2018. However, the overall content of each session remained consistent.

Measures

Covariates

 Medical and demographic factors. At Time 1, parents completed a demographics form assessing camper's age, gender, race, and medical characteristics (i.e., lesion level, SB type, number of shunt surgeries, and ambulation status). Total household income was reported on a 21-point scale, from <$10,000 per year to >$200,000 per year, with increments of $10,000.

 Youth intellectual functioning. Youth were administered the Vocabulary and Matrix Reasoning subtests of the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) while at camp (Time 2). 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. Raw subtest scores were converted to norm-reference scores, yielding an estimated Full Scale IQ score (FSIQ). These subtests have demonstrated high levels of internal consistency for

individuals aged 6 to 16 years (α = .89 for Vocabulary, α = .92 for Matrix Reasoning; Wechsler, 1999). If already administered within the past two years, FSIQ scores were extracted from previous camp evaluations.

Outcome Measures

   Independence in completing sb-related tasks. The Sharing of Spina Bifida Management Responsibilities (SOSBMR) was adapted from the Diabetes Family Responsibility Questionnaire (DFRQ; Anderson, Auslander, Jung, Miller, & Santiago, 1990), a measure that has shown adequate internal consistency and concurrent validity. The SOSBMR was used to assess responsibility for SB tasks across several domains (e.g., health appointments, communication about SB, medications). Campers and parents indicated who was responsible for 34 SB-related tasks (1 = parent, 2 = shared, 3 = camper, 4 = not applicable). Mean total scores were computed, with higher scores indicating greater camper responsibility for condition-related tasks. Because items deemed "not applicable" by respondents are considered missing data when total scores are calculated, alphas for this adapted scale were not computed (because reliability programs only include participants who respond to all items).

   Mastery of self-management skills. The 48-item SB Independence Survey (SBIS) was adapted from a validated diabetes questionnaire (Diabetes Independence Survey (DIS); Wysocki et al., 1996). Parents responded "yes," "no," "not sure," or "N/A" regarding their child's mastery of condition-related skills (e.g., medication management, catherization). Ratio scores of "yes" responses to the total number of item responses were calculated to determine the degree to which a camper had mastered condition-related tasks, with high scores indicating greater mastery of tasks. Similar to the SOSBMR questionnaire, items deemed "not applicable" by respondents on

this scale are considered missing data. Therefore, reliability alphas are not able to be calculated

for the SBIS.

Parent Measures

Parent distress. Parents 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) was used

in this study. The GSI is the average of all items from all subscales, with higher scores indicating

higher global distress. Traditional psychometric analyses of the SCL-90-R have consistently

reported acceptable levels of internal consistency (Derogatis et al., 1976), and internal

consistency was found to be acceptable in this sample ($\alpha = .98$).

General parenting stress. Subscales from the Parenting Stress Index (PSI, Abidin, 1990)

were used to assess parenting stress (e.g., stress an individual experiences as a direct result of

being a parent). The current study used three parenting subscales: Perceived Parental

Competence, Restriction of Role, and Social Isolation. These three subscales were chosen

because they are believed to best capture the functioning of parents in their role as parents. These

three subscales are comprised of 24 items total; 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. In creating total scores, raw item scores were converted to z-scores so

that 4- and 5-point scale items could be totaled together. A total parenting general parenting

stress score was computed, with higher scores on this measure indicating higher reported

parenting stress. In samples of parents of youth with SB, this total score has previously shown adequate internal consistency (α = .79-.89; Friedman, Holmbeck, Jandasek, Zukerman, & Abad, 2004). In this study's sample, internal consistency was acceptable (α = .62).

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 six 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 used a total score comprised of the 6 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), and the SB-specific scale showed good internal consistency in this sample (α = .83).

Parental perceptions of child vulnerability. The 16-item Vulnerable Child Scale (VCS; Perrin, West, & Culley, 1989) is a modified version of Forsyth's Child Vulnerability Scale (Forsyth, 1987). The current study employed a 15-item version; the item "I sometimes worry that my child will die" was dropped due to the pediatric nature of the study population. All statements on this measure reflect specific concerns about the child's health. Parents responded to each item on a four-point scale (1 = definitely true, 4 = definitely false). The measure has an inter-item reliability of .75 with a test-retest reliability of .96 (Perrin et al., 1989). Internal consistency in this study was found to be good (α = .80).

Parental illness uncertainty. Parents completed the Parents' Perception of Uncertainty in Illness Scale (PPUS), which was adapted from the Mishel Uncertainty in Illness Scale (MUIS)

and measures a parents' evaluation of their uncertainty experienced concerning their child's

medical condition (Mishel, 1983). The scale asks parents to respond to 31 items regarding the

uncertainty of their child's condition and treatment (e.g., "I am unsure if my child's illness is

getting better or worse") that are rated on a 5 point Likert scale (5 = Strongly Agree, 1 =

Strongly Disagree). The PPUS is comprised of four factors: Ambiguity, Lack of Clarity, Lack of

Information, and Unpredictability. However, this study utilized a single total scale score. This

total score has demonstrated adequate internal consistency previously (α = .81 - .93; Mishel,

1983), and good internal consistency in this study (α = .88).

Parental overprotection. Parents completed the Parent Protection Scale (PPS;

Thomasgard, Metz, Edelbrock, & Shonkoff, 1995), a 25-item self-rating scale that assesses

parenting behaviors across the subscales of Supervision, Dependence, Separation Problems, and

Control. Each item is rated on a four-point Likert scale (ranging from 0 = never to 3 = always).

Seven items are reverse-coded. Higher total scores represent greater levels of protection. The

internal reliability of the total score has been found to be satisfactory in previous research (α =

.73; Thomasgard et al., 1995). Internal consistency in this study's sample was acceptable (α =

.70).

Parental optimism. Parents completed the Life Orientation Test – Revised (LOT-R;

Scheier, Carver & Bridges, 1994; Herzberg, Glaesmer, & Hoyer, 2006), a 10-item self-report

questionnaire that assesses traits of optimism and pessimism using a 5-point Likert scale

(ranging from 1 = I Disagree a Lot to 5 = I Agree a lot). Three items are scored for optimism,

three items for pessimism, and remaining four items are not scored (i.e., "filler" items). Given

differences in reliability among the two factors, only the three-item optimism factor (α = .71;

Herzberg et al., 2006) was used in this study and demonstrated adequate internal consistency with this study's sample ($\alpha = .67$).

Parental expectations for the future. Parental expectations for their children with SB's future functioning were assessed using a measured designed for a previous study by the same principal investigator (Questions about the Future [QAF] described in Holbein et al., 2015). Parents rated the extent to which they believed their children would be capable of attaining specific adulthood milestones (e.g., attaining a full-time job, living independently, marriage, parenthood) on a 4-point Likert scale (ranging from 1 = very unlikely to 4 = very likely). For this study, a total expectations for the future score was calculated, with higher scores indicating higher parental expectations for campers' futures. Internal consistency for this measure was found to be excellent in this study's sample ($\alpha = .95$).

Statistical Treatment

Preliminary Analyses

Data was analyzed using SPSS for Windows (V. 26) and Mplus (V 8.1). Prior to hypothesis testing, the psychometric properties (e.g., alphas) of all measures were evaluated (reported above). All variables were examined for outliers using stem-and-leaf plots. Analyses were also performed to determine if any variables 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 time points and both years. Therefore, attrition analyses were performed to evaluate differences between families who discontinued participation and those who did not.

Primary Analyses

All analyses included youth IQ, lesion level, camper age, reported family income, and previous participation in the intervention as covariates, as all of these may contribute to parent adjustment factors, parent perceptions and attitudes, youth medical self-management, and youth response to intervention. To have a broad understanding of the associations among the independent and dependent variables, Pearson correlations were performed, and a correlation matrix was created prior to hypothesis testing. Further, both parents and campers reported on the dependent variable of camper independence in completing self-management tasks. If these reports were found to be highly correlated, a composite variable would be created and used in all subsequent analyses.

Analytic plan for objective 1. A series of hierarchical multiple regression analyses were conducted to examine associations between all parent variables and youth self-management variables at Year 1 Time 1 (see Figure 1). When running these cross-sectional regression analyses, independent variables were entered in the following order: (Step 1) covariates – IQ, lesion level, camper age, income, years of prior camp participation; (Step 2) individual predictors in blocks (Block 1 [adjustment]: general parenting stress, SB-specific parenting stress, parental distress; Block 2 [perceptions and attitudes]: parental perception of child vulnerability, parental illness uncertainty, overprotection, optimism, and expectations for the future). Separate regressions were run for both blocks of predictor variables .

An a priori power analysis was conducted using G*Power 3.1 (Faul, Erdfelder, Buchner, & Lang, 2009). For the proposed regression analyses, assuming a power of .95 and an alpha of .05, a sample size of 1,050 was required to detect small effect sizes, a sample of 146 was required to detect medium effect sizes, and a sample of 67 was required to detect large effect

sizes. The final sample size for the current study (of campers participating for at least one summer) was 89 families. Thus, the proposed study was adequately powered to detect large effect sizes.

Analytic plan for objective 2. A series of hierarchical multiple regression analyses were conducted to assess if there were significant parent predictors of changes in youth self-management variables from Time 1 to Time 3 after controlling for the effects of demographic covariates. Residual scores were created by controlling for Time 1 of the dependent variable, and these residuals were used as the dependent variable in subsequent analyses.

Independent variables were entered in the following order: (Step 1) covariates – Time 1 of dependent variable, IQ, lesion level, camper age, income, years of prior camp participation; (Step 2) individual predictors in blocks (Block 1 [adjustment]: general parenting stress, SB-specific parenting stress, parental distress; Block 2 [perceptions and attitudes]: parental perception of child vulnerability, parental illness uncertainty, overprotection, optimism, and expectations for the future). Separate regressions were run for each block of predictor variables. The self-management dependent variables were entered at Time 3.

An a priori power analysis was conducted using G*Power 3.1 (Faul et al., 2009). For the proposed regression analyses, assuming a power of .95 and an alpha of .05, a sample size of 1050 is required to detect small effect sizes, a sample of 146 is required to detect medium effect sizes, and a sample of 67 is required to detect large effect sizes. The final sample size for the current study (of campers participating for at least one summer) was 89 families. Thus, the proposed study was adequately powered to detect large effect sizes.

Analytic plan for objective 3. To examine individual trajectories of youth self-management variables (independence in completing SB-related tasks, mastery over self-

management skills) with parent variables as predictors of these trajectories, multilevel modeling was conducted with Mplus. Multilevel modeling was chosen, as this approach allows for examination of both change over time in self-management variables within each individual (Level 1) as well as the role of the parent variables in explaining differences in trajectories between individuals (Level 2). This approach was deemed to be the most appropriate for this study's longitudinal data for two reasons: (1) multilevel modeling in Mplus allows for variability in intervals between measurements (e.g., assessments do not need to be completed at regular intervals), and (2) multi-level modeling can utilize maximum likelihood estimation to manage missing data (Gray et al., 2018). Rather than eliminating a participant due to missing data (as in listwise deletion), maximum likelihood estimation uses all available data for a participant to estimate his or her trajectory. This approach allowed for trajectories of the entire sample to be examined, not just those for a potentially biased subsample of only those individuals with complete data. These analyses utilized data from four assessment time points across both years of data collection: Year 1, Time 1; Year 1 Time 3; Year 2 Time 1; Year 2 Time 3 (see Figure 3).

