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A Concurrent and Longitudinal Examination of a Bio-Neuropsychosocial Model for Predicting Medical Adherence and Responsibility in Youth with Spina Bifida

Alexandra Psihogios
Loyola University Chicago

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LOYOLA UNIVERSITY CHICAGO

A CONCURRENT AND LONGITUDINAL EXAMINATION OF A BIO-
NEUROPSYCHOSOCIAL MODEL FOR PREDICTING MEDICAL ADHERENCE
AND RESPONSIBILITY IN YOUTH WITH SPINA BIFIDA

A DISSERTATION SUBMITTED TO
THE FACULTY OF THE GRADUATE SCHOOL
IN CANDIDACY FOR THE DEGREE OF
DOCTOR OF PHILOSOPHY

PROGRAM IN CLINICAL PSYCHOLOGY

BY

ALEXANDRA M PSIHOGIOS, M.A.

CHICAGO, ILLINOIS

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CHAPTER ONE

INTRODUCTION

Medical nonadherence, or discordance between an individual's behavior and medical recommendations, is a major public health concern that poses significant risks for patients, families, and the larger healthcare system (Modi, Pai, Hommel, et al., 2012). Nonadherence with medical regimens can adversely affect the physical and psychological health of patients, the cost-effectiveness of medical care and clinical decisions, and the results of clinical trials (Rapoff, 2011). In 2009, the New England Healthcare Institute (NEHI) estimated that \$290 billion in avoidable medical spending was generated annually by nonadherence. For youth with chronic health conditions, medical nonadherence is a primary cause of treatment failure and reduced quality of life (Sabaté, 2003, Fredericks, Magee, Oipari-Arrigan, et al., 2008). Considering the escalating prevalence rates of adult and child chronic health conditions (Van Cleave, Gortmaker, & Perrin, 2010), it is likely that nonadherence will continue to burden patients, patients' families, and the economy in the next decade.

Medical adherence is distinct from other related medical self-management constructs, including perceived barriers or facilitators to adherence, medication knowledge, medical self-efficacy, and attitude or beliefs about medications (Stirratt, Dunbar-Jacob, Crane, et al., 2015). The transfer of medical responsibilities from parent to child is another aspect of medical self-management and is defined as an interpersonal

family process in which an adolescent assumes greater responsibility with healthcare tasks (O'Hara & Holmbeck, 2013; Reed-Knight, Blount, & Gilleland, 2014). Aspects of medical self-management may be best conceptualized as antecedents or consequences of medical adherence (Stirratt et al., 2015). For example, medical nonadherence can be the result of transitioning responsibilities to children before they are developmentally capable of following through with medical recommendations.

Gaining insight into medical adherence and responsibility behaviors during the adolescent years is essential, as long-term health behaviors, including engagement in the health system, diet, exercise, and drug/alcohol use, are often established and consolidated during this developmental period (Williams, Holmbeck, & Greenley, 2002). During adolescence, many youth with chronic health conditions gain increased responsibility for their medical regimen (Anderson, Ho, Brackett, et al., 1997; Modi et al., 2008; Stepansky et al., 2010). Unfortunately, rates of adherence amongst adolescents are generally much lower than adherence rates in younger children and adults (i.e., 50% adherence rate among adolescents; La Greca & Mackey, 2009). This drop in adherence is thought to reflect a transitional period in which the adolescent assumes increased responsibility for his or her medical care (while parents become less involved; Miller & Harris, 2012), as well as other salient developmental issues of adolescence that may negatively impact medical self-management and adherence (e.g. individuation and separation from the family and greater affiliation with peers; Rapoff, 2011). Not surprisingly, research suggests that increased parental responsibility and supervision are associated with higher levels of adherence during the adolescent period (e.g., Anderson et al., 1997; Ellis, Podolski, Naar-King, et al., 2007; Helgeson, Reynolds, Siminerio, et al., 2008).

Current research on correlates of adherence in pediatric populations is fairly broad, ranging from patient characteristics (e.g., demographics, socioeconomic status, adjustment/coping, and neurocognitive functioning) to family (e.g., coping, parental involvement, and family functioning), peer (e.g., friendship quality and peer victimization), biological (e.g., severity) and regimen characteristics (e.g., complexity; Rapoff, 2011). Despite knowledge regarding how these correlates concurrently relate to adherence, research has yet to establish enduring and predictive variables that relate medical adherence across time (Simons, McCormick, Devine, & Blount, 2010; Helgeson et al., 2008; Holmbeck & Devine, 2010; La Greca & Mackey, 2009). By understanding why patients and their families do or do not follow medical or health recommendations over salient developmental periods, such as adolescence, meaningful interventions can target specific barriers to reduce long-term negative outcomes (e.g., disease-related morbidity, mortality, and unnecessary health costs). Further, analyses of these factors may enable clinicians and researchers to tailor interventions to specific at-risk groups and to unveil meaningful targets for intervention (such as parenting or the development of self-efficacy; Berg, King, Butler et al., 2011; Rapoff, 2011).

The Current Study

Youth with spina bifida (SB) are considered an understudied and underserved population who endure physical, neuropsychological, developmental, and psychosocial challenges during the adolescent years (Holmbeck & Devine, 2010). While preliminary investigations suggest that up to 50% of children and adolescents with nonadherence (Psihogios, Kolbuck, & Holmbeck, 2015), existing research has not adequately described the individual and contextual factors that promote or impede adherence to SB treatments

(Sawin, Betz, & Linroth, 2010). To address this gap in the research literature, the current study tested the utility of a bio-neuropsychosocial model of adjustment for pediatric spina bifida (SB; Holmbeck & Devine, 2010) through the evaluation of salient biological, neuropsychological, and social (i.e., family and social adjustment) predictors of concurrent and longitudinal medical adherence and responsibility in a pediatric SB population.

To investigate adolescent medical adherence and responsibility simultaneously as outcomes, we created a categorical variable with four levels, from two questionnaires (a medical adherence questionnaire and a medical responsibility questionnaire; see Figure 1) in two core medical domains essential to SB medical care: clean intermittent catheterization and completing a bowel program. By creating four variables for two primary medical domains, this study evaluated rates of adherence/nonadherence (e.g., adherence for catheterization or bowel program) depending on the child's level of responsibility in that specific domain (e.g., responsibility with catheterization or bowel program). Furthermore, the creation of these variables allowed investigation of hypotheses regarding the most optimal level of functioning in terms of adherence and responsibility (i.e. child responsible and adherent), and less optimal medical outcomes (i.e., limited child responsibility and/or nonadherent). Salient predictors of less optimal outcomes may unveil targeted foci for adherence and medical self-management interventions.

Figure 1. Evaluating Adherence and Nonadherence for Catheterization/Bowel Program Based on Child Responsibility with Catheterization and Bowel Program at Time 1 and Time 2

		<u>Responsible</u>	
		Yes	No
<u>Adherent</u>	Yes	Group 1: Adherent, Child Responsible (with catheterization or bowel program)	Group 2: Adherent, Child Not Responsible (with catheterization or bowel program)
	No	Group 3: Nonadherent, Child Responsible (with catheterization or bowel program)	Group 4: Nonadherent, Child Not Responsible (with catheterization or bowel program)

Adolescent medical adherence and self-management of SB has been identified as a “gap” in the pediatric psychology evidence-based intervention literature (Sawin, Betz, & Linroth, 2010), with few studies describing the factors that promote or impede SB health behaviors in this age group. Individuals with SB may struggle with acquiring appropriate medical self-management for several reasons. First, youth with SB typically demonstrate Low Average to Average cognitive capabilities (Riddle et al., 2005; Wills, 1993) and struggle with aspects of executive functioning (Dennis, Barnes, & Heatherington, 1999; Fletcher, Brookshire, Landry, et al., 1996). This pattern of cognitive

challenges may make it more difficult for youth with SB to plan and follow through with multi-step medical procedures (such as catheterization). For example, youth may struggle to remember to catheterize every 3-4 hours or neglect an important step of the catheterization process (e.g., washing hands). Second, it has also been observed that children with SB tend to achieve lower overall levels of autonomy during adolescence compared to typically developing children (Davis, Shurtleff, Walker & Seidel, 2006; Friedman et al., 2009). Given difficulties youth with SB have in achieving independence in general independence domains (e.g., doing chores around the house or cooking meals), it is likely that they will also struggle with gaining the skills and practice necessary to become independent and adherent with a complex medical regimen. Understanding the unique medical self-management demands for children and adolescents with SB, including how these youth negotiate these demands within the context of families, will better guide the selection and adaption of existing evidence-based self-management interventions (e.g., Kahana, Drotar, & Frazier, 2008) that match targets for youth with SB.

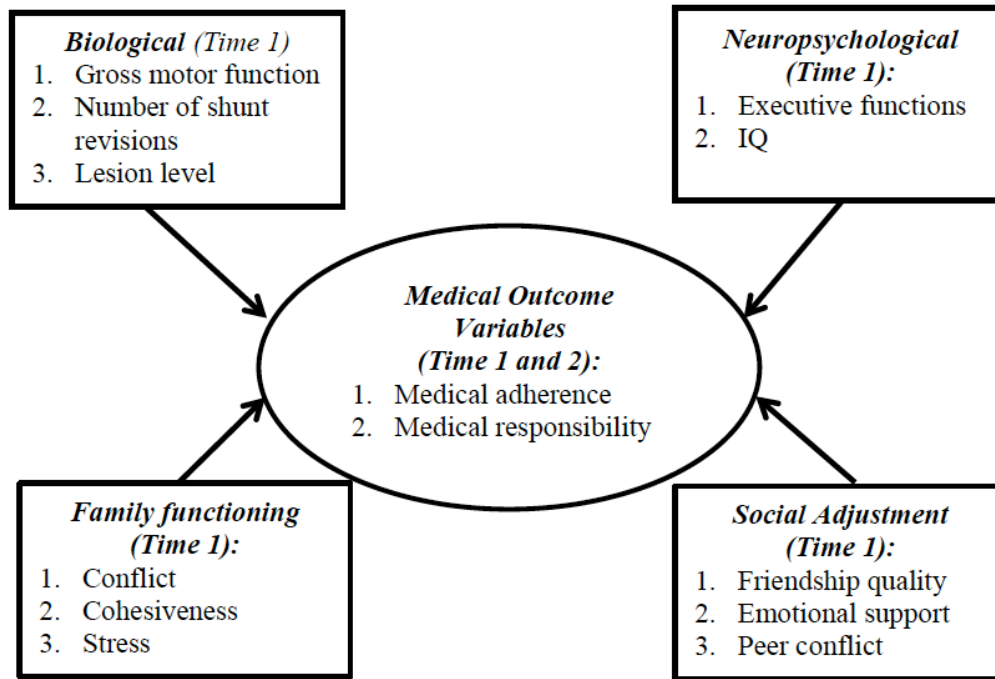
Evidenced-based models of pediatric medical management underscore the influence of modifiable family factors on medical adherence (Grey, Schulman-Green, Knafl, & Reynolds, 2015; Modi et al., 2012; Schwartz, Tuchman, Hobbie, & Ginsberg, 2011). These models are supported by a large body of literature that has linked aspects of family functioning (i.e. family conflict and cohesion; Stepansky, Roache, Holmbeck, & Schultz, 2010) to medical adherence in SB and other childhood chronic health conditions (e.g., Hanson, Henggeler, & Burghen, 1987; Lewin et al., 2006; Rapoff, 2011). In SB, research suggests that families with high levels of family conflict have lower levels of

adherence during early to late childhood (Psihogios & Holmbeck, 2013; Stepansky et al., 2010) and high levels of family cohesion are related to more optimal adherence (Stepansky et al., 2010). Adaptive parenting styles (e.g., high levels of acceptance and behavioral control) and higher levels of executive functioning among youth have also been linked to higher levels of concurrent levels of adherence (e.g., O'Hara & Holmbeck, 2013).

Research on youth with SB, however, has yet to evaluate the development of medical adherence and responsibility during adolescence in relation to other salient predictors, such as condition severity or social functioning (the latter being associated with adherence outcomes in other pediatric populations; e.g., La Greca et al., 1995; Bearman & La Greca, 2002). Additionally, the existing body of literature is limited by a lack of an organizational framework to study the impact of several different areas of functioning on adolescent medical adherence and responsibility in SB. Holmbeck and Devine (2010) proposed a bio-neuropsychosocial model of adjustment for individuals with SB, where adjustment in medical and non-medical domains is influenced by biological (e.g., SB severity), neuropsychological (e.g., executive functioning), and social (e.g., family and peer functioning) variables. These authors recommended that studies examine biological, neuropsychological, and social predictors of adjustment variables (including medical adherence) across time, particularly during critical stages of development (such as the transition to adolescence). The current study utilized this framework to study medical adherence and responsibility in a pediatric SB population, over the course of two biennial study time points.

This study extended the current literature by utilizing a bio-neuropsychosocial model of adjustment (Holmbeck & Devine, 2010) to evaluate the predictive value of four different domains of functioning on the development of medical responsibility and adherence in youth with SB: 1) biological (gross motor functioning, number of shunt revisions, and lesion level), 2) neuropsychological (IQ and assessment and questionnaire-based measures of executive functioning), 3) family (multi-method and multi-source measures of conflict, cohesion, and stress), and 4) peer (friendship quality, emotional support from peers, and peer conflict). We expected that each of these domains would relate to adherence and responsibility in unique ways (see Figure 2) and together, these domains would account for significant variability in concurrent and longitudinal health care behaviors. Although these domains are unlikely to be independent from one other (e.g., neuropsychological functioning and SB severity may be intertwined), further understanding the domains that most closely relate to the development of SB adherence and responsibility may reveal important intervention targets (e.g., family or peer dynamics) or at-risk groups (e.g., children with particular disease or neuropsychological characteristics). Thus, examining these predictor groups separately is clinically useful to healthcare teams working with affected youth in multi-disciplinary medical settings.