A multistep approach was used when applying multilevel modeling to the data. First, a null model was built to determine if the data were appropriate for multilevel modeling. Next, linear and quadratic models were created with self-management variables (independence in completing self-management tasks, mastery over condition-related tasks) at each time point entered as the dependent variable, time entered as within-subjects Level 1 variable, and demographic covariates (IQ, lesion level, camper age, income, years of prior camp participation) and parent factors at Year 1 Time 1 entered as between-subjects Level 2 variables. Fit indices were compared across equivalent models to identify the model that best fit the data.

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). Results indicated that two variables were positively skewed: parent-report on the SCL-90 (skewness value = 3.02) and parent-report on the Family Stress Scale (skewness value = 1.11). One variable, parent-report on the QAF, was found to be negatively skewed (skewness value = -1.17). Scoring on this variable was reversed prior to using data transformation techniques. These variables were first transformed using the square root transformation; however, they continued to be skewed significantly. These variables were transformed using log transformations and were found to be acceptable. Lastly, as parent- and camper-reports on the SOSBMR ($r = .78$-$.85$) and health-related goal attainment ($r = .55$-$.66$) were found to be highly correlated, composite total scores for these variables were created and used in subsequent analyses.

Attrition Analyses

As was anticipated, not all families participated at all four data collection time points. As mentioned previously, 89 families participated for at least one time point and were included in this study. Of those 89 families, 25 (28.1%) completed Time 1 questionnaires only and 64

43

(71.9%) completed questionnaires at Time 1 and Time 3. Analyses revealed that families who

participated at both time points reported significantly higher parent report of illness uncertainty

(t(86)=2.15, p<.05) than those who participated only at Time 1. Of the total sample, 48 families

(53.9%) participated across two summers. There were no significant differences between

families who participated for one or two summers on any of the covariate, independent, or

dependent variables (all p's > .05). For the second summer of data collection, 14 families

(29.2%) participated at Time 1 only and 34 families (70.8%) participated at both Time 1 and

Time 3. There were no significant differences between families who participated at one or both

time points on any of the covariate, independent, or dependent variables (all p's > .05).

Primary Analyses

Correlation Matrix

Prior to hypothesis testing, a series of Pearson correlations were performed, and a

correlation matrix was created (see Table 2). This matrix shows the correlations between

covariates, independent variables, and dependent variables at Year 1 Time 1. Means and standard

deviations for all variables at Year 1 Time 1 are also included in Table 2.

The following covariates were found to be significantly positively correlated with camper

condition-related responsibility: camper age (r = .59, p < .01) and previous years at camp (r =

.40, p < .01). These covariates were also significantly positively correlated with camper mastery

over condition-related tasks (age: r = .45, p < .01; previous years at camp: r = .49, p < .01).

Camper IQ, camper lesion level, and family income were not significantly correlated with either

outcome variable at Year 1 Time 1. Two of the independent parent predictor variables were

significantly correlated with camper condition-related responsibility: parental perception of child

vulnerability (r = -.32, p < .01) and parental overprotection (r = -.38, p < .01). Parental

overprotection was also significantly negatively correlated with camper condition-related

mastery ($r = -.26$, $p < .05$). There were no other significant associations between the independent

and dependent variables.

A number of significant associations existed among the independent parent adjustment

and perception/attitude/behavior variables (see Table 2). All of these associations were in the

expected direction. Maternal distress was significantly correlated with general parenting stress (r

$= .30$, $p < .01$), SB-specific parenting stress ($r = .30$, $p < .01$), PPCV ($r = .36$, $p < .01$), illness

uncertainty ($r = .23$, $p < .05$), and optimism ($r = -.38$, $p < .01$). Maternal general parenting stress

was significantly correlated with overprotection ($r = .27$, $p < .05$). SB-specific parenting stress

was significantly correlated with PPCV ($r = .22$, $p < .05$), illness uncertainty ($r = .29$, $p < .01$),

overprotection ($r = .26$, $p < .05$), optimism ($r = -.23$, $p < .05$), and expectations for the future ($r =$

$-.24$, $p < .05$). Maternal PPCV was significantly cross-sectionally associated with illness

uncertainty ($r = .43$, $p < .01$), overprotection ($r = .42$ $p < .01$), and expectations for the future ($r =$

$-.32$, $p < .01$). Maternal illness uncertainty was significantly correlated with overprotection ($r =$

$.23$, $p < .05$) and optimism ($r = -.24$, $p < .05$). Despite significant correlations among the

independent variables, because the constructs were conceptualized as separate entities, composite

scores were not created.

Table 2. Descriptives and Correlation Matrix of Covariates, Independent, and Dependent Variables at Year 1 Time 1

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15
1. IQ	89.52 (17.00)	2.24 (.46)	12.20 (3.00)	9.23 (5.82)	2.67 (2.66)	.26 (.40)	74.84 (7.26)	11.41 (4.61)	1.67 (.42)	60.44 (16.65)	1.01 (.27)	11.65 (2.45)	3.31 (.71)	1.98 (.40)	.60 (.25)
2. Lesion Level (p)	.002	1													
3. Camper Age (p)	-.16	.006	1												
4. Family income (p)	.17	.001	-.03	1											
5. Previous years at camp (p)	-.22*	.06	.59**	-.11	1										
6. Distress (p)	.20	-.06	.08	-.18	-.001	1									
7. Parenting Stress (p)	-.03	-.05	-.08	-.23	-.11	.30**	1								
8. SB-specific Parenting Stress (p)	.07	-.15	-.02	.07	-.14	.30**	.07	1							
9. PPCV (p)	-.02	-.15	-.18	-.17	-.24*	.36**	.18	.22*	1						
10. Illness Uncertainty (p)	-.11	-.10	.07	-.03	-.03	.23*	-.02	.29**	.43**	1					
11. Overprotection (p)	-.06	-.07	-.38**	-.10	-.29**	.22	.27*	.26*	.42**	.23*	1				
12. Optimism (p)	.04	.14	-.10	.08	-.05	-.38**	-.11	-.23*	-.12	-.24*	.02	1			
13.Expectations for the Future (p)	.35**	-.08	-.25*	.13	-.11	-.15	.05	-.24*	-.26*	-.14	-.06	.21	1		
14. Camper Condition-Related Responsibility (p + c)	.11	-.06	.59**	-.05	.40**	.04	-.02	-.14	-.32**	-.06	-.38**	.03	.09	1	
15. Camper Condition-Related Mastery (p)	.20	.04	.45**	-.15	.49**	.02	-.11	-.17	-.19	-.06	-.26*	.08	.11	.70**	1

Each cell underneath the diagonal indicates Pearson Correlation value (*r*). Values in the top row indicated means and standard deviations for each variable in the following format: *M(SD)*

Letters after variable name indicate reporters on variables: (p) = parent; (p + c) = parent and camper

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

Hypothesis Testing

Based on the results of Pearson correlations among this study's proposed covariates, independent, and dependent variables presented (above and in Table 2), some changes were made to the originally proposed analytic plan. In an effort to maximize sample size (by limiting the number of participants removed from analyses due to missing data via listwise deletion) and due to their lack of significant cross-sectional associations with the outcome variables, the proposed covariates of camper IQ, lesion level, and family income were excluded from this study's primary analyses. Camper age and previous years attending camp were included in analyses as covariates, and camper age was examined as a moderator of significant effects.

Objective 1. A series of hierarchical multiple regression analyses were conducted to examine the associations between all parent variables and youth self-management variables at Year 1 Time 1 (see Table 3). Analyses were run both without covariates and with the covariates of age and previous years at camp.

Condition-related responsibility. When covariates were not included in analyses, parental distress (β=.16, p=.17), general parenting stress (β=-.09, p=.48), and SB-specific parenting stress (β=-.17, p=.16) were not significantly associated with youth's condition-related responsibility. When including the covariates of age and previous years at camp, cross-sectionally, parental distress (β=.12, p=.23), general parenting stress (β=.01, p=.93), and SB-specific parenting stress (β=-.12, p=.20) were again not significantly associated with youth's condition-related responsibility.

Table 3. Summary of Cross-Sectional Regression Analyses Showing Associations Between Parent Variables and Camper Self-Management Variables (Objective 1)

Variable	Step	Condition-Related Responsibility			Step	Condition-Related Mastery		
		b	β	ΔR2		*b*	β	ΔR2
Camper Age	1	.07	.55	.41**	1	.02	.19	.32**
Previous Years at Camp		.02	.14			.04	.44	
SB-specific Parenting Stress	2	-.30	-.12	.01	3	-.14	-.09	.01
Distress	3	.09	.12	.01	2	.04	.09	.01
General Parenting Stress	4	.00	.01	.00	4	-.01	-.09	.01
Camper Age	1	.07	.54	.38**	1	.02	.23	.32**
Previous Years at Camp		.02	.13			.04	.40	
Expectations for the Future	2	.63	.26	.06**	2	.33	.22	.04*
PPCV	3	-.13	-.13	.01	5	.04	.06	.00
Overprotection	4	-.11	-.08	.01	6	-.05	-.05	.01
Optimism	5	-.01	-.03	.00	3	.01	.09	.01
Illness Uncertainty	6	-.00	-.02	.00	4	-.01	-.06	.00

Note: All predictor and outcome variables are measured at Time 1, and separate regressions were run for two blocks of predictors (Block 1: adjustment variables, Block 2: perceptions/attitudes). For all analyses, the covariates of camper age and previous years at camp were entered in Step 1.
*$p < .05$, **$p < .01$

When considering parental perceptions/attitudes/behaviors without covariates, parental overprotection was significantly associated with youth with SB's condition-related responsibility (β=-.39, p<.001), such that higher levels of reported overprotection were associated with lower levels of youth condition-related responsibility. PPCV (β=-.20, p=.08), illness uncertainty (β=.08 p=.52), optimism (β=-.04, p=.70), and expectations for the future (β=.02, p=.85) were not significantly associated with youth's condition-related responsibility. When including the covariates of camper age and previous years at camp in this analysis, parental expectations for the future were significantly associated with youth's condition-related responsibility (β=.63,

$p<.01$), such that higher expectations for the future were related to greater youth responsibility for condition-related tasks. PPCV (β=-.13, p=.15), overprotection (β=-.08, p=.43), optimism (β=-.03, p=.72), and illness uncertainty (β=-.02, p=.82) were not significantly associated with youth's condition related responsibility.