Figure 2. The Bio-Neuropsychosocial Model for Predicting Spina Bifida Medical Adherence and Responsibility at Time 1 and Time 2.



Hypotheses

In terms of the biological domain, we expected that children with more severe SB (i.e., youth with upper-level lesions, more shunt revisions, and decreased gross motor functioning) would be more likely to fall in the “Adherent, Child Not Responsible” or “Not Adherent, Child Not Responsible) categories (i.e. Group 2 and 4 respectively, see Figure 1), compared to the reference group, “Adherent, Child Responsible” (i.e., Group 1) at Time 1 and Time 2. That is, parents of children with more severe SB would be more likely to maintain responsibility for SB medical management. For a subset of families (i.e., families in Group 4), we hypothesized that parents would struggle to adhere to treatments for more severe SB. We did not expect that SB severity would significantly differentiate Group 3 (“Not Adherent, Child Responsible”) from Group 1.

In neuropsychological domains, we expected that poorer executive functions (as measured by a questionnaire and test data) and lower IQ would relate to nonadherent categories (i.e., “Non-Adherent, Child Responsible” and “Non-Adherent, Child Not Responsible”, Groups 3 and 4 respectively, see Figure 1), compared to Group 1 at Time 1 and Time 2. Support for this hypothesis stems from research that indicates cognitive deficits (i.e., lower IQ, inattention, and executive dysfunction; Stepansky et al., 2010; O’Hara & Holmbeck, 2013) are related to suboptimal adherence. We also expected that neuropsychological functioning would differentiate participants in Group 2 (i.e., “Adherent, Child Not Responsible”) from Group 1. That is, for some families, parents who recognized child neuropsychological deficits would be more likely to maintain medical responsibility, as well as successfully follow the medical regimen.

Socially, we predicted that youth with more family conflict, less family cohesion, and more family stress would be more likely to fall in the “nonadherent” groups (i.e., “Non-Adherent, Child Responsible” and “Non-Adherent, Child Not Responsible”, Groups 3 and 4 respectively, see Figure 1) compared to Group 1 at Time 1 and Time 2. This hypothesis is guided by past research in a separate population of youth with SB that found that family functioning is an important predictor of medical adherence across time (Stepansky et al., 2010). We did not expect family variables to significantly differentiate Group 2 from Group 1, as we hypothesized high levels of family functioning in both of these groups.

We hypothesized that poorer friendship quality, lower peer emotional support, and higher levels of peer conflict would differentiate individuals in the “nonadherent” groups (i.e., “Non-Adherent, Child Responsible” and “Non-Adherent, Child Not

Responsible”, Groups 3 and 4 respectively, see Figure 1) from youth in Group 1 at Time 1 and Time 2. That is, youth who were responsible and adherent to their medical regimens would display higher levels of social adjustment than youth with poor adherence. In addition, we expected that levels of social adjustment would differentiate children in Group 2 (i.e., “Adherent, Child Not Responsible”) compared to Group 1. Although adherence is high in both groups, we expected that children who struggle socially would also demonstrate difficulties with medical autonomy development.

Finally, we sought to examine which domain (biological, neuropsychological, family, or peer) accounted for the most variance in SB medical adherence and responsibility for catheterization and bowel programs two years later. As past research consistently cites a link between family functioning and adherence in SB populations (e.g., Stepansky et al., 2010; Psihogios & Holmbeck, 2013), we hypothesized that the family domain would account for the most variance in SB responsibility and adherence for catheterization and bowel programs. We did not expect different findings for catheterization and bowel management, or different findings for concurrent and longitudinal analyses.

CHAPTER TWO

REVIEW OF THE RELEVANT LITERATURE

Overview of Spina Bifida

Despite the relatively large body of literature devoted to studying adherence in youth with conditions such as diabetes (e.g., Berg et al., 2011; Helgeson et al., 2008), the literature on adherence in youth with SB is extremely limited (e.g., Holmbeck et al., 1998; Stepansky et al., 2010). A PubMed literature search utilizing the term “self-management” revealed 18 citations for SB versus 3,008 for diabetes and a search of “adherence” yielded 16 citations for SB versus 5,561 for diabetes. Self-management of pediatric SB requires a complex medical regimen, which is a risk factor for poor adherence (Quittner et al., 2000). Indeed, preliminary research of our sample showed that adolescents and parents struggled with adhering to a variety of SB medical tasks, including the completion of bowel programs, following exercise recommendations, and conducting routine skin checks (Psihogios, Kolbuck, & Holmbeck, 2015). Without intervention, ongoing adherence issues in these domains may result in lasting and fatal health crises. Preventable, life-threatening secondary complications, such as pressure ulcers and urinary tract infections are the most common cause of hospitalizations in individuals with SB (Dicianno & Wilson, 2010; Kinsman & Doehring, 1996), with sepsis and acute renal failure being a common cause of unexpected death (Oakeshott, Hunt, Poulton, & Reid, 2010; Roach, Short, & Saltzman, 2011).

SB is one of the most common and disabling birth defects in the United States, occurring in roughly 3 out of every 10,000 live births (CDC, 2011). Each day, approximately eight infants are born with SB in the United States (SB Association, 2008), and these infants face a multitude of physical, cognitive, psychosocial, and medical challenges throughout their lives. SB is caused by the incomplete closure of the neural tube during the early stages of pregnancy, resulting in malformations of the spinal cord and cerebral cortex (i.e., the Arnold-Chiari malformation). The most common and severe form of SB is myelomeningocele, in which the meninges, spinal cord, and neural elements protrude out from the spinal cord through open vertebrae, resulting in significant nerve damage (Deidrick, Grissom, & Farmer, 2009). A more moderate form of SB, meningocele (which involves no neural elements), is characterized by meninges protruding through the opening of the spine but less severe nerve damage than myelomeningocele. A third type, lipomeningocele, is considered the most mild form of SB. Lipomeningocele is characterized by a benign tumor that consists of fatty tissue over part of the spine and is associated with minimal nerve damage (Menkes & Till, 1995).

In addition to variability in the type of SB, SB severity may vary based on the location of the lesion on the spine (with higher level lesions indicating more severe SB) and the presence or absence of shunted hydrocephalus. Lesions in the sacral region are most common in SB, although lesions may occur at any level of the spine (Wills, 1993). Higher spinal lesions are associated with greater paralysis and limitations in upper- and lower-limb movement quality (e.g., Dennis, Fletcher, Rogers, Hetherington, & Francis, 2002; Landry, Lomax-Bream, & Barnes, 2003), whereas individuals with lower level regions generally have better mobility and muscle strength. Lesions in the thoracic region

result in impairment in trunk, partial or complete impairment in lower extremities, and impairment in bladder and bowel functioning. Lesions in the lumbar region are associated with complete trunk function, but weaknesses in hips, knees, ankles, and feet. Sacral level lesions are associated with weakened hips and feet and bowel/bladder dysfunction.

The severity of ambulatory/functional mobility depends on the location of the spinal lesion, as motor and sensory functioning is typically impaired at and below this spinal insult. Mobility goals vary according to the degree of functional impairment, with individuals with lower-level lesions needing less assistance than individuals with upper-level lesion. Depending on the degree of difficulty with ambulation, individuals with SB often utilize assistive devices including orthotics, braces, and wheelchairs (Children's National Medical Center, 1995). For example, patients with lesions in the thoracic region typically require the use of a wheelchair in the community, as well as a walker/crutches for limited distances, a bath bench, and driving with hand controls. Individuals with lumbar lesions typically require a wheelchair in the community or walker and crutches. Finally, individuals with sacral lesions may utilize ankle-foot orthotics (AFO's) or supra malleolar orthotics (SMO's), shoe inserts, and crutches, or may not require assistive devices at all.

Approximately 80% of patients with SB have hydrocephalus, or abnormal build-up of cerebrospinal fluid around the brain (Spina Bifida Association, 2008). The treatment of hydrocephalus involves diversionary shunting of the cerebrospinal fluid (CSF). Although shunting can improve the long-term functional outcomes of individuals with hydrocephalus and restore brain volume, most researchers have found that reversing neuronal and axonal damage is unlikely (Del Bigio, 1993). Further difficulty managing

CSF also tends to occur due to shunt malfunctions and infections, which are associated with worse neuropsychological outcomes (Dennis et al., 2006; Hetherington, Dennis, Barnes, Drake, & Gentili, 2006).

Children with SB require intense medical care throughout their lifetime and this treatment typically begins in-utero or after birth when the lesion in the spine is surgically repaired. Subsequent medical procedures typically include a surgical placement of a shunt to divert cerebrospinal fluid from the ventricles of the brain to the abdominal cavity and repeated shunt surgeries to repair malfunctions. SB affects communication between the nerves in the spinal cord that control bladder and bowel functioning (i.e., neurogenic bladder and bowel), causing issues such as urinary and bowel incontinence, constipation, increased urinary urgency, reduced bladder capacity, and urinary tract infections (Mayo Clinic, 2014). Individuals with SB also experience physical disabilities and cognitive deficits throughout their lifetime. These medical impairments require complex care, such as clean intermittent catheterization, medications, bowel programs, physical therapy, dietary restrictions, nutritional supplements, and routine skin checks to prevent pressure ulcers. Additional services, such as physical therapy, occupational therapy, school accommodations, neuropsychological testing, and psychological/psychiatric services, may also be necessary.

Inadequate SB self-management and nonadherence is associated with numerous, preventable, secondary complications. For example, the most common presenting problem of emergency department visits in this population is urinary tract infections, which can occur because of poor adherence to catheterization (e.g., long delays between catheterizations and poor hygiene; Caterino et al., 2006). Similarly, fecal incontinence

and constipation may result from nonadherence to bowel programs (e.g., inconsistent program schedule, inadequate intake of dietary fiber, and inconsistent use of laxatives or stool softeners (Dicianno et al., 2008). Pressure ulcers are another common secondary complication of SB, with up to 62% of adults reporting a history of skin difficulties (Long & Green, 2009). Individuals with SB prevent pressure ulcers by engaging in routine skin checks and pressure relief exercises. Approximately one-third of patients with SB are obese (Buffart et al., 2008; Dosa et al., 2009), which is associated with additional health complications (e.g., hypertension, type 2 diabetes, sleep apnea, and cardiovascular disease). In order to prevent obesity and related health difficulties, SB healthcare providers typically recommend healthy meal plans and regular physical activity. Youth with SB also have higher rates of latex allergies, necessitating avoidance of products made from this material (Ausili et al., 2007). Less modifiable secondary complications include scoliosis, tethered cord syndrome, seizure disorders, and shunt malfunctions and infections (Dicianno et al., 2008; Webb, 2010).

The Development of Medical Responsibility and Adherence

For many youth with chronic health conditions, adolescence is characterized by increased responsibility with medical care. For instance, it has been found that parents of children with diabetes (Anderson et al., 1997), cystic fibrosis (Modi et al., 2008), and SB (Stepansky et al., 2010) transfer medical responsibilities to children during early adolescence. By the time children with SB are 12-13 years old, most children have obtained at least partial responsibility for catheterization and bowel programs. Despite these gains in responsibility, rates of adherence amongst adolescents are generally lower than adherence rates in younger children and adults (i.e., 50% adherence rate among

adolescents; La Greca & Mackey, 2009). In SB, rates of nonadherence for particular medical tasks are also comparable to 50%, with one study reporting that 24.1 to 50.0% of 12-13 year olds were nonadherent to skin checks (Psihogios, Kolbuck, & Holmbeck, 2015).

According to Ricker, Delamater, and Hsu (1998), several concerns regarding medical self-management emerge during adolescence and these issues may negatively impact adherence behaviors. First, the developmental strivings of adolescence, such as independence and individuation from the family, may result in less optimal adherence to a demanding daily regimen. Other developmental events, such as socializing with peers or working at one's first job, may also interfere with the scheduling and completion of medical management tasks. Furthermore, the permanent and severe nature of the disease may become more evident and discouraging during adolescence. An adolescent may realize that his or her illness will persist, even if he or she is fully adherent, and this thought process may negatively affect the adolescent's adherence and psychological adjustment. Thus, the changes that an adolescent is experiencing (in terms of increased responsibility for disease management and developmental events) may make it difficult for an adolescent to be successful at caring for their health without additional support.

A successful transfer of medical responsibilities would require that youth have: (1) the requisite skills to initiate and carryout the medical tasks (e.g., catheterization and bowel management), (2) adequate levels of self-efficacy to do so, and (3) caregivers who carefully monitor their readiness to initiate self-management behaviors, while granting them increasing amounts of responsibility as they become more self-reliant and competent (Reed-Knight et al, 2014). This transfer process is facilitated and high levels

of adherence are more likely to be maintained in families where parent-child conflict levels are low and when parents demonstrate flexibility and a willingness to share self-management responsibilities with their children (La Greca & Mackey, 2009; Modi et al., 2008; Reed-Knight et al, 2014).

The existing body of literature on pediatric adherence typically focuses on adherence in general, without attention to who in the family is completing the medical tasks. For example, it is possible that adherence to SB treatments is high, but parents have yet to transfer medical responsibilities to their adolescent despite the adolescent being well into their teens (i.e., growth in medical responsibility is compromised). For other families, the adolescent with SB may be responsible with his or her care, but have low levels of adherence. Indeed, adherence rates are generally lower when adolescents with SB were primarily responsible for their care (rather than parents; Psihogios & Holmbeck, 2013; Psihogios, Kolbuck, & Holmbeck, 2015).

Biological Factors

A relevant domain that may affect the development of medical responsibility and adherence over time is SB biological variables, specifically SB severity. SB severity has been measured in a variety of ways, including lesion level (e.g., thoracic, lumbar, or sacral), ambulation status (crutches, wheelchair and/or braces), gross motor functioning, the presence or absence of hydrocephalus, and the number of shunt reparations (as repeated shunt surgeries typically results in secondary central nervous system insults, e.g., Hommeyer, Holmbeck, & Wills, 1999). Based on a theoretical model of maladjustment in children with chronic health conditions, Wallander and Varni (1989) suggested that disease severity might have an important impact on a child's overall

psychosocial functioning. This relationship may be direct (e.g., increased pain or difficulty may have a direct effect on adjustment) or indirect based on the condition's impact on functional status (e.g., the ability to perform tasks in an age-appropriate manner).

Research on the link between biological factors and medical adherence in pediatric SB is lacking. As there is no research in this area, we speculate that parents of children with more severe SB may maintain responsibility for SB treatment tasks throughout adolescence, which may protect against the normative drop in adherence during this development period. On the other hand, treatment complexity is considered an important risk factor for poor adherence (Quittner et al., 2000), and it is likely that children with more severe SB must follow more complex treatments (e.g., more intensive physical therapy). Thus, even when parents maintain responsibility for SB medical management, parents may struggle to adhere to more complicated medical recommendations.

Neuropsychological Functioning

Children with SB, especially those with myelomeningocele, are at increased risk for neuropsychological difficulties. Neuropsychological functioning is affected by initial and recurrent hydrocephalus (Mataró, Junqué, Poca, & Sahuquillo, 2001), lesion level (Fletcher et al., 2005), the Arnold-Chiari malformation, and repeated shunt surgeries (Tarazi, Zabel, & Mahone, 2008). On neuropsychological tests, children with SB and hydrocephalus often demonstrate low average to average cognitive capabilities, with relatively better performance on verbal than nonverbal tasks (Riddle et al., 2005; Wills,

1993). On the other hand, children with SB, but without hydrocephalus, often do not exhibit as severe of neural morbidity, compared to children with SB and hydrocephalus.

In addition to cognitive deficits, it has been observed that youth with SB struggle with executive functions (EF; Dennis, Barnes, & Heatherington, 1999; Fletcher et al., 1996). Executive functions are thought to include processes such planning, working memory, attention, problem solving, verbal reasoning, inhibition, mental flexibility, multi-tasking, initiation and monitoring of action (Chan et al., 2008). Research examining executive functioning in individuals with SB has found impairments in shifting (Iddon et al., 2004), working memory (Burmeister et al., 2005), planning, organization, goal-directed behavior, and problem solving (e.g., Fletcher et al., 1996). These deficits are hypothesized to have significant effects on medical adherence and autonomy.

The neuropsychological impairments associated with SB can negatively impact a child's ability to develop autonomy, adaptive functioning, and achievement (Tarazi, Zabel, & Mahone, 2008). Given the complexity of SB care, executive functioning encompasses many important skills used in the management of complex SB regimens. For example, children with poor planning abilities may forget to catheterize in 3-4 hour intervals or forget important steps involved in preventing infection (i.e., washing hands). SB youth with lower than average verbal intelligence (measured by the Peabody Picture Vocabulary Test) exhibited slower growth rate in emotional independence and intrinsic motivation across the transition into adolescence, when compared to a typically developing control group (Friedman, Holmbeck, DeLucia, Jandasek & Zebracki, 2009). Another study found that executive functioning problems predicted higher levels of

observed child dependency and lower levels of intrinsic motivation in youth with SB (Tuminello, Holmbeck, & Olsen, 2012).

Regarding medical adherence and responsibility youth with SB, O'Hara and Holmbeck (2013) found that lower executive functions (measured by questionnaires) were associated with poorer medical adherence, and lower executive functions (measured by test data) were associated with lower levels of medical autonomy. Although this study offered preliminary support between neuropsychological functioning and medical adherence and responsibility in youth with SB, this study focused on early adolescence only. To date, no studies have evaluated the link between neuropsychological functioning and medical adherence and self-management during adolescence.

Family Dynamics

The transfer of medical responsibilities from parent to adolescent is a challenging time period for the family unit. For instance, parents must balance two conflicting demands: encouraging their adolescent's autonomy while continuing to monitor the adolescent's adherence and health (Anderson & Coyne, 1991; Holmbeck et al., 2002). Parental constraints on and excessive granting of adolescent medical responsibilities have been associated with poor adherence (Olsen, Berg & Wiebe, 2008), whereas ongoing and positive (rather than intrusive) parental involvement and communication have been associated with more favorable disease-related outcomes (Anderson et al., 1997; Ellis et al., 2007; Helgeson, Reynolds, Siminerio, Escobar, & Becker, 2008; Wiebe et al., 2005; Wysocki et al., 2006). The relationship between family functioning and treatment adherence has been extensively studied in other childhood chronic illnesses, particularly diabetes. A number of studies of family functioning found that relationship factors (e.g.,

communication, problem-solving skills, conflict resolution) significantly predicted adherence behaviors for children and adolescents with diabetes (Bobrow, AvRuskin, & Siller, 1985; Wysocki et al., 1999).

Considering the neuropsychological profile (e.g., poor executive functions) and physical disabilities of those with SB, positive family interactions during early adolescence may be especially important for this population. For instance, ongoing parental support and teaching effective medical management may alleviate some of the difficulties that an adolescent with SB may face if prematurely granted full responsibility for medical care (e.g., trouble planning/organization or accessibility issues). Indeed, research suggests that parental involvement in SB medical care is essential for optimal medical adherence during pre- and early adolescence (Psihogios & Holmbeck, 2013). Furthermore, more adaptive parenting characteristics (i.e. maternal acceptance and behavioral control; O'Hara & Holmbeck, 2013) have been associated with higher levels of adherence (but not medical autonomy) during early adolescence. Taken together, the quantity of parental involvement and the quality of parenting behaviors are important predictors of in youth with SB. These findings provide further evidence that the development of medical adherence and responsibility during adolescence is a dynamic process that is affected by parental involvement.

As children with SB tend to lag behind their typically development peers in terms of autonomy development by approximately two years (Devine et al., 2011), it has been suggested that this high level of dependence on parents (in combination with limited social interactions) makes it highly likely that the family will play a significant role in the management of the child's SB across the span of adolescence (Stepansky et al., 2010).

Previous research suggests that family functioning is a salient predictor of adherence rates across a wide range of pediatric populations (Hanson, Henggeler, & Burghen, 1987; Lewin et al., 2006; Rapoff, 2011), including in families of youth with SB (Stepansky et al., 2010). It has been suggested that the daily management of medical tasks may be more difficult in the presence of conflictive family relationships, since effective communication, supervision, and division of responsibilities may be compromised (Schöbinger et al., 1993; Klemp & La Greca, 1987). This finding has been supported in a SB population, with studies finding that high levels of family conflict predict a decrease in concurrent (Psihogios & Holmbeck, 2013) and later adherence (Stepansky et al., 2010). According to Stepansky and colleagues (2010), the longitudinal association between family conflict and medical adherence suggests that family conflict and medical adherence become increasingly intertwined during adolescence.

On the positive side, studies on family cohesiveness (including warmth, acceptance, emotional health, and closeness; see DiMatteo, 2004 for a meta-analysis, Klemp & La Greca, 1987), support, expressiveness, organization, and expressiveness in pediatric populations have linked these variables to higher adherence to regimens (Rapoff, 2011). This finding has also been supported in the SB literature, with family cohesion being positively associated with medical adherence across adolescence (Stepansky et al., 2010). In general, this literature suggests that children with more structured, cohesive, and supportive family environments are in better control of their disease. Possibly, these families are better able to navigate developmental issues that may compromise adherence (e.g., through effective communication and support).

Although it appears to be clear that family functioning has an effect on the management of pediatric conditions (including SB), the existing body of literature has yet to evaluate whether family functioning relates to an adolescent's ability to develop medical responsibility and demonstrate adherence in the face of this increased responsibility. In past research studies, adherence has been evaluated broadly, without consideration to who is completing the task (e.g., a family may have high levels of adherence, but the child is completely dependent on his or her parents). As at the end of adolescence, the most optimal outcome for a teen is to be independent and adherent to their disease care, this study extended the literature by examining different levels of medical responsibility and adherence simultaneously (see Figure 1).

Social Adjustment

During adolescence, peer relationships become an influential force that may positively or negatively impact a child's psychological and academic functioning (Hartup, 1996). For youth with chronic health conditions, peer relationships can buffer against the stress of having a chronic health condition and affect adherence behaviors (Bearman & La Greca, 2002). Specifically, it has been suggested the high quality friendships have a positive effect on the adolescent's medical behaviors by reducing stigma about the illness, improving self-esteem and self-efficacy, and by providing additional resources for coping with their care (La Greca, 2002).

Similar to the literature on adherence and family functioning, most research investigating adherence behaviors and peer correlates has been conducted in diabetes populations. In general, research on the link between social functioning variables and adherence behaviors in diabetes populations have been mixed, with some studies finding

positive relation between health care behaviors and peer support, and other studies finding no relation between these two variables (see Palladino & Helgeson, 2012, for a review). In contrast, in the same review, a more robust relation has emerged between high levels of peer conflict and poorer diabetes outcomes, which might suggest that peer conflict has a larger effect on adherence outcomes than social support. The relation between medical adherence and less optimal peer support has been further studied in pediatric inflammatory bowel disease, which has found a positive relation between peer victimization and medical adherence (Janicke et al., 2009). In the same study, it was found that prosocial support moderated the relation between peer victimization and medical adherence, suggesting that emotional support may lessen the impact of harmful peer interactions.

Although youth with SB have shown lower levels of social functioning compared to other pediatric populations (Pinquart & Teubert, 2012), the impact of peer support on medical responsibility and adherence has yet to be explored. Considering that research on the relation between peer support and diabetes has been relatively inconclusive (Palladino & Helgeson, 2012), it is possible that research on these relations in a SB population may have important clinical implications (as these children have more profound impairments in social functioning). Possibly, the relation between friendship quality (e.g., level of companionship, security, closeness, and support) and medical outcomes may be especially salient for youth with SB, who have demonstrated lower levels of social adjustment in terms of the quality of their best friendships (e.g., Devine et al., 2012). However, this hypothesis has yet to be tested in this population.

Hypotheses

Taken together, biological traits, neuropsychological abilities, and family and peer relationships are likely to play a role in the development of medical adherence and responsibility over the course of adolescence. However, these variables have largely been investigated in isolation without a theoretical basis. Furthermore, much of the existing body of literature investigated these variables in relation to diabetes outcomes rather than in populations of youth with physical disabilities such as SB. This study represents the first to evaluate biological, neuropsychological, and social variables in relation to medical adherence and responsibility in youth with SB. Several hypotheses were tested to understand these relationships:

Hypothesis 1

Regarding biological characteristics, we expected that children with more severe SB (as indicated by higher lesion levels, more shunt revisions, and decreased gross motor functioning) would be more likely to fall in the “Adherent, Child Not Responsible” category (i.e. Group 2, see Figure 1), compared to the “Adherent, Child Responsible” category (i.e., Group 1) at Time 1 and Time 2. In other words, parents of children with more severe SB would be more likely to maintain responsibility for SB medical management, as well as successfully adhere to the SB regimen. We also expected that SB severity would differentiate Group 1 from Group 4 (“Non-Adherent, Child Not Responsible”). That is, for a subset of families, we hypothesized that families would struggle to adhere to treatments for more severe SB. We did not expect that SB severity would significantly differentiate Group 3 (“Not Adherent, Child Responsible”) from Group 1.

Hypothesis 2

In neuropsychological domains, we expected that poorer executive functions (as measured by a questionnaire and test data) and lower IQ would distinguish individuals in the nonadherent categories (i.e., “Non-Adherent, Child Responsible” and “Non-Adherent, Child Not Responsible”, Groups 3 and 4 respectively, see Figure 1) from youth in Group 1 at Time 1 and Time 2. This hypothesis is supported by research that suggest that cognitive deficits (i.e., lower IQ, inattention, and executive dysfunction; Stepansky et al., 2010; O’Hara & Holmbeck, 2013) are related to suboptimal adherence. We also expected that neuropsychological functioning would differentiate participants in Group 2 (i.e., “Adherent, Child Not Responsible) from Group 1. That is, for some families, parents who recognized child neuropsychological deficits would be more likely to maintain medical responsibility, as well as successfully follow the medical regimen.

Hypothesis 3

Socially, we predicted that youth with more family conflict, less family cohesion, and more family stress would be more likely to fall in the “nonadherent” groups (i.e., “Non-Adherent, Child Responsible” and “Non-Adherent, Child Not Responsible”, Groups 3 and 4 respectively, see Figure 1) compared to Group 1 at Time 1 and Time 2. This hypothesis is guided by past research in a separate population of youth with SB that found that family functioning is an important predictor of medical adherence across time (Stepansky et al., 2010). We did not expect family variables to significantly differentiate Group 2 from Group 1, as we hypothesized high levels of family functioning in both of these groups.

Hypothesis 4

We hypothesized that poorer friendship quality, lower peer emotional support, and higher levels of peer conflict would differentiate individuals in the “nonadherent” groups (i.e., “Non-Adherent, Child Responsible” and “Non-Adherent, Child Not Responsible”, Groups 3 and 4 respectively, see Figure 1) from youth in Group 1 at Time 1 and Time 2.. That is, youth who were responsible and adherent to their medical regimens would display higher levels of social adjustment than youth with poor adherence. In addition, we expected that levels of social adjustment would differentiate children in Group 2 (i.e., “Adherent, Child Not Responsible) compared to Group 1. Although adherence is high in both groups, we expected that children who struggle socially would also demonstrate difficulties with medical responsibility obtainment.

Hypothesis 5

As a group, the four domains (family, peer, neuropsychological, and biological variables) would account for significant variability in medical adherence and responsibility at Time 1 and Time 2. However, given the robust relationship between family functioning and condition management in the pediatric psychology literature (e.g., Hanson, Henggeler, & Burghen, 1987; Lewin et al., 2006; Rapoff, 2011), family functioning would account for the most variance in the development of catheterization and bowel program responsibility and adherence.