Camper age was examined as a moderator of the relationships between parent factors and camper condition-related responsibility. There were no significant interactions between camper age and any of the three parent adjustment factors or camper age and PPCV, overprotection, illness uncertainty, or optimism (all p's > .05). The interaction between camper age and parental expectations for the future was significant (β=.20, $p<.05$). Post hoc probing revealed that there was not a significant relationship between parental expectations for the future and youth's condition-related responsibility for younger campers (β=.16, p=.19), but that this relationship was significant for parents of older campers (β=.36, $p<.01$; see Figure 4). In other words, younger campers with different parental expectations for the future did not have differing levels of condition-related responsibility. However, for older campers, different levels of parental expectations for the future were associated with differing levels of camper condition-related responsibility (e.g., higher reported expectations associated with higher levels of responsibility).

Condition-related mastery. Parallel hierarchical multiple regression analyses were conducted to evaluate the associations between individual parent variables and youth with SB's mastery of condition-related tasks.

In analyses without covariates, parental distress (β=.14, p=.25), general parenting stress (β=-.18, p=.16), and SB-specific parenting stress (β=-.15, p=.23) were not significantly associated with youth's mastery over SB tasks. When including covariates, these variables

remained non-significantly related to youth's condition-related mastery (distress: β=.09, p=.36; general parenting stress: β=-.09, p=.40; SB-specific parenting stress: β=-.09, p=.41).

a.   b.
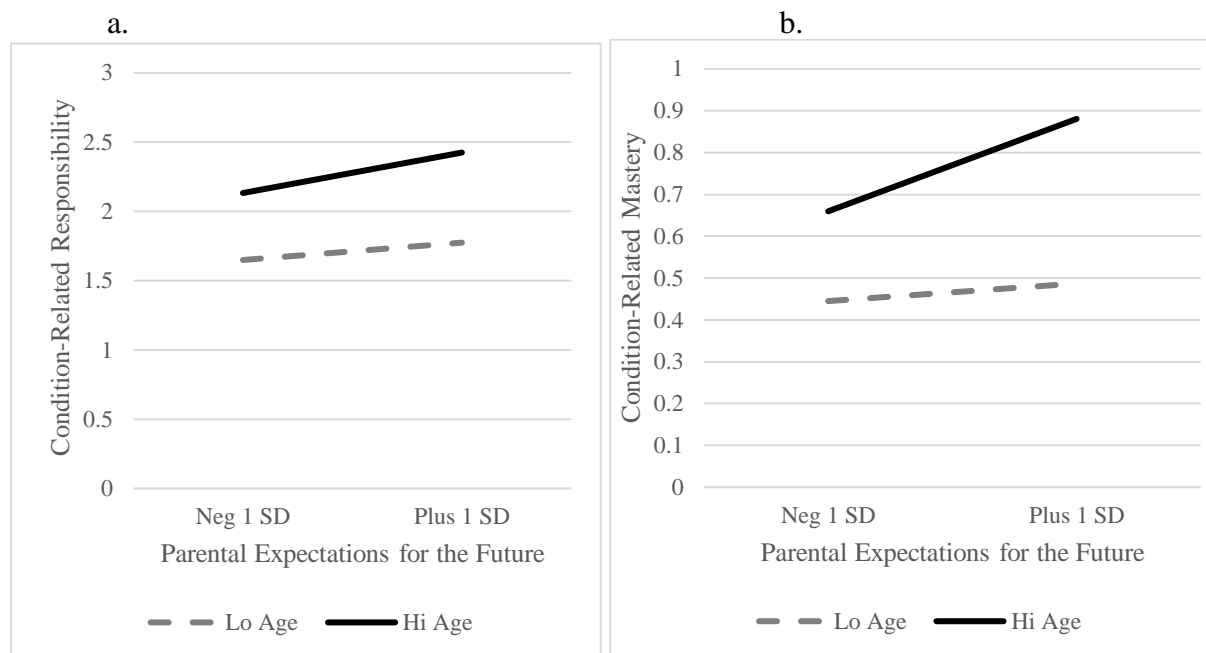
Figure 4. Simple slopes analyses of youth age as a moderator of the relationships between parental expectations for the future and youth condition-related responsibility (a) and condition-related mastery (b)

In the analysis without covariates, parental overprotection was significantly associated with youth's condition-related mastery (β=-.28, p<.05). When parents reported higher levels of overprotection, they also reported lower levels of camper mastery over condition-related tasks. PPCV (β=-.11, p=.37), optimism (β=.07, p=.50), expectations for the future (β=.04, p=.71), and illness uncertainty (β=.03, p=.84) were not significantly associated with camper condition-related mastery. With camper age and previous years at camp included as covariates, parental expectations for the future were found to be significantly associated with campers' mastery over condition-related tasks (β=.22, p<.05). Parents who reported greater expectations for campers' futures also reported higher levels of camper mastery over condition-related tasks. No other

parent perceptions/attitudes were significantly associated with campers' condition-related master (optimism: β=.09, p=.34; illness uncertainty: β=-.06, p=.54; PPCV: β=.06, p=.60; overprotection: β=-.05, p=.65).

Again, camper age was examined as a moderator of the relationships between parent factors and youth condition-related mastery. There were no significant interactions between camper age and any of the three parent adjustment factors or camper age and PPCV, overprotection, illness uncertainty, or optimism (all p's > .05). The interaction between camper age and parental expectations for the future was significant (β=.24, p<.05). Post hoc probing revealed that there was not a significant relationship between parental expectations for the future and youth's condition-related mastery for younger campers (β=.08, p=.53), but that this relationship was significant for parents of older campers (β=.43, p<.01; see Figure 4). In other words, younger campers with different parental expectations for the future did not have differing levels of mastery. However, for older campers, different levels of parental expectations for the future were associated with differing levels of camper condition-related mastery (e.g., higher reported expectations associated with higher levels of condition-related mastery).

Testing for suppression effects. Because there were no significant cross-sectional bivariate associations between parental expectations for the future and either of the self-management outcome variables (condition-related responsibility, condition-related mastery), additional analyses were completed to determine if a suppression effect contributed to the significant results (Gaylord-Harden, Cunningham, Holmbeck, & Grant, 2010). To assess for suppression, parental expectations for the future was first allowed to predict responsibility and mastery individually and then this analysis was repeated with each included covariate. When parental expectations for the future alone predicted the outcomes, the results were non-

significant (responsibility: β=.12, p=.26; mastery: β=.16, p=.14), but when including age as an additional independent variable, the associations between parental expectations for the future and the two self-management variables became significant (responsibility: β=.28, p<.01; mastery: β=.28, p<.01). Sobel tests were conducted using the unstandardized coefficients to determine the significance of these suppression effects. The suppression effect of camper age on condition-related responsibility was non-significant ($z = 1.10$, $p = .27$), as was the suppression effect of camper age on condition-related mastery ($z = 1.37$, $p = .17$). When models included previous years at camp as an independent variable, the associations between parental expectations and self-management remained non-significant (p's>.05).

Objective 2. The second objective of this study was to determine the relationships between the independent parent variables and changes in health-related goal attainment, condition-related responsibility, and condition-related mastery as a result of participating in the psychosocial camp-based intervention targeting self-management. Results are summarized in Table 4.

Condition-related responsibility. Parental distress (β=.004, p=.98), general parenting stress (β=.10, p=.47), and SB-specific parenting stress (β=.18, p=.19) were not significantly associated with changes in campers' condition-related responsibility. When including the covariates of age and previous years at camp, parental distress (β=-.01, p=.94), general parenting stress (β=.08, p=.58), and SB-specific parenting stress (β=.14 p=.35) remained not significantly associated with changes in youth's condition-related responsibility.

Table 4. Summary of Regression Analyses Showing Associations Between Parent Variables and Changes in Camper Self-Management Variables and Goal Attainment (Objective 2)

Variable	Condition-Related Responsibility				Condition-Related Mastery				Health-Related Goals			
	Step	b	β	ΔR_2	Step	b	β	ΔR_2	Step	b	β	ΔR_2
Camper Age		.01	.13			.01	.04			-.01	-.03	
Previous Years at Camp	1	-.01	-.22	.04	1	.01	.07	.01	1	-.03	-.08	.01
SB-specific Parenting Stress	2	.14	.14	.02	2	-.17	-.17	.03	3	-.91	-.15	.02
Distress	4	-.01	-.01	.00	4	-.05	-.18	.02	4	-.02	-.01	.00
General Parenting Stress	3	.01	.08	.01	3	.01	.13	.03	2	-.02	-.14	.02
Camper Age		.01	.22			.00	.03			.04	.12	
Previous Years at Camp	1	-.01	-.14	.03	1	.01	.01	.01	1	-.02	-.05	.01
Expectations for the Future	2	-.17	-.17	.03	2	-.29	-.35	.11*	4	-.44	-.08	.01
PPCV	3	.11	.26	.05	4	.08	.24	.03	2	.26	.12	.01
Overprotection	6	.03	.05	.00	6	-.06	-.11	.01	5	-.09	-.03	.00
Optimism	4	-.01	-.11	.01	5	.01	.14	.04	6	-.01	-.01	.00
Illness Uncertainty	5	-.00	-.07	.01	3	-.01	-.18	.02	3	-.01	-.09	.01

Note: All predictor variables are measured at Time 1, and separate regressions were run for two blocks of predictors (Block 1: adjustment variables, Block 2: perceptions/attitudes). Outcome variables indicate changes in self-management variables and goal attainment (e.g., residuals after regressing outcomes at Time 1 on outcomes at Time 3). For all analyses, the covariates of camper age and previous years at camp were entered in Step 1.
*$p < .05$, **$p < .01$

Without covariates, none of the parental perceptions/attitudes/behaviors were significantly associated with youth's condition-related responsibility (PPCV: β=.14, p=.27; overprotection: β=-.07, p=.60; optimism: β=-.13, p=.32; illness uncertainty: β=-.05, p=.76; expectations for the future: β=.17, p=.21). When including the covariates of camper age and previous years at camp in this analysis, parent perceptions/attitudes remained non-significant (PPCV: β=.26, p=.07; overprotection: β=.05, p=.75; optimism: β=-.11, p=.40; illness uncertainty: β=-.07, p=.63; expectations for the future: β=.173, p=.22).

Age was examined as a moderator of the relationships between parent factors and changes in youth condition-related responsibility. There were no significant age x parent factor interactions (all p's > .05).

Condition-related mastery. Without covariates, parental distress (β=-.16, p=.28), general parenting stress (β=.12, p=.39), and SB-specific parenting stress (β=-.17, p=.23) were not significantly associated with changes in camper mastery over condition-related tasks. When including covariates, these associations remained non-significant (parental distress: β=-.18, p=.25; general parenting stress: β=.13, p=.36; SB-specific parenting stress: β=-.17, p=.26).

Similar analyses were completed using parental perceptions/attitudes as independent variables. Parental expectations for the future were found to be significantly associated with changes in campers' condition-related mastery (β=.31, p<.05). When parents reported greater expectations for the future, campers exhibited larger increases in reported mastery following participation in the intervention. This association remained significant when including the covariates of age and previous years at camp (β=.35, p<.01). Other parental perceptions/attitudes were not associated with changes in condition-related mastery with covariates (illness uncertainty: β=-.18, p=.16; PPCV: β=.24, p=.11; optimism: β=.14, p=.28; overprotection: β=-.11, p=.45) or without covariates (illness uncertainty: β=-.19, p=.13; PPCV: β=.20, p=.17; optimism: β=.13, p=.30; overprotection: β=-.13, p=.31) in the analysis.

Age was examined as a moderator of the relationships between parent factors and changes in youth condition-related mastery. There were no significant age x parent factor interactions (all p's > .05).

Health-related goal attainment. Without covariates, parent adjustment variables where not significantly associated with changes in campers' health-related goal attainment (distress:

β=-.02, p=.92; general parenting stress: β=-.13, p=.38; SB-specific parenting stress: β=-.10, p=.49). When including covariates, parental distress (β=-.01, p=.94), general parenting stress (β=-.14, p=.33), and SB-specific parenting stress (β=-.15, p=.33) were not significantly associated with changes in campers' health-related goal attainment. Additionally, parent perceptions/attitudes were not significantly associated with changes in campers' health-related goal attainment both with (PPCV: β=.12, p=.40; illness uncertainty: β=-.09, p=.55; expectations for the future: β=-.08, p=.61; overprotection: β=-.03, p=.87; and optimism: β=-.01, p=.96) and without (PPCV: β=.10, p=.45; illness uncertainty: β=-.08, p=.62; expectations for the future: β= -.02, p=.90; overprotection: β=-.09, p=.53; and optimism: β=-.01, p=.93) covariates.

Objective 3. The third objective of this study was to examine trajectories of the youth self-management variables (independence in completing SB-related tasks, mastery over self-management skills) over the course of two summers, with parent variables as predictors of these trajectories. This was accomplished using multilevel modeling in Mplus.

Prior to testing the hypotheses that parent factors would affect trajectories of condition-related responsibility and mastery, the appropriateness of using a multilevel modeling approach was examined by building null models which contained only the dependent variables and no predictors. Two null models were built: (1) examining trajectories of youth condition-related responsibility, (2) examining trajectories of youth condition-related mastery. To evaluate all subsequent models' fit, standard fit indices were used (Kline, 2006) The robust Satorra-Bentler scaled chi-square statistic (S-B χ2) should be significant; RMSEA and SRMR should be <.08, and CFI and TLI should be < 1.0.

Condition-related responsibility. The null latent growth curve model for condition-related responsibility was found to fit at the latent basis level. Overall, families tended to report

increases in campers' responsibility over time (from Year 1 Time 1 to Year 2 Time 3), however, the fit of a latent basis growth model indicated that this change was represented by a nonlinear pattern (Ram & Grimm, 2013). The mean intercept was 1.98 (SE=.04, $p < .001$), and mean slope was .25 (SE=.04, $p<.001$). An additional model was built including only the initially proposed covariates (camper age, IQ, lesion level, family income, and prior years at camp) in the model. There were significant paths from age (β=.07, $p<.001$) and IQ (β=.01, $p<.05$) to the intercept, indicating that campers with higher ages and IQs had higher initial levels of responsibility. None of the covariates were significantly related to the slope.

Two additional models were created to examine parent adjustment and parent perception/ attitude variables as predictors of change in campers' condition-related responsibility over time. There were no significant effects when including parent adjustment variables (distress, general parenting stress, SB-specific parenting stress) as predictors in the model. When building a model including the parent perceptions/attitudes/behaviors as predictors, none of these parent variables were significantly related to the intercept of the growth curve. However, the path from PPCV to slope was significant (β=-.01., $p<.05$); when parents reported higher levels of PPCV, the slope of the line of growth over time was less steep (e.g., campers acquired responsibility at a slower rate).

Condition-related mastery. The null latent grown curve model for condition-related mastery was found to fit at the linear level. Overall, families tended to report increases in campers' condition-related mastery over time (from Year 1 Time 1 to Year 2 Time 3). The mean intercept was .61 (SE = .03, $p<.001$), and mean slope was .03 (SE = .004, $p<.001$). An additional model was built including only the initially proposed covariates (camper age, IQ, lesion level, family income, and prior years at camp) in the model. There were significant paths from age

(β=.02, p<.05), IQ (β=.094, p<.01), and prior years at camp (β=.04, p<.001) to the intercept. This indicates that older campers, those with higher IQs, and those who had more years of previous camp participation had higher initial levels of mastery. There were also significant paths from age (β=-.003, p<.05) and family income (β=.002, p<.05) to the slope, indicating that younger campers and those from families of higher SES acquired mastery over condition-related tasks at a higher rate. All of the initially proposed covariates were included in subsequent models, as multi-level modeling eliminated concerns about missing data/listwise deletion.

Two additional models were created to examine parent adjustment and parent perception/attitude/behavior variables as predictors of change in campers' condition-related mastery over time. There were no significant effects when including parent adjustment variables (distress, general parenting stress, SB-specific parenting stress) as predictors in the model. When building a model including the parent perceptions/attitudes/behaviors as predictors, none of these parent variables were significantly related to the intercept of the growth curve. However, the path from expectations for the future to slope was significant (β=.05., p<.05); when parents reported greater expectations for the future, the slope of the line of growth over time was steeper (e.g., campers mastery increased more quickly).

Exploratory Analyses

In the current study, many significant relationships were identified among the individual parent factors via Pearson correlations (see Table 2), and research has found parent adjustment variables to be significantly related to parent perceptions in families of youth with SB (e.g., distress/parenting stress → PPCV, Driscoll et al., 2018). IFSMT proposes a pathway from individual/family adjustment to condition self-management outcomes through perceptions, beliefs, and expectations (Ryan & Sawin, 2009), and recent research with families of youth with

SB has extended this model. Specifically, longitudinal analyses have found maternal distress to be significantly negatively associated with youth with SB's condition-related responsibility when this relationship was mediated by maternal PPCV (Driscoll et al., under review). Therefore, exploratory analyses were performed to better understand potential *processes* by which parent adjustment (e.g., distress, general parenting stress, and SB-specific parenting stress) may be related to youth self-management outcomes through parent perceptions/attitudes.

Mediation models were tested using the PROCESS bootstrapping macro for SPSS (Hayes, 2017). The models tested were chosen based on significant cross-sectional correlations between parent adjustment variables and perceptions/attitude variables. The models tested are shown in Figure 5. All analyses included the covariates of camper age and previous years at camp. In all models, both independent and mediating variables were assessed at Time 1, while the outcome variables were assessed at Time 3. Models identified as significant were run a second time, using the residual variables (e.g., those created by controlling for the outcome variable at Time 1). It is important to recognize limitations of these exploratory analyses, given that they only utilize two time points and models were initially run without controlling for the outcome variable at the previous time point. Therefore, these results should be interpreted with extreme caution and viewed as indicators of relationships that require further investigation with more robust data sets.

A significant indirect effect was identified between parental distress at Time 1 and youth mastery over condition-related tasks at Time 3, with parental optimism mediating this relationship (β=-.04, CI: -.10 to -.01). However, this pathway was no longer significant when condition-related mastery at Time 1 was accounted for (e.g., when using the Time 3 residual as the dependent variable; β=-.02, CI: -.08 to .01).

a)
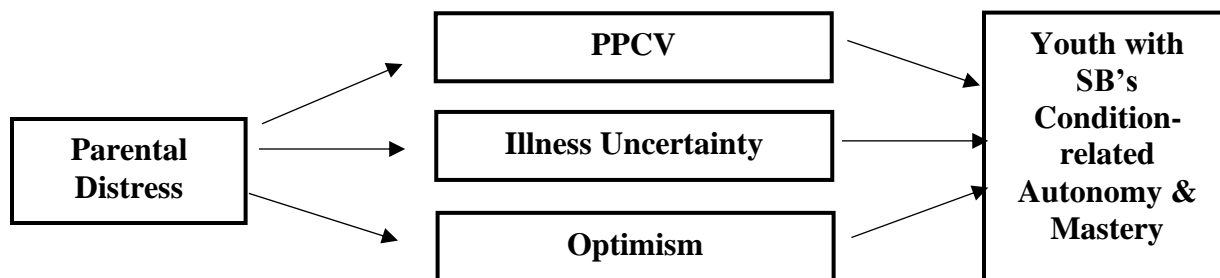
b)
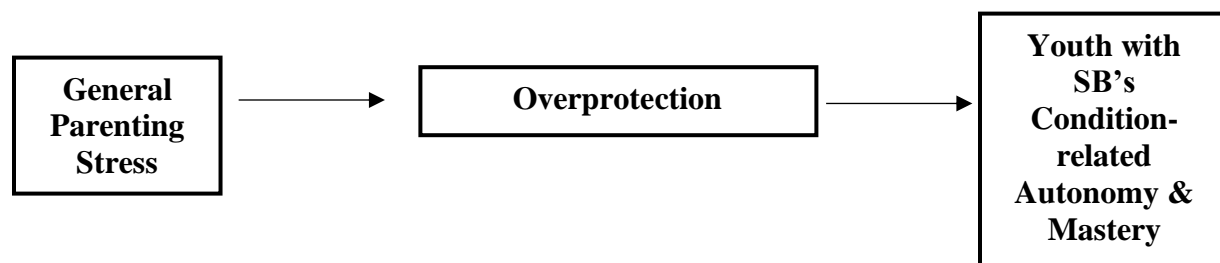
c)
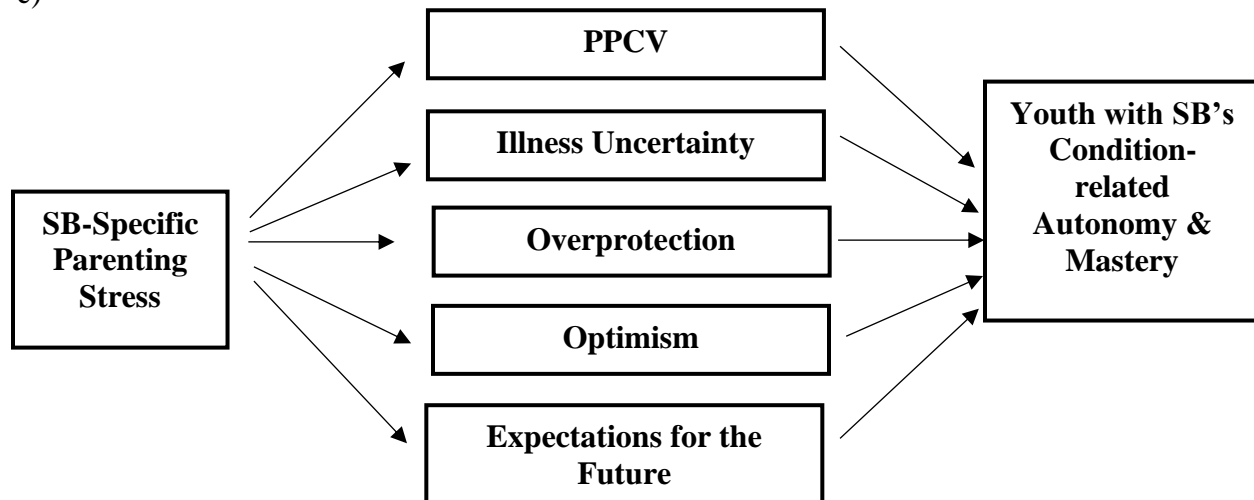
Figure 5. Theoretical models of exploratory mediation analyses using a) parental distress, b) general parenting stress, and c) SB-specific parenting stress as predictors

When using SB-specific parenting stress as the independent variable, significant indirect effects were identified. Specifically, a significant indirect effect was identified between SB-specific parenting stress at Time 1 and youth medical responsibility at Time 3, with parental expectations for the future mediating this relationship (β=-.25, CI: -.70 to -.01). However, this pathway was no longer significant when medical responsibility at Time 1 was accounted for (e.g., when using the Time 3 residual as the dependent variable; β=-.07, CI: -.22 to .01). Parental expectations for the future was also found to mediate a significant indirect effect between SB-specific parenting stress at Time 1 and youth condition-related mastery at Time 3 (β=-.14, CI: -.40 to -.01). This effect remained significant when using the Time 3 mastery residual as the dependent variable (β=-.08, CI: -.21 to -.01).