This study extended an earlier, cross-sectional investigation of medical adherence and autonomy in the same sample of youth with SB (O’Hara & Holmbeck, 2013). This earlier study found that inattention and executive dysfunction predicted lower levels of medical adherence and autonomy among youth with SB. As SB medical tasks require

higher order cognitive abilities, such as planning, organizing, attending to detail, and problem solving, the authors concluded that more severely impaired children have a lower likelihood of completing medical tasks proficiently and independently (i.e., these children require increased scaffolding and parental support). This study adds to O'Hara and Holmbeck's (2013) initial investigation of the link between neuropsychological functioning and medical responsibility/adherence by evaluating the association between the cognitive profiles of youth (i.e., during late childhood to early adolescence) on medical outcomes across adolescence (i.e., by mid- to late adolescence). Additionally, the current study examined the utility of investigating the link between child neuropsychological functioning and medical responsibility/adherence in comparison to the links for other domains (i.e., family/peer functioning and biological variables).

CHAPTER THREE

METHODS

Participants

Participants were part of a larger longitudinal study at Loyola University Chicago examining family, psychosocial, and neurocognitive functioning among children with SB (e.g., Devine et al., 2011). This study utilized data regarding medical responsibility and adherence, and family, peer, biological, and neuropsychological characteristics from Time 1 and data regarding medical responsibility and adherence at Time 2 (approximately two years after participant data from Time 1 was collected). Families of children with SB were recruited from four hospitals and a statewide SB association in the Midwest. Inclusion criteria consisted of: (1) diagnosis of SB (types included myelomeningocele, lipomeningocele, and meningocele); (2) age eight to 15 years at time 1; (3) ability to speak and read English or Spanish; (4) involvement of at least one primary caregiver; and (5) residence within 300 miles of lab (to allow for home visits for data collection). During recruitment, 246 families who met inclusion criteria were approached. Of the original 246 families, 163 families agreed to participate but 21 of those families could not be contacted or later declined, and two families did not actually meet inclusion criteria (i.e., one child was too young and one for having a milder form of SB). The final participants included 140 families of children with SB (53.6% female; *M* age = 11). Of these children, 60.4% identified as Caucasian, 22.6% were Hispanic,

12.3% were African American, and 4.7% identified as an “other” race. The average Hollingshead Four Factor Index for the sample was approximately 39.44 (SD = 15.90), suggesting a generally middle class sample with some variability. Children of families who declined participation did not differ from those who accepted participation with respect to type of SB (e.g. myelomeningocele or other), $\chi^2(1) = .0002, p > .05$, shunt status, $\chi^2(1) = .003, p > .05$, or occurrence /nonoccurrence of shunt infections, $\chi^2(1) = 1.08, p > .05$.

Child medical information about their physical status was gathered from their medical chart (medical chart release was obtained during the home visit) and from a mother questionnaire. Of the 140 participants, medical chart data indicated that 87.9% had a diagnosis of myelomeningocele, 8.3% lipomeningocele, and 3.8% other. Additionally, over half of the children had spinal lesions in the lumbosacral or lumbar spinal regions (62.9%), 19.0% were sacral and 18.1% thoracic. Also, 80.3% of the children had a shunt. Mother questionnaire data indicated that 81.1% of the children used braces to ambulate and 61.4% used a wheelchair. The average number of shunt surgeries among children with shunts was 3.14 (SD = 5.07). Similar to past studies (e.g., Wills et al., 1990), youth with SB demonstrated a low average IQ (M FSIQ = 85.68, SD = 16.58). Of the 140 children that participated in this study, 26 children (19.7%) had an IQ score less than 70.

As a part of the study, each family was asked to invite a peer to participate. Inclusion criteria for peers were (1) within 2 years of the target child’s age, and (2) ability to speak and read English or Spanish. Families were strongly encouraged to invite friends who were not related to the target child and who were within two years of the target

child's age (ages 6-17 years at Time 1, 8-19 years at Time 2). One hundred twenty-one families were able to recruit a peer within the specified age range. Fifteen peers were related to the child with SB (e.g., cousins, siblings, etc.) and were removed from analyses involving peer data. Thus, 106 children with SB (76% of the entire sample) and their friends were included in the peer analyses. The mean age of friends was 10.98 years ($SD = 2.75$), and 55.7% were female. Regarding racial background, 64.2% were Caucasian, 17.9% were Hispanic, 8.5% were African American, and 6.6% reported they belonged to an "other" racial background. Socioeconomic data were not available for peers.

Design and Procedures

Time 1

Trained undergraduate and graduate student research assistants collected data over the span of two home visits that each lasted about 3 hours. Families and peers who completed all parts of the study received monetary compensation (\$150 for families, \$50 for peers) and gifts (e.g. t-shirts and pens). For participant families, informed consent from parents and assent from children were obtained prior to the start of the first home visit at the participant's house. For peers, informed consent from parents and assent from children were obtained prior to the start of the second home visit at the participant's house. Parents of participants were asked to complete release of information forms to allow for additional data collection from teachers, health professionals, and medical charts.

During the first home visit, children with SB and their parents independently completed questionnaires. To maintain confidentiality, family members were asked to fill out questionnaires independently. If needed, research assistants read the questionnaires

out loud to the child to ensure that he/she understood the questions. Likert scale responses on a laminated card were also available for the child to use in selecting desired responses.

Families also participated in audio- and video-taped structured interaction tasks. The videotaped interactions consist of four structured tasks: (1) an interactive game, (2) discussion of two age-appropriate vignettes about social situations, (3) discussion of transferring disease-specific responsibilities to the child, and (4) discussion of family conflict issues that were frequently endorsed on questionnaires by family members (Smetana, Yau, Restrepo, & Braeges, 1991). The last three tasks were counterbalanced for each family.

For the interactive game, parents and children were asked to play the game “Uno-Stacko”. A research assistant explained the rules to the family and then provided a laminated card of the rules for reference. Families were instructed to play until someone won. For the discussion of two age-appropriate vignettes, families were given two cards that contained two short stories and were asked to answer a series of questions together about the stories. Specific cards were given to families based on child gender (e.g., male children were given stories with male characters). In one story, a child with SB had to attend a new school where the other children do not know him/her or that he/she has SB. In the other story, a child discovers his/her friend does not want to spend time with him/her. Families were asked to read each story out loud, and then discuss all of the questions together in order. Examples of questions included: “How do you think [the character] is feeling?”, “Should [the character] tell anyone about his SB”, and “If

something like this were to happen to you in the future, what would you do?” Families were given 10 minutes to complete this task.

For the discussion of the sharing of SB responsibilities, families were asked to identify one SB related responsibility that is currently managed by the parent but for which the child would have to take responsibility in the future. After identifying this responsibility, families were asked to discuss how the transfer of this responsibility would take place (e.g., how it will be done and by when it will need to be done). If families were unable to identify a SB responsibility, they were asked to think of other responsibilities that would need to shift from the parent to the child. Families were given five minutes to complete this task.

Prior to the conflict task, families were asked to complete the Parent-Adolescent Conflict Scale (PAC; Robin & Foster, 1989). Mother, father, and child reports on this questionnaire were examined and scored by a research assistant. Scores were computed for each item by multiplying discussion frequency by conflict intensity. Items with the five highest scores across respondents were selected for the conflict task. The family was then given 10 minutes to discuss three of these five issues (considered to be “hot” topics; Smetana et al., 1991).

During the first home visit, neuropsychological testing of the child was also done. Assessments of the child’s IQ, executive functioning, motor functioning, and emotion recognition (i.e., where one was required to identify emotions based on pictures and voices) was conducted. Finally, families were asked to select a peer to participate in the second home visit.

During the second visit, the child and peer individually completed questionnaires

and audiotaped interviews about general friendship characteristics and the specific friendship of the participating target child and peer. Target children and peers engaged in four, videotaped, and structured interaction tasks. All but one of the tasks was counter-balanced across dyads. Tasks included: (a) Toy Ranking (the dyad was asked to rank a set of toys based on how much the children enjoyed playing with them), (b) Unfamiliar Object Task (develop a commercial advertising an ambiguous object; five minutes), (c) Plan an Adventure (discuss what the pair would do and where they would go), and (d) Conflict Task (discuss previous peer conflicts and brainstorm problem-solving ideas that could have been used to resolve conflict; this task was always presented last).

Time 2

The second time point of the study took place approximately two years after each participant had completed their first home visit. One hundred eleven of the original 140 participants (M age = 13) completed Time 2 (i.e., 80% of the sample). At this time point, 5 families withdrew from the study and 1 participant passed away. Nine families declined participation at Time 2, but agreed to be re-contacted for Time 3. Eleven families had missing contact information and 3 families consented to the study, but failed to return study materials. Participants at Time 2 did not differ from youth who did not participate with respect to gender, $\chi^2 = 0.28, p > .05$, SES, $t(128) = 1.86, p > .05$, type of SB (myelomeningocele or other), $\chi^2(1) = 1.19, p > .05$, lesion level (thoracic or other), $\chi^2(1) = 0.72, p > .05$, or shunt status, $\chi^2(1) = 2.73, p > .05$. However, youth who did not participate at Time 2 were significantly older at Time 1 ($M = 12.62$ compared to $M = 11.12$), $t(138) = 3.02, p < .01$.

For Time 2, data were collected over the course of one visit to the family's home. Similar to the Time 1, families and peers who completed all parts of the study received monetary compensation (\$150 for families, \$50 for peers). For participant families, informed consent from parents and assent from children were obtained prior to the start of the first home visit at the participant's house. For peers, informed consent from parents and assent from children were obtained upon arrival to the participant's house (approximately an hour and a half after the start of the visit). The structure and content of the Time 2 home visit were essentially the same as Time 1. Namely, the visit consisted of: the completion of child, parent, and peer questionnaires, audio-taped parent, child, and peer interviews, neuropsychological testing, and videotaped family and peer interactions. However, this study only utilized data obtained from mother and father questionnaires regarding medical responsibility and adherence for catheterization and bowel management at Time 2.

Measures

Medical Responsibility (Time 1 and Time 2)

We utilized the Sharing of Spina Bifida Management Responsibilities (SOSBMR), an adaptation from the Diabetes Family Responsibility Questionnaire (DFRQ; Anderson et al., 1990), to examine who takes primary responsibility over SB medical tasks. The SOSMBR consists of 34- items that describe SB or general health-related issues or tasks relevant to children with SB (e.g. "Remembering to catheterize regularly, every 2-4 hours"). Parents independently rated who was primarily responsible for each task (e.g. Parent, Child, Equal, or Not Applicable). On this measure, mother and father-reports were significantly correlated at Time 1 ($r = .76$) and Time 2 ($r = .75$).

Thus, for data reduction purposes, mother and father reported were combined to form one parent score (i.e., by computing the mean). This new, combined parent-report of medical responsibility was found to have acceptable alphas for the bowel (i.e., two items related to bowel program) and catheterization (i.e., three items related to catheterization) subscales at Time 1 ($\alpha = .97$ for the bowel subscale and $\alpha = .94$ for catheterization subscale) and Time 2 ($\alpha = .95$ for the bowel subscale and $\alpha = .97$ for catheterization subscale).

This project evaluated responsibility with catheterization and bowel programs only, as these tasks are the most prominent components of SB medical care. Responsibilities for catheterization and bowel programs were dichotomized to create categorical variables: “Responsible” with catheterization or bowel programs or “Not Responsible” with catheterization or bowel programs. For this measure, a score of “1” was assigned to tasks that parents are primarily responsible, “2” to tasks that are shared between the parent and child, and “3” to tasks that children are primarily responsible. In this way, higher scores indicated higher child responsibility. For catheterization and bowel programs, total mean scores (ranging from 1 to 3) were calculated for subscale items. Means above or equal to 2.1 (i.e., slightly above “shared responsibility”) was assigned a score of “1” or child “Responsible”, whereas means below 2.1 (i.e. scores ranging from “shared responsibility” to “parent responsibility”) was assigned a “0” or child “Not Responsible”.

Medical Adherence (Time 1 and Time 2)

The SB Self-Management Profile (SBSMP; Wysocki & Gavin, 2006) measured adherence to SB medical treatments. The SBSMP is a 14-item, structured interview that

addresses seven dimensions of SB medical regimen, including appointment keeping, bowel control program, skin and wound care, exercise, medications, clean intermittent catheterization, and dealing with urinary tract infections. On this measure, higher scores indicate higher levels of SB medical adherence. When developing this measure, item content, wording, and scoring was developed based on a consultation with Dr. Wysocki (the developer of the original version of this measure for youth with type 1 diabetes). Internal consistency for this measure is adequate, with a Cronbach's alpha reliability coefficient of .66 for mothers of children with SB (Wysocki & Gavin, 2006). For this study, the SBSMP was administered as a questionnaire rather than an interview. We evaluated mother and father responses on the SBSMP. Due to significant correlation at Time 1 ($r = .45, p < .05$; see Table 1) and 2 ($r = .35, p < .05$) responses were combined to form one score for each time point (i.e., by computing a mean score). Scale reliability could not be computed for this sample due to a low number of participants who completed every item (i.e., parents can endorse "not applicable" for certain items).