No significant mediational models including general parenting stress as the independent variable were identified. Additionally, no other models using parental distress of SB-specific parenting stress (apart from those described above) were identified as significant.

CHAPTER FIVE

DISCUSSION

Many pediatric health conditions, including SB, require lifelong adherence to a medical regimen (Modi et al., 2012). The development and maintenance of self-management behaviors for youth with these conditions are important steps towards independence (Bakaniene, Ziukiene, Vasilaiskiene, & Prasauskiene, 2018). However, the acquisition of condition-related skills and the transfer of condition-related responsibilities from caregivers to individuals with SB are often challenging processes. The purpose of the current study was, broadly, to identify parent factors that may affect the development of condition-related autonomy for youth with SB. This study examined the relationships among parent personal adjustment factors, parental perceptions/ attitudes/behaviors, and youth condition-related responsibility and mastery cross-sectionally and longitudinally in the context of a summer camp independence intervention study.

A number of significant relationships between the independent parent factors and dependent youth self-management variables were identified, and these will be discussed below. However, first, it should be noted that, though conceptualized at separate constructs, the dependent self-management variables of condition-related responsibility and condition-related mastery were highly positively correlated. Further, growth analyses indicated that youth with SB's condition-related responsibility and mastery each increased over time for many youth. This indicates that youth with greater levels of responsibility also had higher levels of mastery over condition-related tasks. These results are promising, because increase in responsibility without

increase in skill may lead to poor adherence (Psihogios et al., 2015), and, subsequently, the occurrence of secondary medical complications. Given their conceptualization as unique constructs, findings for condition-related responsibility and mastery will be discussed individually below. However, findings should be interpreted and applied in the context of this significant relationship between the self-management outcome variables.

Parental Expectations for the Future

There were a number of significant findings for the parent perception of expectations for the future, including significant findings in cross-sectional, longitudinal (over one summer), and growth (across two summers) analyses. When including covariates in (camper age and previous years at camp) in cross-sectional analyses, parental expectations for the future were significantly associated with both youth with SB's condition-related responsibility and mastery. Moreover, this relationship was moderated by camper age – when parents of *older* campers held higher expectations for campers' milestone attainment, campers also had greater responsibility and parents reported higher mastery of condition-related skills. Additionally, higher parental expectations for the future were associated with larger increases over time in campers' *mastery* over condition-related skills following participation in the camp intervention regardless of camper age. Further, growth analyses revealed that when parents reported higher expectations for the future at baseline, youth's mastery over SB-related tasks improved more rapidly. Research with typically-developing youth and youth with disabilities suggests that parental expectations regarding an adolescent child's abilities and milestone achievement are strong predictors of adolescent and young adult outcomes and proposes that parents behave and relate to their children in ways that align with their expectations (Doren, Gau, & Lindstrom, 2012). In youth with SB, this may manifest as parents promoting condition-related autonomy, as this is a

necessary step for youth with SB to become more independent and meet developmentally-normative milestones. This relationship may become significant for older campers, as these campers are closer to reaching the age at which some milestones are expected to be met.

One interpretation of these findings is that when parents expect their children to reach certain milestones, they likely create an environment that promotes the skills necessary to meet these milestones (e.g., by providing opportunities to practice skills, by utilizing a scaffolding approach). In line with this, youth's mastery over condition related tasks (e.g., clean intermittent catheterization, bowel programs) has been found to be significantly related to youth's community participation (Bakaniene et al., 2018). Community participation can be considered a precursor to broader autonomy.

Overall, parents of young people with SB have been found to decrease their expectations for future milestone achievement over time (Holbein et al., 2016). This same pattern was observed with the current study's data, as camper age was significantly negatively associated with parental expectations for the future cross-sectionally. Notably, bivariate correlations in the current study found that camper IQ was significantly positively associated with parents' expectations for the future, and parents of youth with SB's expectations for the future have been found to vary based on youth's cognitive abilities (Holbein et al., 2016; Wasserman & Holmbeck, 2016; Zukerman et al., 2011). Youth with SB demonstrate a unique profile of neuropsychological strengths and challenges, but a range of functioning within these domains has also been documented (Copp et al., 2015; Wasserman & Holmbeck, 2016). Additionally, the neurological sequelae of SB (e.g., hydrocephalus, Chiari II malformation) and their subsequent medical treatment have potential to impact neuropsychological outcomes. For example, the presence of hydrocephalus and differential treatment approaches (e.g., shunt-treated versus non),

have been associated with differing performances on tests across numerous neuropsychological domains (Hampton et al., 2011). Future research should examine how youth's cognitive abilities (including overall intellectual functioning, attention, executive functioning, and learning) are related to both parental expectations for their child's future and youth with SB's self-management behaviors. Executive dysfunction has been identified as a particularly important factor, both in terms of individuals with SB's milestone achievement and self-management (Stern et al., 2018; Zukerman et al., 2011). A better understanding of the contributions of cognitive abilities to specific self-management behaviors and, ultimately, autonomy would be useful for the development of interventions tailored specifically to individuals with SB.

However, though not examined in the current study, it is probable that this relationship between parent expectations and youth self-management is bidirectional. There are a number of other, condition-related factors that contribute to youth with SB's *potential* for condition-related independence, including attention abilities, executive functioning, gross motor functioning, and fine motor coordination (Copp et al., 2015; Stern et al., 2018). Campers with dysfunction in these areas may struggle to master condition-related skills and assume responsibility for condition-related tasks, and, subsequently, their parents may have lowered expectations for campers' future milestone achievement. It is also possible that when parents have higher expectations for campers, the campers have higher expectations for themselves. Campers may then have intrinsic motivation (internal drive to perform an activity for its inherent interest and enjoyment) to utilize opportunities, such as those presented at camp, to improve their condition-related skills (Zukerman et al., 2011). Future research is needed to better understand all factors that contribute to this, likely bidirectional, relationship.

Other Significant Parental Perceptions and Behaviors

Cross-sectional analyses yielded different results when including or excluding covariates. Without covariates, parental overprotection was the only parent factor found to be significantly associated with both youth with SB's condition-related responsibility and condition-related mastery. In youth with other chronic health conditions, parental overprotection has both positive and negative effects. Parental overprotection and structure are associated with improved physical and medical outcomes (Crandell, Sandelowski, Leeman, Havill, & Knafl, 2018). However, overprotection has also been associated with negative psychosocial outcomes, including limited youth autonomy and social competence (Hoehn, Foxen-Craft, Pinder, & Dahlquist, 2016; Holmbeck et al., 2002). These negative psychosocial outcomes most likely occur when parents are overly protective, imposing limits that youth perceive as unnecessary (Stiernman, Osterlind, Rumsey, Becker, & Persson, 2019). Parents of youth with SB have been found to be more overprotective than parents of children without a medical condition, and these youth with SB have been described as having limited decision making autonomy, social immaturity, few interactions outside of school, and dependence on adults for guidance (Holmbeck et al., 2002; Holmbeck et al., 2003) When age was included in analyses, parental overprotection was no longer a significant predictor. This indicates that for campers with SB, parental overprotection seems to, appropriately, decrease with changes in camper age. This also indicates that this significant finding is likely not clinically meaningful as age is a very important factor when working with children.

Another parental perception was identified as a potentially significant predictor of self-management for youth with SB in this study's growth analyses. Parent factors did not influence the intercepts of growth in these analyses (e.g., the initial level of reported responsibility and

mastery). However, when parents reported higher levels of PPCV, youth with SB acquired condition-related skills at a slower rate. When parents perceive their children with SB to be more vulnerable, they may be hesitant to allow them to take responsibility for medical tasks. This finding is line with the study's hypotheses and previous research. In fact, in another longitudinal study with families of youth with SB, higher maternal PPCV was associated with lower subsequent levels of youth condition-related responsibility (Driscoll et al., under review).

Parent Adjustment

Across all analyses, the three parent adjustment factors (distress, general parenting stress, SB-specific parenting stress) were not significantly directly associated with youth self-management variables. Previous research with youth with SB has found that youths' interaction styles with their parents during observed tasks are consistent, regardless of family or parent characteristics or behaviors (Holmbeck et al., 2002; Lennon, Murray, Bechtel, & Holmbeck, 2015). Specifically, youth are more passive and less actively involved during family discussions. It is possible that youth with SB's unique interpersonal/interaction style protects them from the potential negative impact of poor parental adjustment. It is notable that the baseline levels of distress, general parenting stress, and SB-specific parenting stress reported by parents in this study were each low. It is possible that the parents of campers with SB included in this study did not experience the challenges with adjustment that has been reported by other parents of youth with SB or that these parents have found adaptive ways to manage distress and stress. Alternatively, is possible that parental distress and parenting stress decreased as a result of camp itself. For example, parents may experience some relief at the start of camp knowing that they will have fewer responsibilities for the week and this may affect their report on the measures of adjustment used in this study.

Additionally, previous studies have demonstrated that mothers and fathers take on differential roles and adjust differently when parenting a child with SB (Brekke, Fruh, Kvarme, & Holstrom, 2017; Driscoll et al., 2018). Though a majority of the current study's parent participants were female caregivers, all caregivers (male and female; biological parent versus other caregiver) were included in the same group in this study's analyses. Given his heterogeneity may have affected our ability to identify specific relationships between parent personal adjustment and youth self-management variables.

Campers' Response to Intervention: A Single Summer

Many of the study's analyses examining change over a single summer were non-significant. Specifically, parent factors were not related to changes in youth responsibility for condition-related tasks or health-related goal achievement following participation in the camp-based intervention. While these outcome variables were both found to increase significantly from pre- to post-camp, the hypothesized parent factors were not significantly related to these changes over a single summer. It is possible that, because changes in youth with SB's behavior occurred in the summer camp setting (e.g., away from their parents with repeated reinforcement and assistance with goal-directed problem solving from an alternate caregiver [e.g., camp counselor]), the proposed parent factors were unrelated to gains made. It is also possible that youth with SB's unique interpersonal style observed in the home (described above; Holmbeck et al., 2002; Lennon et al., 2015), allowed them to make gains in responsibility and goal achievement following camp regardless of parent characteristics. Future research should explore other camper-, parent-, and family-level variables that may affect campers' response to the summer-camp based intervention.

Exploratory Analyses

The exploratory analyses identified some pathways through which parent adjustment may be related to youth self-management. These pathways are important to consider, as specific family processes have been identified as a target area of research in the broader field of pediatric psychology (Alderfer et al., 2017). However, given limitations (discussed in greater detail below), these results should be interpreted with extreme caution and considered preliminary evidence to support further investigation with more robust samples.

Parental expectations for the future were identified as a significant mediator of the indirect relationship between SB-specific parenting stress and both youth self-management variables. These relationships were significant when including covariates of camper age and prior years at camp and they were in the anticipated direction: as parents experience higher levels of condition-specific stress, they have lower expectations for their child with SB's future milestone attainment, and youth demonstrate lower levels of responsibility and poorer condition related mastery.

Parental distress was significantly related to youth's responsibility for condition-related tasks when this relationship was mediated by parental optimism. A similar negative relationship between parental depression and optimism has been found in parents of children with type 1 diabetes (Zayas, Guil, Guerrero, Gil-Olarte, & Mestre, 2016), and it was proposed that intervening to reduce distress and increase optimism in parents may lead to positive youth outcomes as well. In the current study, when parents reported higher levels of distress and lower levels of optimism, youth had lower levels of condition-related responsibility. It has been recommended previously that pediatricians screen caregivers (mothers, specifically) for depressive symptoms (Olson et al., 2005). However, the results of these exploratory analyses

indicate that it may be beneficial to regularly assess other parental factors (stress, optimism, expectations for the future) as the *interaction* of these factors may lead to non-optimal outcomes for family and youth. Further, it has been suggested that the clinicians work to "match" families to psychosocial interventions that meet their specific needs (Canter, 2019; Kazak, Schneider, Didonato, & Pai, 2015). While a more thorough screening may be more time-intensive, it also may help facilitate more appropriate "matching" of families to appropriate interventions.

Study Limitations

There are limitations to be considered when interpreting the results of this study as a whole. First, there is potential for sample bias, as the participants were self-selected attendants at a summer camp. Second, due to a high number of repeat (versus new) campers in the second summer, the overall sample size was smaller than anticipated and may have limited the ability to detect significant effects (specifically for Objective 2). Third, it is possible that common-method variance contributed to some significant findings in this study, as parents reported using paper-and-pencil questionnaires for many of the study's variables. Fourth, the time between assessments (one month) may not have been long enough to detect significant effects, especially given the time of year that the study was completed, as many families are out of their typical routines during the summertime. Future research with a larger sample size in a more generalizable setting could address these limitations.

There are additional limitations to be considered when interpreting the results of the exploratory analyses. First, the mediation models only utilized two time points (assessing the independent and mediator variables at the same time point). Second, youth participated in the camp intervention between Time 1 and Time 3, meaning that changes in self-management skills may not necessarily be those that would occur naturally without intervention. Lastly, two of the

three identified pathways were no longer significant when controlling for the self-management variable at the previous time point. Therefore, further research is necessary to identify and understand these pathways from parent adjustment → parent perceptions → youth self-management outcomes.

Directions for Future Research

The results of the current study identify areas for future research. Future work could expand on this study as discussed previously, (e.g., by using a larger sample size, assessing mothers and fathers of youth with SB separately, considering the different neuropsychological domains and/or other external factors that may impact both parent factors and youth self-management).

Most notably, it would be beneficial for future research to explore the potential bidirectional relationships between this study's parent factors and self-management. Research with families of youth with SB has found that ambulation status (e.g., independent vs. assisted ambulation) significantly impacts parent adjustment such that when youth can ambulate independently, parents report lower levels of stress (Antiel et al., 2016). It could be extrapolated, then, that when youth gain independence in other domains, parental psychological adjustment would also improve. However, independence would require both youth *responsibility* for condition-related tasks and *mastery* over these tasks. For example, it is probable that if youth have poor mastery over condition-related skills and require substantial supervision to complete SB-related tasks, that parents could experience higher levels of distress and stress and more negative perceptions (e.g., perceiving their child as more highly vulnerable). Additionally, there are likely other youth factors (e.g., youth adjustment, youth perceptions/attitudes, parent coping)

that contribute both to self-management behaviors and to parent adjustment, perceptions, and attitudes. However, these types of youth factors were not assessed in the current study.

Further, the current study utilized total scores for the included self-management variables. However, it may be beneficial for future research to examine the associations between parent variables and specific condition-related tasks (e.g., clean intermittent catheterization). Specific aspects of the management of SB have been found to be particularly stressful for parents (e.g., limited mobility, managing bowel and bladder dysfunction; Antiel et al., 2016; Kanaheswari et al., 2011). Therefore, it may be beneficial to break down the self-management variables (e.g., responsibility and mastery) into individual components and examine the associations between these specific components of self-management and the parent factors. This could allow for future interventions to be designed to address specific processes that are most challenging or stressful for families.

Clinical Implications

The results of the current study also have important implications for work with families of youth with SB. Parental overprotection, PPCV, and parental expectations for the future have been identified as potential targets for assessment. It would be beneficial for these perceptions and behaviors to be assessed when families present to SB health-care providers, as they have the potential to affect SB outcomes, including aspects of self-management.

These perceptions and behaviors are also potential targets for parent- and family-based interventions. Given the heterogeneous nature of the complications and medical sequelae associated with SB, it would be difficult to design a "one-size-fits-all" intervention. However, parents of youth with SB could benefit from cognitive-behavioral interventions that target potentially maladaptive thoughts/perceptions and unhelpful behaviors (e.g., Levy et al., 2016).

The exploratory analyses highlight potential processes by which parent factors interact to affect youth self-management. Additionally, regardless of parent variables, youth with SB respond positively to the goals-focused camp intervention (Driscoll et al., 2019; Holbein et al., 2013; O'Mahar et al., 2009). Families may benefit from similar problem-solving focused interventions that draw on both family and patient strengths to make up for specific challenges (e.g., Narad et al., 2019). These types of interventions could be delivered in a brief, skills-focused, group format (e.g., in a single session) or in an individual, tailored format. For the latter type of therapy, it may be helpful to collect information from the medical team about the child with SB's physical and cognitive functioning, as well as the medical team's estimation of youth with SB's potential for independence, as parents and the medical team frequently disagree on these issues (DeMaso et al., 1991). Information from the medical team may provide support for challenging potentially unhelpful beliefs and perceptions.

Conclusions

In summary, this study has identified specific parent perceptions and behaviors that are associated with aspects of self-management for youth with SB. Parental expectations for their child with SB's future milestone attainment was consistently associated with youth's condition-related responsibility and mastery. Parental overprotection and PPCV were also identified as potentially significant contributors to self-management. These may be important targets of assessment and intervention when working with families of youth with SB as well as areas for future research.

APPENDIX A

MEASURES

Demographic Information

1. Camper gender *(circle one)*   Male   Female

2. Camper birthdate (Month/Day/Year): _____/_____/_____

3. Camper age: _____

4. Camper ethnicity:
      a. _____ African American
      b. _____ Asian
      c. _____ Caucasian
      d. _____ Hispanic
      e. _____ Other (*please list*): -
      _____

4. **Your** gender *(circle one)*   Male   Female

5. Check the highest level of education that **you** completed:
      a. ___ Some grade school
      b. ___ Finished grade school
      c. ___ Some high school
      d. ___ Finished high school
      e. ___ Business or technical school
      f. ___ Some college
      g. ___ Finished college
      h. ___ Attended graduate school or professional school after college
      i. ___ Received a professional degree
      j. ___ I am currently enrolled in the following: _____

6. Check the highest level of education that your **SPOUSE / SIGNIFICANT OTHER** completed:
      a. ___ Some grade school
      b. ___ Finished grade school
      c. ___ Some high school
      d. ___ Finished high school
      e. ___ Business or technical school
      f. ___ Some college
      g. ___ Finished college
      h. ___ Attended graduate school or professional school after college
      i. ___ Received a professional degree
      j. ___ S/he is currently enrolled in the following: _____

7. What is **your** current EMPLOYMENT status? (*please circle one*)
      a. Full-time homemaker (does not work outside the home)

b. Retired
c. On disability from work
d. Employed part-time
e. Employed full-time
f. Other (please explain):

_____

8. If **you** are EMPLOYED part-time or full-time, please describe your job:
    a. Where do you work? _____
    b. What kind of work do you do? _____
    c. How many hours per day do you work? _____

9. What is your **SPOUSE / SIGNIFICANT OTHER's** current EMPLOYMENT status?
(*Please circle one.*)
    a. Full-time homemaker (does not work outside the home)
    b. Retired
    c. On disability from work
    d. Employed part-time
    e. Employed full-time
    f. Other (*Please explain*):

_____

10. If your **SPOUSE / SIGNIFICANT OTHER** is EMPLOYED part-time or full-time, please describe
    his/her job:
    a. Where does s/he work? _____
    b. What kind of work does s/he do? _____
    c. How many hours per day does s/he work? _____

11. What is your family's total yearly income? *(Please circle one.)*

1.	Under $10,000	12.	$110,000 - $119,999
2.	$10,000 - $19,999	13.	$120,000 - $129,999
3.	$20,000 - $29,999	14.	$130,000 - $139,999
4.	$30,000 - $39,999	15.	$140,000 - $149,999
5.	$40,000 - $49,999	16.	$150,000 - $159,999
6.	$50,000 - $59,999	17.	$160,000 - $169,999
7.	$60,000 - $69,999	18.	$170,000 - $179,999
8.	$70,000 - $79,999	19.	$180,000 - $189,999
9.	$80,000 - $89,999	20.	$190,000 - $199,999
10.	$90,000 - $99,999	21.	Over $200,000
11.	$100,000 - $109,999		

Previous Camp Experience Information

12. The total number of times camper has attended Camp Ability/Camp Independence is _____
(Note: If this is their first year, please put 0)

Spina Bifida Related Information

13. What type of spina bifida does your camper have?
    a. _____ Occulta
    b. _____ Lipomeningocele (lipo)
    c. _____ Meningocele
    d. _____ Myelomeningocele (MM)
    e. _____ Not sure

14. What is the level of your camper's lesion?
    a. _____ Thoracic
    b. _____ Lumbar
    c. _____ Sacral
    d. _____ Not sure

15. Does your camper have a shunt?
    a. _____ Yes
    b. _____ No
    c. _____ Not sure

    IF YES, Total Number of Shunt Surgeries/Revisons: _____

16. What does your camper use to get from place to place (ambulation)? *Please check all that apply*:
    a. _____ Nothing.
    b. _____ Ankle-foot braces (AFOs)
    c. _____ Knee-ankle-foot braces (KFOs)
    d. _____ Hip-knee-ankle-foot braces
    e. _____ Reciprocating brace
    f. _____ Swivel walker
    g. _____ Parapodium
    h. _____ Twister cables
    i. _____ Night splint
    j. _____ Back brace
    k. _____ Crutches
    l. _____ Walker
    m. _____ Wheelchair

17. If your camper uses more than one mobility device, please write down the percentage of time that s/he uses each device (please make sure that the percentages add up to 100%):

   ____ % Unassisted walking (no braces)
   ____ % Braces alone (no crutches or walker)
   ____ % Braces with crutches or walker
   ____ % Wheelchair

Sharing of Spina Bifida Management Responsibilities-P

For each of the following parts of spina bifida care, choose the number of the answer that best describes the way you handled things at home **during the last month**.