Similar to how we analyzed the SOSMBR, this study formed categorical variables from items assessing adherence to catheterization and bowel programs. For catheterization, two items were used to assess adherence. The first item asked parents to rate how often families had catheterized to schedule in the past 6 months. Based on clinical recommendations (that children with SB catheterize every 3-4 hours), a score of "1" or "adherent" was given to families who indicate that they miss catheterizing 4-5 times per week or less (i.e., less than once per day). A score of "0" or "nonadherent" was given to families who indicate that they miss catheterizing one time (i.e., failing to catheterize within a 6-8 hour window) or more per day. The second catheterization item

asks parents to rate how well they follow the five steps of clean intermittent catheterization in the past 6 months (i.e., having all the supplies together, washing hands first, correct positioning of the child, and inserting the catheter with slow steady pressure until urine begins to flow). Based on clinical judgment, a score of ‘1’ or adherent was assigned to families who indicate that they complete three or more of the five steps (i.e., more than 50% of the steps). We assigned a score of “0” or “nonadherent” to families who indicate that they complete two steps or fewer (i.e., less than 50% of the steps). If the family indicated that their child is not required to catheterize (e.g., “N/A” responses for all three items), families were dropped from the analyses. For overall adherence to catheterization, families who reported non-adherence to one or more of the catheterization items were considered “non-adherent”.

For assessing adherence to bowel programs, two items were utilized. The first items asked parents to rate how often their child has stayed within prescribed diet recommendations over the past 6 months. Or score of “1” or “adherent” was assigned to families who indicated that their child eats according to diet recommendations 50% of the time or greater. A score of “0” or “nonadherent” q assigned to families who indicated that the child eat according to diet recommendations less than 50% of the time. The second item related to adherence to bowel program asked parents to rate how often their child adheres to taking medication to reduce constipation (i.e., suppositories, enemas, or stool-softening medications). A score of “1” or “adherent” was assigned to families who indicated that their child takes bowel medications 50% of the time or greater. A score of “0” or “nonadherent” was assigned to families who indicated that the child takes bowel medications less than 50% of the time. If the family indicates that their child is not

required to complete a bowel program (e.g., “N/A” responses to both items), families were dropped from the analyses. For overall adherence to bowel recommendations, families who reported non-adherence to one or more of the bowel program items were considered “non-adherent”.

Demographic and Biological Information

Demographic information was obtained from responses by parents that included gender of the child, ethnicity of family members, parental occupation, parental educational attainment, family annual income, developmental milestones, and family structure. Furthermore, medical records were utilized to determine the physical status of each participant at Time 1, including lesion level and number of shunt revisions/infections. Health professionals and research assistants conducted medical chart reviews for each participant that provided consent. For this project, we analyzed lesion level as a continuous variable. Specifically, we assigned participants a score that ranged from 1 to 30, with lower numbers representing lower-level lesions (e.g., a score of ‘1’ represented a lesion in the S5 region, whereas a score of ‘30’ represented a C1 lesion). Among participants, the average lesion level was approximately in the L4 region.

To assess the target’s gross motor functioning, we utilized a modified version of *The Gross Motor Function Classification System Expanded and Revised* (GMFCS-E&R) developed by Palisano, Rosenbaum, Bartlett, & Livingston (2007). Specifically, we utilized information from mother-report on a Medical History Questionnaire (and utilized father-report in the absence of mother data) to assign the target a gross motor classification scale level based on the following: Level I: No braces, crutches, walker, or wheelchair (i.e., 100% unassisted walking), Level II: Uses braces, crutches, or walker,

Level II: Some wheelchair use, but able to walk with braces (> 50% walking), Level IV: Uses wheelchair at school and/or long outings, may walk for short distances with a walker (< 50% walking), and Level V: all areas of motor functioning limited, no means of independent mobility.

Neuropsychological Functioning

General intellectual functioning. The Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) was utilized as a proxy for general intellectual functioning. The WASI includes tasks within the performance and verbal domains, and is frequently utilized to provide an intelligence quotient (IQ). Specifically, the Vocabulary and Matrix Reasoning subtests were administered to participants in the present study to obtain an estimate of IQ. The Vocabulary subtest is a 42-item measure that assesses for expressive vocabulary, verbal knowledge, and fund of information. In addition, it is a reliable measure of crystallized intelligence and general intelligence (e.g., Wechsler, 1999). On items one through four, the examinee is required to name pictures (e.g., bucket). On items five through 42, words are orally and visually presented, and the examinee is required to provide a definition (e.g., what is a car?). The Matrix Reasoning subtest assesses nonverbal abstract problem solving, inductive reasoning, and spatial reasoning skills. In addition, it has been found to be a reliable and valid measure of fluid intelligence, correlating .81 with another common measure of fluid intelligence (Wechsler, 1999). Matrix Reasoning consists of 35-items, each consisting of an incomplete pattern. The examinee is asked to complete the pattern by selecting the best choice from five options. In general, higher scores on these measures represent higher levels of intellectual abilities. Standardized norms for both of these subtests have been

obtained across 2,245 individuals aged six through 89, and average test-retest reliability coefficients of .89 (Vocabulary) and .92 (Matrix Reasoning) were obtained for children 6 to 16 years old (Wechsler, 1999).

Executive functions. The Behavior Rating Inventory of Executive Function (BRIEF; Gioia et al., 2000a, 2000b) is a parent- and teacher-report questionnaire that measures several domains of executive functions of children. It is composed of eight subtests including Inhibit (i.e., the ability to resist or not act on an impulse; e.g., Interrupts others), Shift (i.e., the ability to move freely from one situation, activity or aspect of a problem to another demand; e.g., Becomes upset with new situations), Emotional Control (i.e., the capacity to modulate emotional responses; e.g., Overreacts to small problems), Initiate (i.e., the capacity to begin a task or activity or independently generate ideas, responses, or problems solving strategies; e.g., Does not take initiative), Working Memory (i.e., the ability to hold information in mind for the purpose of completing a task; e.g., Has trouble remembering things, even for a few minutes), Plan/Organize (i.e., the ability to manage current and future-oriented task demands; e.g., Has good ideas but cannot get them on paper), Organization of Materials (i.e., orderliness of work, play, and storage spaces; e.g., Keeps room messy), and Monitor (i.e., work-checking habits; e.g., Makes careless errors) subtests. These subtests fall within two broad indices, Behavioral Regulation and Metacognition, which make up the overall Global Executive Composite Score. Mothers, fathers, and teachers completed all 86 items that comprise the BRIEF subtests. On each item, parents and teachers were instructed to circle whether their child has never, sometimes, or often demonstrated a particular behavior during the past six months. Higher scores on the BRIEF represent higher levels

of executive dysfunction. Using the same data, O'Hara and Holmbeck (2013) found that mother, father, and teacher-reports for the item mean scores were moderately correlated ($r = .30$ to $.57$). Thus, for this study, we employed the mean across reporters. O'Hara and Holmbeck (2013) reported adequate internal consistency ($\alpha = .98$) for the entire combined scale at Time 1 (e.g., mother, father, and teacher report).

Several neuropsychological measures were utilized as an assessment of performance-based executive functions. The Cognitive Assessment System (CAS; Naglieri & Das, 1997) is an assessment battery of tests that measure cognitive processing in children 5 to 17 years of age. Specifically, the Planned Connections subtest of the CAS was utilized as an assessment of nonverbal executive function (i.e., planning). On this test, the examinee was first required to sequentially connect numbers that appear in a quasi-random order on a page, and then the examinee was required to connect both numbers and letters in serial order alternating between numbers and letters (e.g., 1-A-2-B-3, etc.). Each test was timed to provide an estimate of task efficiency. Scores were then transformed into age scaled scores, with higher scores representing higher levels of executive function ability.

Selected subtests from the Delis Kaplan Executive Function System (D-KEFS; Delis, Kaplan, & Kramer, 2001) were also utilized as an assessment of executive function. The D-KEFS is a comprehensive battery of tests that measure higher-level cognitive functions including reasoning, problem solving, and planning. This study utilized the Verbal Fluency Test of the D-KEFS as a measure of verbal executive functions. The Verbal Fluency Test is comprised of three subtests including Letter Fluency, Category Fluency, and Category Switching. For each of these three conditions,

the examinee was given 60 seconds to generate words that start with a particular letter (e.g., the letter 'F') fluently in an effortful, phonetic format (Letter Fluency), from over learned concepts (e.g., types of animals and names of boys/girls; Category Fluency), and while shifting between over learned concepts (e.g., name a fruit, then a piece of furniture, then a fruit, etc.; Category Switching). Letter and Category fluency scores were computed based on the total number of correct responses. On the Category Switching subtest, two scores were computed: total number of correct responses and total number of correct switches between concepts. Across all subtests, higher scores represented higher levels of executive function ability. All scores were computed as age scaled scores.

We created a composite score based on the mean age scaled scores across the neuropsychological test data that measure executive function (with higher scores indicating higher levels of executive function skills). Scales in this composite score included the D-KEFS Verbal Fluency subtests (i.e., Letter Fluency, Category Fluency, and Category Switching) and the Planned Connections subtest from the CAS. Adequate internal consistency was demonstrated across these scales ($\alpha = .89$ at Time 1) in an earlier study (O'Hara & Holmbeck, 2013). As stated above, higher scores on the BRIEF indicate higher levels of executive dysfunction, whereas higher scores on performance-based executive function tasks indicated higher executive function.

Family Functioning

Family conflict (questionnaire data; time 1). Mothers and children separately completed The Parent-Adolescent Conflict scale (PAC), a brief version of the Issues Checklist (IC; Robin & Foster, 1989). The PAC measures conflict by asking informants to respond to 15 potential conflict issues that are commonly discussed in all families

during adolescence (e.g. whether or not the child does chores around the house) and 10 potential conflict issues that are typically discussed in families of children with SB (e.g. how he/she does his/her catheterization). For each issue, respondents are asked to indicate whether or not the issue was discussed in the past 2 weeks. If the issue had been discussed, respondents are asked how many times the issue was discussed and how intense those conversations were. Intensity is rated on a Likert scale (ranging from “calm” to “angry”). Items on the PAC are organized into two subscales: medical conflict and non-medical conflict. Alpha coefficients were not computable for this measure, as each family member only answers items they have personally discussed and each respondent rarely answers *every* item (i.e. the SPSS algorithm for reliability employs listwise deletion and only includes participants that responded to *all* items).

This study utilized mother, father, and child reports of conflict intensity on the PAC. Since mother- ($r = .46, p = .00$), father ($r = .45, p = .00$), and child-reports ($r = .46, p = .00$) of medical and non-medical conflict were significantly correlated at Time 1, medical and non-medical conflict scores were combined to form a general measure of conflict (i.e., mother-reported overall conflict, father-reported overall conflict, and child-reported overall conflict). Mother- and father-reports of overall conflict were correlated ($r = .48, p = .00$), thus, we computed the mean intensity to form a parent-reported conflict. Child-reported conflict was significantly correlated with father-report ($r = .23, p < .05$), but not mother-report ($r = .15, p > .07$). Thus, we separately examined parent- and child-reported overall family conflict.

Family cohesion (time 1). The Family Environment Scale (FES; Moos & Moos, 1994) measures social and environmental characteristics of the family. The current study

revised Form R, which measured true-false responses, to measure parents' perceptions of their family environments on a Likert scale. The FES includes three main dimensions, comprising a total of ten subscales. The subscales are grouped according to domains, including the Relationship dimension (cohesion, expressiveness, and conflict subscales), Personal Growth dimension (independence, achievement orientation, intellectual-cultural orientation, active-recreational orientation, and moral-religious emphasis subscales), and the System Maintenance dimension (organization and control subscales). We employed mother and father reports on the Family Cohesion subscale. The Family Cohesion subscale is a nine-item scale intended to measure the degree of commitment, help, and support family members provide to one another. Due to a high correlation ($r = .46$, $p < .001$), mother- and father-reports were combined (i.e., by computing a mean score) to form one, cohesion score. Internal consistency was adequate for the parent-reported cohesion subscale ($\alpha = .62$).

Family conflict, family cohesion (observational data; time 1). This study also investigated family conflict and cohesion by evaluating family interaction tasks from Time 1. A trained undergraduate or graduate research assistant coded each family interaction task using the Family Interaction Macro-coding System (FIMS; Holmbeck et al., 2007; Kaugars et al., 2011), an adaptation of the coding system developed by Smetana et al. (1991). Research assistants received approximately 10 hours of training prior to coding the videotapes. Training included the coding of previously coded interactions and discussing each code with an expert coder. Coders were instructed to view one interaction at a time and then rate the interaction on a variety of dimensions. Once training was complete, research assistants independently scored five videos and

were required to reach a reliability of 90% agreement with an expert coder. The FIMS consists of 113 separate codes. Coded items assess interaction style, conflict, affect, control, and problem solving at the individual-, dyadic- (mother/father, mother/child, father/child), and systemic-level (family) using 5-point ratings. For example, the item assessing “Level of Conflict” captures signs of conflict in a dyadic relationship as shown through verbal or nonverbal behaviors (1 = “Not At All” and 5 = “Very Much”). Two research assistants independently coded each interaction task (game, conflict, transfer of responsibility, and vignettes).

For this project, we examined The Family Cohesion and Family Conflict subscales. The Family Cohesion subscale includes the following 7 items: Requests Input (dyadic), Involvement (individual), Collaboration (systemic), Openness (systemic), Reaches Agreement (systemic), Parents Present as United Front (systemic), and Disengagement (systemic, reverse-coded; $\alpha = .90$). The Family Conflict subscale consists of the following 2 items: Conflict (dyadic) and Attempts Resolution (individual; reverse-coded; $\alpha = .66$; Kaugars et al., 2011). Interrater reliability between the two coders was adequate for the Family Cohesion ($ICC = .77$) and Family Conflict subscales ($ICC = .60$).

Family stress (time 1). The Family Stress Scale (FSS; Quittner, Glueckauf, & Jackson, 1990) consisted of 19 items assessing common stressors in families with a child with SB on a five-point scale. The measure includes 13 non-disease specific items (e.g., outings in the community) and 6 disease-specific items (e.g., catheterization). Mother- and father-reported family stress was significantly correlated at Time 1 ($r = .40, p < .001$); thus, we computed the mean across the raters to form one, parent-reported stress score. Internal consistency was adequate for parent-reported family stress ($\alpha = .92$).