CAMPER-Camper took or initiated responsibility for this almost all of the time, by him/herself.
EQUAL-Parent(s) and camper shared responsibility for this about equally.
PARENT-Parent(s) took or initiated responsibility for this almost all of the time.
N/A- Not Applicable. This does not describe a part of your camper's spina bifida care.

Who Has Responsibility?

	CAMPER	EQUAL	PARENT	N/A
1. Remembering day of clinical appointment.	☐	☐	☐	☐
2. Making appointments with doctors.	☐	☐	☐	☐
3. Talking with doctors about medical questions and requests (e.g., medication refill).	☐	☐	☐	☐
4. Explaining absences from school/work to teachers or other personnel.	☐	☐	☐	☐
5. Telling teachers/supervisors about spina bifida.	☐	☐	☐	☐
6. Telling relatives about spina bifida.	☐	☐	☐	☐
7. Telling camper's friends about spina bifida.	☐	☐	☐	☐
8. Remembering to take medication, as prescribed.	☐	☐	☐	☐

N/A- Not Applicable. This does not describe a part of your camper's spina bifida care.

Who Has Responsibility?

	CAMPER	EQUAL	PARENT	N/A
9. Checking expiration dates on medical supplies.	☐	☐	☐	☐
10. Taking proper care of wheelchair and braces.	☐	☐	☐	☐
11. Wearing orthotics (braces) as prescribed by doctor/physical therapist.	☐	☐	☐	☐
12. Getting around in wheelchair from place to place inside of the home.	☐	☐	☐	☐
13. Getting around in wheelchair from place to place outside of the home.	☐	☐	☐	☐
14. Getting in and out of wheelchair.	☐	☐	☐	☐
15. Taking care of basic needs (e.g., bathing, dressing).	☐	☐	☐	☐
16. Avoiding products that may contain latex, if allergic to latex.	☐	☐	☐	☐
17. Protecting his/her skin from temperature, textures, and injury.	☐	☐	☐	☐
18. Conducting daily skin checks.	☐	☐	☐	☐
19. Taking medications for urinary tract infection.	☐	☐	☐	☐

20. Noticing differences in urine that could indicate a urinary tract infection.   ☐   ☐   ☐ | ☐

N/A- Not Applicable. This does not describe a part of your camper's spina bifida care.

Who Has Responsibility?

	CAMPER	**EQUAL**	**PARENT**	**N/A**
21. Remembering to catheterize regularly, every 2-4 hours.	☐	☐	☐	☐
22. Washing hands and genital area before catheterizing.	☐	☐	☐	☐
23. Gathering appropriate catheterization equipment (e.g., catheter, lubricant)	☐	☐	☐	☐
24. Lubricating catheter.	☐	☐	☐	☐
25. Properly inserting catheter.	☐	☐	☐	☐
26. Draining bladder completely and removing catheter.	☐	☐	☐	☐
27. Cleaning, storing, and discarding catheterization equipment properly.	☐	☐	☐	☐
28. Following a regular physical exercise routine.	☐	☐	☐	☐
29. Remembering to eat foods with fiber and avoiding other foods (e.g., chocolate).	☐	☐	☐	☐
30. Remembering to drink fluid.	☐	☐	☐	☐
31. Taking suppositories, enemas, stool softeners, or laxatives as needed.	☐	☐	☐	☐
32. Maintaining a regular bowel toileting time.	☐	☐	☐	☐

33. Cleaning up after him/herself, if an accident occurred.   ☐   ☐   ☐ | ☐

34. Monitoring bowel functioning by keeping a log.   ☐   ☐   ☐ | ☐

Sharing of Spina Bifida Management Responsibilities-C

For each of the following parts of spina bifida care, choose the number of the answer that best describes the way you handled things at home **during the last month**.

CAMPER-Camper took or initiated responsibility for this almost all of the time, by him/herself.
EQUAL-Parent(s)/Other(s) and camper shared responsibility for this about equally.
PARENT-Parent(s)/Other(s) took or initiated responsibility for this almost all of the time.
N/A- Not Applicable. This does not describe a part of your camper's spina bifida care.

Who Has Responsibility?

	CAMPER	EQUAL	PARENT/OTHER	N/A
1. Remembering day of clinical appointment.	☐	☐	☐	☐
2. Making appointments with doctors.	☐	☐	☐	☐
3. Talking with doctors about medical questions and requests (e.g., medication refill).	☐	☐	☐	☐
4. Explaining absences from school/work to teachers or other personnel.	☐	☐	☐	☐
5. Telling my teachers/supervisors about spina bifida.	☐	☐	☐	☐
6. Telling my relatives about spina bifida.	☐	☐	☐	☐

7. Telling my friends about spina bifida.　□　□　□　│　□

8. Remembering to take medication, as prescribed.　□　□　□　│　□

N/A- Not Applicable. This does not describe a part of your camper's spina bifida care.

Who Has Responsibility?

	CAMPER	EQUAL	PARENT/OTHER	N/A
9. Checking expiration dates on medical supplies.	□	□	□	□
10. Taking proper care of wheelchair and braces.	□	□	□	□
11. Wearing orthotics (braces) as prescribed by doctor/physical therapist.	□	□	□	□
12. Getting around in wheelchair from place to place inside of the home.	□	□	□	□
13. Getting around in wheelchair from place to place outside of the home.	□	□	□	□
14. Getting in and out of wheelchair.	□	□	□	□
15. Taking care of basic needs (e.g., bathing, dressing).	□	□	□	□
16. Avoiding products that may contain latex, if allergic to latex.	□	□	□	□
17. Protecting my skin from temperature, textures, and injury.	□	□	□	□

18. Conducting daily skin checks.

☐	☐	☐	☐

19. Taking medications for urinary tract infection.

☐	☐	☐	☐

20. Noticing differences in urine that could indicate a urinary tract infection.

☐	☐	☐	☐

N/A- Not Applicable. This does not describe a part of your camper's spina bifida care.

Who Has Responsibility?

	CAMPER	EQUAL	PARENT/OTHER	N/A
21. Remembering to catheterize regularly, every 2-4 hours.	☐	☐	☐	☐
22. Washing hands and genital area before catheterizing.	☐	☐	☐	☐
23. Gathering appropriate catheterization equipment (e.g., catheter, lubricant)	☐	☐	☐	☐
24. Lubricating catheter.	☐	☐	☐	☐
25. Properly inserting catheter.	☐	☐	☐	☐
26. Draining bladder completely and removing catheter.	☐	☐	☐	☐
27. Cleaning, storing, and discarding catheterization equipment properly.	☐	☐	☐	☐
28. Following a regular physical exercise routine.	☐	☐	☐	☐

29. Remembering to eat foods with fiber and avoiding other foods (e.g., chocolate). ☐ ☐ ☐ ☐

30. Remembering to drink fluids. ☐ ☐ ☐ ☐

31. Taking suppositories, enemas, stool softeners, or laxatives as needed. ☐ ☐ ☐ ☐

32. Maintaining a regular bowel toileting time. ☐ ☐ ☐ ☐

33. Cleaning up after myself if an accident occurred. ☐ ☐ ☐ ☐

34. Monitoring bowel functioning by keeping a log. ☐ ☐ ☐ ☐

Spina Bifida Independence Survey

Instructions: For each of the following spina bifida skills, please check "YES" if your camper has mastered that skill, "NO" if your camper has not mastered that skill, or "NOT SURE" if you do not know. Mastery of a given skill means that your camper can perform it correctly without any kind of help from another person. Please remember that we are interested in what your camper is able to do and not in what he or she actually does. Try to ignore your camper's cooperation with treatment as you fill out this survey. If the skill is not relevant to your camper's medical management, please mark "N/A." If you are not sure about whether your camper is able to do the skill, please mark "NOT SURE."

Can your camper:	Yes	No	Not Sure	N/A
1. Recognize symptoms of hydrocephalus/shunt malfunction and tell someone else about it (e.g., headache, change in appetite, deterioration in school performance)	☐	☐	☐	☐
2. Notice changes in health (e.g., weight gain, skin, stool)	☐	☐	☐	☐
3. Ask for help for health-related issues	☐	☐	☐	☐

Can your camper:	Yes	No	Not Sure	N/A
4. Identify appropriate professionals for specific problems	☐	☐	☐	☐
5. Arrange for transportation to and from a health care facility if such a clinic visit is necessary	☐	☐	☐	☐
6. Take medications appropriately (e.g., timing, dose)	☐	☐	☐	☐
7. Fill prescriptions	☐	☐	☐	☐
8. Recognize and discard expired medication products	☐	☐	☐	☐
9. State each type of medication he/she uses	☐	☐	☐	☐
10. State the reasons why it is especially important for an individual with spina bifida to follow a healthy diet (e.g., bowel functioning)	☐	☐	☐	☐
11. Identify foods that are important to include in his/her diet (e.g., fiber and calcium-rich foods) and foods to avoid (e.g., chocolate, cheese)	☐	☐	☐	☐
12. Maintain a healthy diet, including appropriate fluid intake	☐	☐	☐	☐
13. Perform a physical exercise routine on a regular basis	☐	☐	☐	☐
14. Understand the benefits of exercise for an individual with spina bifida	☐	☐	☐	☐

Can your camper:	Yes	No	Not Sure	N/A
15. Maneuver in and out of his/her wheelchair	☐	☐	☐	☐
16. Maintain wheelchair and orthotic devices and use them properly.	☐	☐	☐	☐
17. Dress him/herself independently.	☐	☐	☐	☐
18. Bath him/herself independently.	☐	☐	☐	☐
19. State different products that may contain latex.	☐	☐	☐	☐
20. Conduct daily skin checks	☐	☐	☐	☐
21. Understand why skin care is especially important for individuals with spina bifida (e.g., pressure sores, infection)	☐	☐	☐	☐
22. Protect skin from potential damage (e.g., extreme temperature, cuts)	☐	☐	☐	☐
23. Recognize skin warning signs (e.g., redness, swelling, fever, blister and sores)	☐	☐	☐	☐
24. Recognize symptoms of a urinary tract infections (e.g., fever, stomach ache, smelly and/or cloudy urine, or blood in urine)	☐	☐	☐	☐
25. State catheterization steps	☐	☐	☐	☐