Social Adjustment

Peer conflict (observational data; time 1). The Peer Interaction Macro-Coding Scale (PIMS) is an adaptation of several previous coding systems (Holmbeck, Belvedere, Gorey-Ferguson, & Schneider, 1995; Johnson & Holmbeck, 1999; Smetana, Yau, Restreppo, & Braeges, 1991) and draws upon codes used in other systems (Allen et al., 1998; Allen, Porter, & McFarland, 2002; Buhrmester, Camparo, Christiansen, Gonzalez, & Hinshaw, 1992; Julien, Markman, Lindahl, Johnson, & Van Widenfelt, 1987; Levy, 1943; Paikoff, 1992). Coders were undergraduate and graduate student research assistants who viewed an entire peer interaction task before rating the target child and peer on a multitude of items. For all codes, a five-point Likert scale was used with descriptive anchors specific to each code. For example, the item “eye contact” featured the following anchors: 1 – *not at all*; 2 – *rarely*; 3 – *sometimes*; 4 – *frequently*; 5 – *very often*.

Coders were trained for ten hours before coding the videotapes. They were required to achieve a 90% agreement rate on practice items before they were authorized to code study videotapes (i.e., “agreement” = concordance across coders within one point on the Likert scale). For each of the four interaction tasks, two coders rated behaviors, and item level means across coders for each task were averaged across the tasks to produce a single score for each target child and friend separately (for codes assessing individual constructs) or for each pair (for codes assessing dyadic constructs). For this project, only the dyadic conflict code was utilized. The dyad conflict code consists of 5 items: ratings of their ability to reach an agreement/resolution, resolution of issues, level of conflict within dyad, negative escalation, and toleration differences and disagreements. At Time 1, inter-rater reliability for the Conflict scale was adequate (SB target: ICC =

.75; peer: ICC = .77; Holbein, Zebracki, & Holmbeck, 2015), as was the internal consistency (SB target $\alpha = .86$; peer $\alpha = .89$).

Overall friendship quality (time 1). The Friendship Activity Questionnaire (FAQ) is based on Bukowski's Friendship Qualities Scale (Bukowski, Hoza, & Boivin, 1994) and consists of 46-items. This measure instructs participants to rate their best friend across five scales of friendship qualities: companionship (e.g. "My friend and I spend a lot of our free time together"), conflict (e.g. "I can get into fights with my friend"), help (e.g. "If other kids were bothering me, my friend would help me"), security (e.g. "If I have a problem at school or at home, I can talk to my friend about it"), and closeness (e.g. "I think about my friend even when my friend is not around"). Respondents are asked to rate how true each statement is for his/her friendship on a five-point Likert scale with responses ranging from '1-not true' to '5-really true.' Internal consistencies for all subscales are high, with alpha reported between .71 and .86 (Bukowski et al., 1994). This study utilized the target child's total score on this measure, which showed adequate internal consistency ($\alpha = .88$).

Emotional support from peers (time 1). The Emotional Support Questionnaire (ESQ) is an extension of Slavin's Perceived Emotional/Personal Support Scale (PEPSS; Slavin, 1991), a measure of emotional support among individuals 14-19 years old in which adolescents nominated three individuals from three broad social categories: family members, non-family adults, and friends, and rate them across four dimensions (i.e., degree to which a child talks to this person about personal concerns, how close the child feels to them, how much they talk about the child's concerns, and how satisfied the child is with how much help and support they give). The three items added for this study

included (a) how much do the respondent and individual rated get upset with or mad at each other, (b) how much does the respondent play around and have fun with the individual rated, and (c) how sure the respondent is that this relationship will last no matter what. Respondents rated each relationship across these 7 items (on a Likert scale with responses ranging from '1- hardly at all', '2- a little', '3- pretty much', and '4- very much'). This study utilized the target child's mean score from the friend category only across all seven dimensions. Internal consistency for emotional support from peers was good ($\alpha = .88$).

CHAPTER FOUR

RESULTS

Preliminary Analyses

Means, standard deviations, and scale ranges for variables utilized in the analyses are presented in Table 1. For descriptive analyses, we examined medical outcome variables (i.e., medical adherence and child responsibility) as continuous variables (see Tables 1 and 2). Skewness analyses were conducted for all variables using guidelines established by Tabachnick and Fidell (2001). Conservative alpha levels (.001) were employed to evaluate the significance of skewness, in which z-score values greater than 3.29 were considered significantly skewed. These analyses revealed that the following variables were significantly positively skewed: number of shunt revisions (z-score = 7.22), lesion level (z-score = 4.82), the observational measure of peer conflict (z-score = 5.28), parent-reported family conflict (z-score = 5.6), child-reported family conflict (z-score = 3.33), and parent-reported family stress (z-score = 3.97). First, square root transformations were conducted on these variables. The observational peer conflict variable continued to be significantly skewed after square root transformations (z-score = 3.72). Thus, logarithm transformations were computed on this variable only, and this transformed variable was no longer significantly skewed. Preliminary analyses were conducted to determine the degree of association among demographic (i.e., child age and SES), biological, neuropsychological, family, and peer variables (see Table 2) from Time

1. Among the demographic variables, child age was associated with number of shunt revisions ($r = .31$), IQ ($r = -.24$), performance measures of executive functions ($r = -.25$), observed peer conflict ($r = -.34$), and friendship quality ($r = -.19$). Thus, older children had more shunt revisions, higher performance-based executive functions, less peer conflict, and lower friendship quality. Socioeconomic status was significantly associated with IQ ($r = .47$), performance measures of executive functions ($r = .27$), and peer conflict ($r = -.22$). That is, youth with higher SES had higher scores on IQ and performance measures of executive functions, and lower peer conflict.

Correlations among the biological variables showed an association between lesion level and gross motor functioning ($r = .57$). That is, higher lesion levels were associated with greater limitation in gross motor functioning. In terms of neuropsychological variables, IQ was positively related to performance-based executive functions ($r = .77$) and negatively related to the combined parent- and teacher-report on the BRIEF ($r = -.024$). In other words, higher IQ was associated with better performance on executive function measures and lower parent- and teacher-reported executive dysfunction.

Regarding family variables, the observational measure of family conflict was positively associated with family stress ($r = .19$) and negatively correlated with observed family cohesion ($r = -.43$). Parent-reported conflict was also positively associated with family stress ($r = .28$) and child-reported conflict ($r = .25$), and negatively correlated with the observational measure of family cohesion ($r = -.18$). The observational and questionnaire measure of family cohesion were negatively correlated with family stress ($r = -.23$ for both measures). Taken together, correlational data revealed that families, who

were low in conflict, demonstrated high cohesion and low family stress. Among the social variables, friendship quality was positively correlated with emotional support from peers ($r = .43$).

Although we utilized categorical variables of catheterization and bowel program adherence/responsibility to explore the main study objectives (see Figure 1), we conducted preliminary analyses to explore associations among continuous medical adherence and responsibility variables at Time 1 and Time 2 (i.e., total adherence and child responsibility scores, across all medical domains; see Table 2). Since subscales for the medical adherence measure are not yet established (i.e., subscales for catheterization and bowel program), we utilized total scores, across all medical domains.

Regarding adherence, mother-reported medical adherence at Time 1 was positively correlated with father-reported adherence at Time 1 ($r = .48$) and mother-reported adherence at Time 2 ($r = .51$). Mother-reported adherence at Time 2 was positively correlated with father-reported adherence at Time 2 ($r = .35$). Thus, in most cases, adherence ratings were positively correlated across reporter (mother and father) and time point. As stated in the methods section, due to significant correlations for mother- and father-reported medical adherence at Time 1 and 2, responses were combined to form one score for each time point (i.e., by computing a mean score). In terms of medical responsibility, mother-reported child responsibility at Time 1 was positively associated with father-reported responsibility at Time 1 ($r = .71$) and Time 2 ($r = .67$), and mother-reported responsibility at Time 2 ($r = .75$), suggesting a strong correlation between mother- and father- ratings of child responsibility and across time. Thus, we combined mother and father responses to form one score for each time point

(i.e., by computing a mean score). As described in a previous study with the same dataset (Psihogios, Kolbuck, & Holmbeck, 2015), mother- and father-reported child responsibility at Time 1 were negatively associated with mother-reported adherence at Time 1 (i.e., poorer adherence at Time 1 was associated with higher levels of child responsibility).

Correlational data also revealed several significant relationships between continuous medical outcome variables, study predictors, and demographic data (see). Specifically, higher mother-reported adherence at Time 1 was associated with more limitations in gross motor functioning ($r = .25$), suggesting higher adherence in families of children with more severe gross motor deficits. In addition, higher levels of mother-reported adherence at Time 2 were associated with higher lesion levels ($r = .31$), fewer executive function problems ($r = -.32$), and lower parent-reported family conflict ($r = -.22$). Father-reported adherence at Time 1 related to more limitations in gross motor functioning ($r = .25$), and lower child-reported family conflict ($r = -.34$). At Time 2, higher mother-reported adherence related to the lower executive dysfunction on the BRIEF ($r = -.20$), lower parent-reported family conflict ($r = -.22$), and higher child-reported friendship quality ($r = .21$). Higher father-reported medical adherence at Time 2 correlated with more shunt revisions ($r = .29$), lower executive dysfunction on the BRIEF ($r = -.26$) and lower family stress ($r = -.25$). Taken together, higher adherence was associated with indicators of more severe SB (e.g., higher lesion level, more shunt revisions, and more limitations in gross motor functioning), higher executive function skills (as measured by fewer problems reported on the BRIEF), lower family conflict, and higher friendship quality.

In terms of medical responsibility, higher mother-reported child medical responsibility at Time 1 correlated with older child age ($r = .36$), higher IQ ($r = .25$), higher performance on executive function measures ($r = .24$), and higher friendship quality ($r = -.20$). Higher father-reported child medical responsibility at Time 1 significantly related to older child age ($r = .41$), higher SES ($r = .23$), lower lesion level ($r = -.24$), greater gross motor functioning ($r = -.29$), IQ ($r = .32$), lower executive dysfunction on the BRIEF ($r = -.24$), higher performance-based executive functions ($r = .27$), and lower family stress ($r = -.25$). Higher mother-reported child responsibility at Time 2 correlated with older child age ($r = .41$), higher SES ($r = .23$), higher IQ ($r = .32$), lower executive dysfunction on the BRIEF ($r = -.30$), higher performance-based executive functions ($r = .29$), lower family stress ($r = -.21$), lower peer conflict ($r = -.23$), and higher emotional support from peers ($r = .24$). In sum, correlational data revealed higher levels of child responsibility related to older age, higher SES, less severe SB (e.g., fewer gross motor functioning problems and lower lesion level), higher intellectual abilities (e.g., lower executive dysfunction on the BRIEF, higher performance-based executive function skills, and higher IQ), lower family stress, and higher social adjustment (e.g., higher friendship quality, lower peer conflict, and higher emotional support from peers).

Table 1. Means, Standard Deviations, and Ranges for Demographic, Biological, Neuropsychological, Family, Peer, and Continuous Medical Variables

Variable	N	Mean	SD	Min - Max
Demographic Variables (Time 1, 2)				
Child Age (Time 1)	132	11.28	2.42	8.00-15.00
SES (Time 1)	130	39.44	15.90	8.00-66.00
Child Age (Time 2)	99	13.41	2.41	10.00-18.00
SES (Time 2)	106	39.86	15.91	8.00-66.00
Biological Variables (Time 1)				
Lesion Level	121	7.37	3.41	1.00-16.00
Number of Shunt Revisions	94	2.95	3.61	0.00-16.00
GMF Classification Scale	133	2.89	1.07	1.00-4.00
Neuropsychological Variables (Time 1)				
Parent/Teacher-Report (BRIEF)	138	1.69	0.32	0.90-2.68
Executive Function Test Data	128	6.88	3.13	1.00-13.75
Intellectual Function Test Data (WASI)	134	85.85	19.70	55.00-137.00
Family Variables (Time 1)				
Family Conflict Subscale (Observed)	139	2.76	0.21	1.00-3.35
Family Cohesion Subscale (Observed)	139	3.36	0.40	2.24-4.19
Parent-Report Family Conflict (PAC)	133	1.70	0.51	1.00-3.91
Child-Report Family Conflict (PAC)	128	1.64	0.59	1.00-3.10
Parent-Report Family Cohesion (FES)	127	3.11	0.10	2.22-3.89
Parent-Report Family Stress (FSS)	127	1.99	0.85	1.00-4.21
Peer Variables (Time 1)				
Peer Conflict (Observational)	123	1.93	0.42	1.15-3.55
Emotional Peer Support (ESQ)	119	3.08	0.58	1.00-4.00
Friendship Quality (FAQ)	126	3.62	0.54	1.87-4.64
Medical Outcome Variables (Time 1, 2)				
Z-SBSMP (M, Time 1)	121	-.02	.50	-1.69-1.15
Z-SBSMP (F, Time 1)	97	-.01	.48	-2.04-0.88
Z-SBSMP (M, Time 2)	100	.01	.48	-2.46-0.81
Z-SBSMP (F, Time 2)	77	.03	.46	-1.13-1.05
SOSBMR (M, Time 1)	120	1.76	.41	1.00-3.00
SOSBMR (F, Time 1)	97	1.69	.41	1.00-3.00
SOSBMR (M, Time 2)	100	1.96	.44	1.00-3.00
SOSBMR (F, Time 2)	76	1.85	.47	1.00-3.00

Notes. SD = standard deviation; BRIEF = Behavioral Rating Inventory of Executive Function; WASI = Wechsler Abbreviated Scale of Intelligence; PAC (Parent-Adolescent Conflict Scale); FES = Family Environmental Scale; FSS = Family Stress Scale; ESQ = Emotional Support Questionnaire; FAQ = Friendship Activity Questionnaire; Z-SBSMP = Z-Score from adherence measure: the Spina Bifida Self-Management Profile; SOSBMR = Measure of SB Medical Responsibility: Sharing of Spina Bifida Management Responsibilities; M = Mother-report; F = Father-report

Table 2. Pearson Correlations for Child Demographic, Biological, Neuropsychological, Family, and Peer Variables (Time 1), and Continuous Medical Outcome Variables (Total Medical Adherence and Responsibility Scores at Time 1 and Time 2)

Variables	1	2	3	4	5	6	7	8	9	10	11	12	13
1. Child Age (T1)	--												
2. SES (T1)	-.09	--											
3. Lesion Level	.20*	-.15	--										
4. Shunt Rev	.31*	.01	-.01	--									
5. GMF	.09	-.15	.57*	.08	--								
6. BRIEF	-.08	.03	-.01	-.00	.07	--							
7. EF Test Data	-.25**	.27*	-	-	-	-	--						
8. WASI	-.24**	.47*	-.23*	-.16	-.20*	-.24*	.77*	--					
9. Family Conflict (O)	-.08	-.16	-.43*	-.07	-.14	.16	-.03	-.12	--				
10. Family Cohesion (O)	-.15	.34*	-.05	-.10	.10	-.14	.32*	.33*	-.43*	--			
11. Family Conflict (P)	.02	.13	-.22*	.21*	-.17*	.34*	-.01	.06	.23*	-.18*	--		
12. Family Conflict (C)	.01	-.05	.01	.16	.00	.15	-.03	-.09	.08	-.24*	.25*	--	
13. Family Cohesion (P)	.02	.08	-.17	-.13	-.10	-.30*	.11	.05	-.06	.15	-.17	.05	--
14. Family Stress (P)	-.01	-.02	.08	.14	.18*	.38*	-.10	.04	.22*	-.23*	.28*	-.06	-.23**
15. Peer Conflict (O)	-.34**	-.22*	-.02	-.04	-.11	.23*	-.07	-.07	.23*	-.18	.04	.05	-.04
16. Emotional Support (C)	.04	.02	.20*	-.09	.13	-.23*	.17	.14	-.14	.05	-.17	-.10	.03
17. Friendship Quality (C)	-.19*	-.01	.06	-.20	-.04	-.03	.03	-.03	.13	.03	-.07	-.01	.09
18. Z-SBSMP (T1, M)	.06	-.10	.31*	.10	.25*	-.33*	-.11	-.10	-.14	.18	-.22*	-.04	.15
19. Z-SBSMP (T1, F)	.06	.00	.17	.19	.25*	-.14	-.15	-.15	-.18	.13	-.12	-.34*	.09
20. Z-SBSMP (T2, M)	.05	-.10	.13	.01	.06	-.20*	-.02	-.10	-.15	-.06	-.22*	-.02	.16
21. Z-SBSMP (T2, F)	.10	-.20	-.00	.29*	-.11	-.26*	-.04	-.08	.03	-.04	-.20	-.08	.01
22. SOSBMR (T1, M)	.36**	.10	-.14	-.08	-.14	-.16	.24*	.25*	-.09	.16	.02	-.15	-.05

23. SOSBMR (T1, F)	.28**	.23*	- .24*	-.06	- .26*	- .24*	.27* *	.32* *	-.01	.04	.19	.06	.05
24. SOSBMR (T2, M)	.41**	.23*	-.21	-.21	-.20	- .30*	.29* *	.32* *	-.06	.15	.07	-.11	.02
25. SOSBMR (T2, F)	.45**	.24*	-.19	-.10	-.19	- .31*	.26* *	.31* *	-.13	.23*	.01	-.14	-.06

Table 2, Continued. Pearson Correlations for Child Demographic, Biological, Neuropsychological, Family, Peer, and Continuous Medical Outcome Variables (Total Medical Adherence and Responsibility Scores)

Variables	14	15	16	17	18	19	20	21	22	23	24	25
1. Child Age (T1)												
2. SES (T1)												
3. Lesion Level												
4. Shunt Revisions												
5. Gross Motor Functioning												
6. BRIEF (M, F, T)												
7. EF Test Data												
8. WASI												
9. Family Conflict (O)												
10. Family Cohesion (O)												
11. Family Conflict (M, F)												
12. Family Conflict (C)												
13. Family Cohesion (M, F)												
14. Family Stress (M, F)	--											
15. Peer Conflict (O)	.06	--										
16. Emotional Support (C)	-.11	-.06	--									
17. Friendship Quality (C)	-.01	.09	.43* *	--								
18. Z-SBSMP (T1, M)	-.14	.10	.06	.02	--							
19. Z-SBSMP (T1, F)	-.04	-.10	.09	.01	.48* *	--						
20. Z-SBSMP (T2, M)	-.06	.06	.18	.21*	.51* *	.07	--					
21. Z-SBSMP	-.26*	.13	.22	-.16	.21	.18	.35*	--				

(T2, F)							*					
22. SOSBMR (T1, M)	-.13	-.17	.09	-.20*	-.20*	-.08	-.07	.05	--			
23. SOSBMR (T1, F)	-.25*	-.04	.15	-.14	-.26*	-.06	-.15	.12	.76*	--		
24. SOSBMR (T2, M)	-.21*	-.23*	.24*	-.08	-.06	.01	-.08	.04	.81*	.71*	--	
25. SOSBMR (T2, F)	-.30*	-.22	.24	.10	-.17	.06	-.20	-.15	.64*	.67*	.75*	--
									*	*	*	

Notes. T1 = Time 1; T2 = Time 2; M = Mother-report; F = Father-report; C = Child-report; P = Combined parent-report; T = Teacher-report; O = Observational; GMF = Gross motor functioning (higher scores indicate higher level of impairment); BRIEF = Behavioral Rating Inventory of Executive Function (higher scores indicate more executive dysfunction); WASI = Wechsler Abbreviated Scale of Intelligence; Z-SBSMP = Z-Score from adherence measure: the Spina Bifida Self-Management Profile; SOSBMR = Sharing of Spina Bifida Management Responsibilities *p < .05; **p < .01

Additionally, we conducted preliminary analyses to determine frequencies in each medical adherence and responsibility group (i.e., four categorical groups, see Figure 1), for bowel and catheterization management at Time 1 and Time 2 (see Tables 3 and 4, respectively). For bowel management, frequency analyses showed that the majority of children were characterized as “Adherent, Not Responsible” for their bowel program at Time 1 (i.e., 50.0% of sample) and Time 2 (i.e., 41.0% of the sample; see Table 3). Interestingly, from Time 1 to Time 2, the percentage of participants who were “Adherent, Responsible” increased from 8.5% to 21.0%, whereas the percentage of participants who were “Non-adherent, Not Responsible” dropped from 28.2% to 18.3%.

Frequency analyses for catheterization adherence and responsibility showed a different trend, with the majority of the children characterized as “Adherent, Responsible” at Time 1 (43.0% of the sample) and Time 2 (46.9%). Very few participants were characterized as “Not-adherent, Not Responsible” at Time 1 and Time 2 (4.2% and 3.7%, respectively). The percentage of participants who were “Adherent, Not Responsible” decreased from Time 1 to Time 2 (37.0% to 29.6%), but the percentage of participants who were “Not Adherent, Responsible” increased from Time 1 to Time 2

(9.9% to 19.8%). Analyses of the McNemar-Bowker Test (i.e., a test for paired, categorical variables) revealed that Adherent/Responsibility groups for bowel management and catheterization did not significantly change from Time 1 to Time 2 ($p > .05$).

Demographic information by group, including average age, SES, and percent female, are also provided in Tables 3 and 4. Multinomial logistic regressions were conducted to determine if the groups differed based on age or SES, using Group 1 (“Adherent/Responsible”) as the reference group. For bowel management at Time 1, participants who fell in Group 2 ($\chi^2(1) = -.53, p < .01$) and Group 4 ($\chi^2(1) = -.55, p < .01$) i.e. “Not Responsible” groups) were younger. At Time 2, participants in Group 2 ($\chi^2(1) = -.29, p < .05$) and Group 4 ($\chi^2(1) = -.36, p < .05$) for bowel management were also younger. Regarding catheterization at Time 1, participants who fell in Group 2 ($\chi^2(1) = -.22, p < .05$) were younger. Similarly, at Time 2, participants in Group 2 were younger ($\chi^2(1) = -.30, p < .05$). At Time 1, participants who fell in Group 2 for catheterization had lower SES ($\chi^2(1) = -.03, p < .05$). At Time 2, participants who fell in Group 2 for bowel management had lower SES ($\chi^2(1) = -.04, p < .05$).

Chi-square tests were conducted to determine if gender differed with respect to group membership at Time 1 and Time 2 (see Tables 3 and 4 for % female by group). Gender differed for bowel management groups at Time 2 only, $\chi^2(3) = 10.55, p < .05$. Further analysis of frequencies by group showed a higher percentage of females in Group 3 (“Non-adherent, Responsible”) for bowel management at Time 2 (100% female; 12 out of 12 participants that fell in this group were female). Catheterization and bowel

management groups did not differ by gender at Time 1, and catheterization groups did not differ by gender at Time 2.

Table 3. Evaluating Frequencies, Percentages, and Demographic Descriptive Information for Medical Adherence and Responsibility with Bowel Program at Time 1 and Time 2

<u>Time 1</u>					
Bowel Program Groups	<i>N</i>	%	Age <i>M (SD)</i>	% Female	SES <i>M (SD)</i>
1. Adherent/Responsible	10	8.5	13.40 (1.65)	70.00	34.95 (20.17)
2. Adherent/Not Responsible	59	50.0	10.98 (2.33)*	50.88	38.44 (16.25)
3. Non-adherent/Responsible	9	7.6	13.00 (2.12)	77.78	42.72 (11.49)
4. Non-adherent/Not Responsible	40	28.2	10.88 (2.41)*	52.50	41.74 (15.52)
Total	118	100.0	11.31 (2.42)	54.31	39.64 (16.01)
<u>Time 2</u>					
Bowel Program Groups	<i>N</i>	%	Age <i>M (SD)</i>	% Female	SES <i>M (SD)</i>
1. Adherent/Responsible	21	21.0	14.47 (2.04)	55.00	47.00 (12.25)
2. Adherent/Not Responsible	41	41.0	13.03 (2.17)*	46.15	37.63
3. Non-adherent/Responsible	12	8.5	14.18 (2.56)	100.00	(17.56)*
4. Non-adherent/Not Responsible	26	18.3	12.65 (2.48)*	50.00	37.86 (12.80)
					37.81 (16.19)
Total	100	100.0	13.33 (2.36)	55.21	39.61 (15.98)

Note. The McNemar-Bowker Test showed that Time 1 and Time 2 group membership did not significantly differ ($p > .05$). The Chi-Square Test showed that there was a significant relationship between gender and group membership at Time 2 ($p < .05$). For continuous demographic variables, * = statistically different than the reference group (Group 1).

Table 4. Evaluating Frequencies, Percentages, and Demographic Descriptive Information for Medical Adherence and Responsibility with Catheterization at Time 1 and Time 2

<u>Time 1</u>					
Catheterization Groups	<i>N</i>	%	Age <i>M (SD)</i>	% Female	SES <i>M (SD)</i>
1. Adherent/Responsible	43	43.0	11.95 (2.32)	58.14	42.50 (15.13)
2. Adherent/Not Responsible	37	37.0	10.76 (2.35)*	59.46	34.89 (15.46)*
3. Non-adherent/Responsible	14	9.9	11.29 (2.34)	61.54	45.00 (15.59)
4. Non-adherent/Not Responsible	6	4.2	11.33 (3.01)	50.00	41.75 (23.45)
Total	100	100.0	11.37 (2.40)	58.59	39.96 (16.13)
<u>Time 2</u>					
Catheterization Groups	<i>N</i>	%	Age <i>M (SD)</i>	% Female	SES <i>M (SD)</i>
1. Adherent/Responsible	38	46.9	13.92 (2.32)	57.89	40.95 (15.15)
2. Adherent/Not Responsible	24	29.6	12.32 (2.46)*	72.72	34.06 (18.78)
3. Non-adherent/Responsible	16	19.8	13.23 (2.20)	53.33	47.53 (9.88)
4. Non-adherent/Not Responsible	3	3.7	10.00 (0.00) ^a	33.33	35.17 (17.96) ^a
Total	81	100.0	13.18 (2.45)	60.26	39.90 (16.07)

Note. The McNemar-Bowker Test showed that Time 1 and Time 2 group membership did not significantly differ ($p > .05$). The Chi-Square Test showed that there was not a significant relationship between gender and group membership at Time 1 or Time 2. For continuous demographic variables, * = statistically different than the reference group (Group 1); ^a = not tested for statistical significance due to Group 4 $n < 5$.

Hypothesis Testing

Multinomial logistic regressions were utilized to explore the main study hypotheses. For all analyses, we ran the analyses with Time 1 predictors (biological, neuropsychological, family, or peer) predicting Time 1 or Time 2 medical adherence/responsibility groups (i.e., four groups, see Figure 1). We utilized group 1, “Adherent, Responsible” as the reference category for all analyses. A power analysis was

used to assess whether the sample size was appropriate for the following statistical analyses (Aiken & West, 1991; Cohen, 1992). Power was computed based on the fewest number of children who had catheterization or bowel program management data at Time 2 (i.e., $n = 81$ for catheterization at Time 2). Assuming a power of .80, an alpha of .05, and an estimated R^2 of .15 (a medium effect size), a sample of 91 is required for the most complex analyses (5 independent variables for family analyses, Cohen, 1992). Therefore, the current study has enough power to detect a medium effect size for analyses of neuropsychological, family, and peer predictors of medical adherence and responsibility for catheterization and bowel programs at Time 1, but only large effects for analyses with biological variables. We had enough power to detect a medium effect size for most bowel program analyses at Time 2, but only large effects for catheterization analyses and select bowel program analyses at Time 2 (e.g., peer and biological variables). Due to limited power, we did not control for Time 1 medical adherence and responsibility variables for Time 2 analyses. Furthermore, we did not evaluate group 4 at Time 2 (i.e., “Not Adherent, Not Responsible”) for catheterization management due to low N (i.e., less than 5 participants).