Can your camper:	Yes	No	Not Sure	N/A
26. Conduct each catheterization step correctly	☐	☐	☐	☐
27. Wash hands and genital area before catheterizing.	☐	☐	☐	☐
28. Remember and gather appropriate catheterization equipment (e.g., catheter, lubricant)	☐	☐	☐	☐
29. Lubricate and hold catheter.	☐	☐	☐	☐
30. Properly insert catheter.	☐	☐	☐	☐
31. Know when and how to remove catheter.	☐	☐	☐	☐
32. Store used catheters properly, in a dry environment	☐	☐	☐	☐
33. Clean, discard, and replace catheters as needed	☐	☐	☐	☐
34. Remember to complete catheterization regularly, every 2-4 hours	☐	☐	☐	☐
35. Understand the importance of hygiene and how it relates to care of catheterization and bowel management equipment.	☐	☐	☐	☐
36. Recognize bowel warning signs (e.g., diarrhea, constipation)	☐	☐	☐	☐
37. Use suppositories, enemas, stool softeners, and/or laxatives correctly	☐	☐	☐	☐
38. Understand the importance of a regular toileting time	☐	☐	☐	☐
39. Use a log sheet to keep a schedule and a record of bowel movements	☐	☐	☐	☐

Can your camper:	Yes	No	Not Sure	N/A
40. Clean up after his/herself, if a bowel or urinary accident occurs	☐	☐	☐	☐
41. Prevent constipation through daily monitoring of stool and bowel functioning	☐	☐	☐	☐
42. Understand that spina bifida causes the bowel not to work in the same way as in typically developing individuals and that special bowel programs help individuals with spina bifida achieve continence	☐	☐	☐	☐
43. Participate in choosing a bowel program that will achieve continence	☐	☐	☐	☐
44. Learn steps of a bowel program	☐	☐	☐	☐
45. Carry out steps of a bowel program	☐	☐	☐	☐
46. Understand the consequences of not following a bowel program (e.g., soiled clothing, social consequences)	☐	☐	☐	☐
47. Understand health risks of choosing not to do a bowel program (e.g., skin breakdown, increased wetness, shunt malfunction, and colon cancer)	☐	☐	☐	☐
48. Call a nurse of doctor for help if bowel accidents, diarrhea, or constipation occur.	☐	☐	☐	☐

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**
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
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

	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
48. Trouble getting your breath	0	1	2	3	4
49. Hot or cold spells	0	1	2	3	4
50. Having to avoid certain things, places, or activities because they frighten you	0	1	2	3	4
51. Your mind going blank	0	1	2	3	4
52. Numbness or tingling in parts of your body	0	1	2	3	4
53. A lump in your throat	0	1	2	3	4
54. Feeling hopeless about the future	0	1	2	3	4
55. Trouble concentrating	0	1	2	3	4
56. Feeling weak in parts of your body	0	1	2	3	4
57. Feeling tense or keyed up	0	1	2	3	4
58. Heavy feelings in your arms or legs	0	1	2	3	4
59. Thoughts of death or dying	0	1	2	3	4
60. Overeating	0	1	2	3	4
61. Feeling uneasy when people are watching or talking about you	0	1	2	3	4

62. Having thoughts that are not your own	**0**	**1**	**2**	**3**	**4**
63. Having urges to beat, injure, or harm someone	**0**	**1**	**2**	**3**	**4**
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	**0**	**1**	**2**	**3**	**4**

achievements					
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**
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**

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.

Strongly Disagree	Disagree	Agree	Strongly Agree
1	2	3	4

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

_____ 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.

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

	Not at all stressful	A bit stressful	Fairly stressful	Quite stressful	Extremely stressful
8. Communication with my child.	1	2	3	4	5
9. My child's relations with other children.	1	2	3	4	5
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

VCS – P

Please answer the following on the scale provided.

	Definitely True 1	Mostly True 2	Mostly False 3	Definitely False 4
1. In general, my child seems less healthy than other children of the same age.	1	2	3	4
2. I often think about calling the doctor about my child.	1	2	3	4
3. When there is something going around, my child usually catches it.	1	2	3	4
4. My child seems to have more accidents and injuries than other children.	1	2	3	4
5. My child usually has a healthy appetite.	1	2	3	4
6. Sometimes I get concerned that my child doesn't look as healthy as he/she should.	1	2	3	4
7. My child usually gets stomach pains or other sorts of pains.	1	2	3	4
8. I often have to keep my child indoors because of health reasons.	1	2	3	4
9. My child seems to have as much energy as other children of the same age.	1	2	3	4
10. My child gets more colds than other children of the same age.	1	2	3	4
11. I get concerned about the circles under my child's eyes.	1	2	3	4
12. I often check on my child at night to make sure he/she is okay.	1	2	3	4

13. I feel anxious about leaving my child with a baby sitter.	1	2	3	4
14. I am sometimes unsure about my ability to care for my child as well as I should.	1	2	3	4
15. I feel guilty when I have to punish my child.	1	2	3	4

MISHEL UNCERTAINTY IN ILLNESS SCALE – PARENT/CHILD FORM

Please read each statement. Take your time and think about what each statement says. Then circle the number under the column that most closely measures how you are feeling about your child TODAY. If you agree with a statement, then you would mark under either "Strongly Agree" or "Agree." If you disagree with a statement, then mark under either "Strongly Disagree" or "Disagree." If you are undecided about how you feel about your child, then mark under "Undecided" for that statement. Please respond to every statement.

	Strongly Agree	Agree	Undecided	Disagree	Strongly Disagree
1. I don't know what is wrong with my child.	5	4	3	2	1
2. I have a lot of questions without answers.	5	4	3	2	1
3. I am unsure if my child's illness is getting better or worse.	5	4	3	2	1
4. It is unclear how bad my child's pain will be.	5	4	3	2	1
5. The explanations they give about my child seem hazy to me.	5	4	3	2	1
6. The purpose of each treatment for my child is clear to me.	5	4	3	2	1
7. I do not know when to expect things will be done to my child.	5	4	3	2	1
8. My child's symptoms continue to change unpredictably.	5	4	3	2	1
9. I understand everything explained to me.	5	4	3	2	1

10. The doctors say things to me that could have many meanings.	5	4	3	2	1
11. I can predict how long my child's illness will last.	5	4	3	2	1
12. My child's treatment is too complex to figure out.	5	4	3	2	1
13. It is difficult to know if the treatments or medications my child is getting are helping.	5	4	3	2	1
14. There are so many different types of staff, it's unclear who is responsible for what.	5	4	3	2	1
15. Because of the unpredictability of my child's illness, I cannot plan for the future.	5	4	3	2	1
16. The course of my child's illness keeps changing. He/she has good and bad days.	5	4	3	2	1
17. It's vague to me how I will manage the care of my child after he/she leaves the hospital.	5	4	3	2	1
18. It is not clear what is going to happen to my child.	5	4	3	2	1
19. I usually know if my child is going to have a good or bad day.	5	4	3	2	1
20. The results of my child's tests are inconsistent.	5	4	3	2	1
21. The effectiveness of the treatment is undetermined.	5	4	3	2	1
22. It is difficult to determine how long it will be before I can care for my child by myself.	5	4	3	2	1
23. I can generally predict the course of my child's illness.	5	4	3	2	1
24. Because of the treatment, what my child can do and cannot do keeps changing.	5	4	3	2	1

25. I'm certain they will not find anything else wrong with my child.	5	4	3	2	1
26. They have not given my child a specific diagnosis.	5	4	3	2	1
27. My child's physical distress is predictable; I know when it is going to get better or worse.	5	4	3	2	1
28. My child's diagnosis is definite and will not change.	5	4	3	2	1
29. I can depend on the nurses to be there when I need them.	5	4	3	2	1
30. The seriousness of my child's illness has been determined.	5	4	3	2	1
31. The doctors and nurses use everyday language so I can understand what they are saying.					

PPS

	Never	Sometimes	Most of the Time	Always
1. I blame myself when my child gets hurt.	0	1	2	3
2. I comfort my child immediately when he/she cries.	0	1	2	3
3. I encourage my child to depend on me.	0	1	2	3
4. I have difficulty separating from my child.	0	1	2	3
5. I trust my child on his/her own.	0	1	2	3
6. I let me child make his/her own decisions.	0	1	2	3
7. I have difficulty leaving my child with a babysitter.	0	1	2	3
8. I decide when my child eats.	0	1	2	3
9. I use baby words when I talk to my child.	0	1	2	3
10. I urge my child to try new things.	0	1	2	3
11. I determine who my child will play with.	0	1	2	3
12. I keep a close watch on my child.	0	1	2	3
13. I feed my child even if he/she can do it alone.	0	1	2	3
14. I feel comfortable leaving my child with other people.	0	1	2	3
15. I protect my child from criticism.	0	1	2	3

16. I let my child choose what he/she wears.	0	1	2	3
17. I make my child go to sleep at a set time.	0	1	2	3
18. I go to my child if he/she cries during the night.	0	1	2	3
19. I encourage my child to play with other children.	0	1	2	3
20. I give my child extra attention when he/she clings to me.	0	1	2	3
21. I decide what my child eats.	0	1	2	3
22. I dress my child even if he/she can do it alone.	0	1	2	3
23. I decide when my child goes to the bathroom.	0	1	2	3
24. I know exactly what my child is doing.	0	1	2	3
25. I allow my child to do things on his/her own.	0	1	2	3

Life Orientation Test-Revised

Please be as honest and accurate as you can throughout. Try not to let your response to one statement influence your responses to other statements. There are no "correct" or "incorrect" answers. Answer according to your own feelings, rather than how you think "most people" would answer.

	1 I DISagree a lot	2 I DISagree a little	3 I neither agree nor disagree	4 I agree a little	5 I agree a lot
1. In uncertain times, I usually expect the best.	1	2	3	4	5
2. It's easy for me to relax.	1	2	3	4	5
3. If something can go wrong for me, it will.	1	2	3	4	5
4. I'm always optimistic about my future.	1	2	3	4	5
5. I enjoy my friends a lot.	1	2	3	4	5
6. It's important for me to keep busy.	1	2	3	4	5
7. I hardly ever expect things to go my way.	1	2	3	4	5
8. I don't get upset too easily.	1	2	3	4	5
9. I rarely count on good things happening to me.	1	2	3	4	5
10. Overall, I expect more good things to happen to me than bad.	1	2	3	4	5

Questions About the Future—P

Please rate the following statements about your child's future on a scale from 1 to 4, 1 being very unlikely and 4 being very likely.

Very Unlikely   Unlikely   Likely   Very Likely
1----------------------------2----------------------3----------------------------4

_____ My child will have a full time job someday.

_____ If my child works, s/he will travel to work by himself/herself.

_____ My child will be able to live on his/her own someday.

_____ My child will get married someday.

_____ My child will have children someday.

_____ If my child has children, s/he will be able to care for them.

_____ My child will drive a car someday.

_____ My child will go to college.

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VITA

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