Hypothesis 1 (Biological)

We expected that children with more severe SB at Time 1 would be more likely to fall in the “Adherent, Child Not Responsible” category (i.e. Group 2, see Figure 1), compared to the “Adherent, Child Responsible” category (i.e., Group 1) at Time 1 and Time 2. In other words, parents of children with more severe SB would be more likely to maintain responsibility for SB medical management, as well as successfully adhere to the SB regimen. We also expected that SB severity would differentiate Group 1 from Group

4 (“Non-Adherent, Child Not Responsible”). That is, for a subset of families, we hypothesized that families would struggle to adhere to treatments for more severe SB. We did not expect that SB severity would significantly differentiate Group 3 (“Not Adherent, Child Responsible”) from Group 1.

Concurrent, Bowel Management

The overall model of biological variables (i.e., lesion level, number of shunt revisions, and gross motor functioning) predicting group membership at Time 1 (see Figure 1) was non-significant $\chi^2(9) = 3.84, p > .05$. Measures of lesion level, number of shunt revisions, and gross motor functioning did not significantly predict group membership at Time 1 (p 's $> .05$).

Longitudinal, Bowel Management

Due to listwise deletion, the number of participants at Time 2 decreased when the shunt revisions variable was included in analyses of bowel program management ($n = 62$ when included shunt revisions, $n = 82$ when excluded shunt revisions). Thus, the lowered n , combined with limited significance of the shunt revision variable, prompted a decision to exclude the shunt variable from the reported analyses. The overall model of biological variables predicting group membership at Time 2 (see Figure 1) was non-significant $\chi^2(6) = 8.66, p > .05$. Despite the overall non-significance of the model, gross motor functioning significantly predicted whether a child was “Adherent, Not Responsible” or “Adherent, Responsible” to bowel programs, $b = .83$, Wald $\chi^2(1) = 5.01, p < .05$ (see Table 6). The odds-ratio indicated that as gross motor functioning limitations increased by one unit, the odds of being “Adherent, Not Responsible” (rather than “Adherent, Responsible”) increased by 2.28 units. Further exploration of gross motor group means

revealed that the mean number of gross functioning classification for youth in Group 1 (i.e., “Adherent, Responsible”) was 2.19, as opposed to 2.97 in Group 2 (i.e., “Adherent, Not Responsible”; see Figure 3). That is, youth with greater gross motor functioning limitations at Time 1 were less likely to be responsible for their bowel program at Time 2 (but maintained high levels of adherence). Lesion level did not significantly predict group membership ($p > .05$).

Table 5. Significant Concurrent Multinomial Logistic Regression Analyses: Biological, Neuropsychological, Family, and Peer Variables as Predictors of Medical Adherence and Responsibility with Bowel Program and Catheterization

Variables	Group 2: Adherent/Not Responsible			Group 3: Non- adherent/ Responsible			Group 4: Non-Adherent/Not Responsible		
Time 1 Bowel Management	<i>b</i>	<i>Wald</i>	<i>OR</i>	<i>b</i>	<i>Wald</i>	<i>OR</i>	<i>b</i>	<i>Wald</i>	<i>OR</i>
<u>Neuro (N = 114)</u> BRIEF	2.85	3.43	17.32^a	<i>ns</i>	<i>ns</i>	<i>ns</i>	4.99	9.10	145.58[*]
<u>Family (N = 110)</u> Family Cohesion	2.75	4.13	15.64[*]	3.67	4.15	39.19[*]	<i>ns</i>	<i>ns</i>	<i>ns</i>
Family Stress (SQRT)	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	4.81	4.54	122.34[*]
<u>Peer (N = 100)</u> Peer Conflict (LOG)	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	10.8 2	3.64	49821.8 7^a
Time 1 Catheterization	<i>b</i>	<i>Wald</i>	<i>OR</i>	<i>b</i>	<i>Wald</i>	<i>OR</i>	<i>b</i>	<i>Wald</i>	<i>OR</i>
<u>Biological (N = 66)</u> Lesion Level (SQRT)	1.43	4.07	4.16[*]	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>
<u>Neuro (N = 97)</u> BRIEF	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	.08	3.67	1.08^a

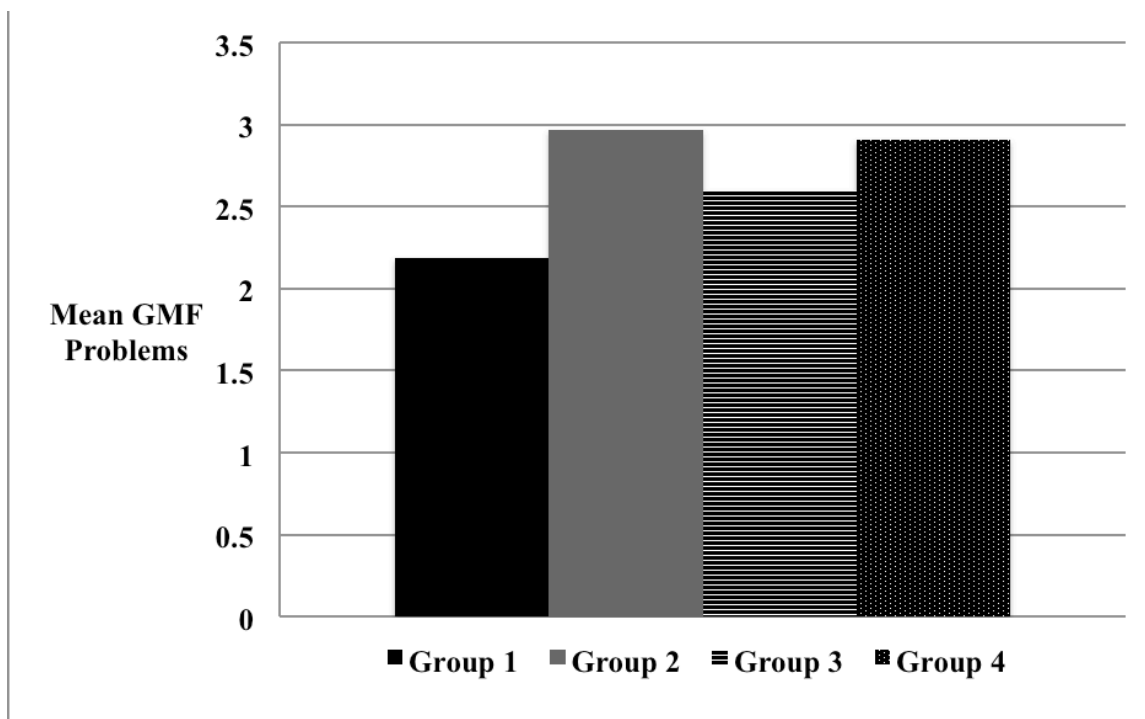
Notes. * = $p < .05$; ** = $p < .01$; ^a = approached significance at $p = .05$; Group 1 (Adherent and Responsible) is the reference group; OR = Odds Ratio; ns = non-significant; Marginally-significant findings included in table; conducted on variable; SQRT = square-root transformation conducted on variable; LOG = logarithm transformation conducted on variable; Large OR observed for transformed variables (SQRT and LOG).

Table 6. Significant Longitudinal Multinomial Logistic Regression Analyses: Biological, Neuropsychological, Family, and Peer Variables as Predictors of Medical Adherence and Responsibility with Bowel Program and Catheterization

Variables	Group 2: Adherent/Not Responsible			Group 3: Non- Adherent/Responsible			Group 4: Non-Adherent/Not Responsible		
	<i>b</i>	<i>Wald</i>	<i>OR</i>	<i>b</i>	<i>Wald</i>	<i>OR</i>	<i>b</i>	<i>Wald</i>	<i>OR</i>
Time 2 Bowel Management									
<u>Biological (N =82)</u>									
GMF	.83	5.01	2.28*	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>
<u>Neuro (N =92)</u>									
IQ	-.06	6.64	.94*	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>
BRIEF	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	3.14	6.25	23.20*
<u>Family (N = 91)</u>									
Family Stress (SQRT)	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	5.05	4.66	55.38*
<u>Peer (N =82)</u>									
Peer Conflict (LOG)	7.48	4.36	1778.14*	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>	<i>ns</i>
Time 2 Catheterization									
<u>Biological (N =67)</u>									
GMF	.75	4.41	2.12*	<i>ns</i>	<i>ns</i>	<i>ns</i>	--	--	--
<u>Family (N = 70)</u>									
Parent-reported Family Conflict (SQRT)	- 3.82	4.20	.02*	<i>ns</i>	<i>ns</i>	<i>ns</i>	--	--	--
Observed Family Cohesion	<i>ns</i>	<i>ns</i>	<i>ns</i>	3.04	4.27	20.88*	--	--	--

Notes. * = $p < .05$; ** = $p < .01$; ^a = approached significance at $p = .05$; Group 1 (Adherent and Responsible) is the reference group; OR = Odds Ratio; ns = non-significant; GMF = Gross motor functioning; *p < .05; Marginally-significant findings included in table; LOG = logarithm transformation conducted on variable; SQRT = square-root transformation conducted on variable; No comparisons done between Group 1 and Group 4 for catheterization due to low N; Large OR observed for transformed variables (SQRT and LOG).

Figure 3. Mean Gross Motor Functioning Classification Scale by Bowel Management Adherence and Responsibility Groups at Time 2



Notes. Group 1 = Adherent/Child Responsible; Group 2 = Adherent/Child Not Responsible; Group 3 = Non-Adherent/Child Responsible; Group 4 = Non-Adherent/Child Not Responsible; GMR = Gross motor functioning; $p < .05$ for Group 1 vs. Group 2 for bowel program; Higher scores indicate higher GMF problems.

Concurrent, Catheterization

The overall model of biological variables predicting medical adherence and responsibility groups at Time 1 was significant, $\chi^2(9) = 24.18, p < .01$. Lesion level significantly related to whether a child was “Adherent, Not Responsible” or “Adherent, Responsible” to catheterization at Time 1, $b = 1.43$, Wald $\chi^2(1) = 4.06, p < .05$. The odds-ratio indicated that as lesion level increased by one unit, the odds of being “Adherent, Not Responsible” (rather than “Adherent, Responsible”) increased by 4.16 units (see Table 5). Further exploration of group means of lesion level revealed $M = 2.50$ for Group 1 (i.e., “Adherent, Responsible”) compared to $M = 3.07$ for Group 2 (i.e.,

“Adherent, Not Responsible”). This finding suggests that youth with higher spinal lesions (and thus, higher SB severity) were less likely to be responsible for their catheterization at Time 1 (but had high levels of adherence). The frequency of shunt revisions and gross motor functioning problems did not significantly predict group membership (p 's > .05).

Longitudinal, Catheterization

Due to listwise deletion, the number of participants at Time 2 decreased when the shunt revisions variable was included in analyses of catheterization ($n = 51$ when included shunt revisions, $n = 69$ when excluded shunt revisions). Thus, the lowered n , combined with limited significance of the shunt revision variable, prompted a decision to exclude the shunt variable from the reported analyses. The overall model of biological variables predicting medical adherence and responsibility groups at Time 2 was non-significant, $\chi^2(4) = 6.28, p > .05$. Similar to our finding with bowel program management, gross motor functioning significantly predicted whether a child was Adherent, Not Responsible or “Adherent, Responsible” to catheterization at Time 2, $b = 0.75$, Wald $\chi^2(1) = 4.41, p < .05$ (see Table 6). The odds-ratio indicated that as gross motor classification increased by one unit, the odds of being “Adherent, Not Responsible” (rather than “Adherent, Responsible”) increased by 2.12 units. That is, youth with greater limitations in gross motor functioning were more likely to be “Adherent, Not Responsible” than “Adherent, Responsible” for their medical regimen. Indeed, further exploration of gross motor means revealed lower gross motor problems for Group 1 ($M = 2.50$; i.e., “Adherent, Responsible”) compared to Group 2 ($M = 3.29$; i.e., “Adherent, Not Responsible”).

Hypothesis 2 (Neuropsychological)

In neuropsychological domains, we expected that poorer executive functioning (as measured by a questionnaire and test data) and lower IQ would distinguish individuals in the nonadherent categories (i.e., “Non-Adherent, Child Responsible” and “Non-Adherent, Child Not Responsible”, Groups 3 and 4 respectively, see Figure 1) from youth in Group 1 at Time 1 and Time 2. We also expected that neuropsychological functioning would differentiate participants in Group 2 (i.e., “Adherent, Child Not Responsible”) from Group 1. That is, for some families, parents who recognized child neuropsychological deficits would be more likely to maintain medical responsibility, as well as successfully follow the medical regimen.

Concurrent, Bowel Management

The overall model of neuropsychological variables (i.e., IQ, BRIEF, and performance-based executive functions) predicting group membership at Time 1 (see Figure 1) was significant, $\chi^2(9) = 26.68, p < .01$. Problems with executive functions (as measured by the BRIEF) significantly predicted whether a child was “Non-Adherent, Not Responsible” or “Adherent, Responsible” to bowel programs, $b = 4.99$, Wald $\chi^2(1) = 9.10, p < .01$ (see Table 5). The odds-ratio indicated that as problem scores on the BRIEF increased by one unit (with higher scores representing higher executive dysfunction), the odds of being “Non-Adherent, Not Responsible” (rather than “Adherent, Responsible”) increased by 147.58 units. That is, youth with more executive dysfunction were less likely to be responsible for their bowel program (although, adherence was high). Indeed, further examination of group BRIEF scores revealed fewer executive functioning problems for participants in Group 1 ($M = 1.51$) compared to Group 4 ($M = 1.83$; see

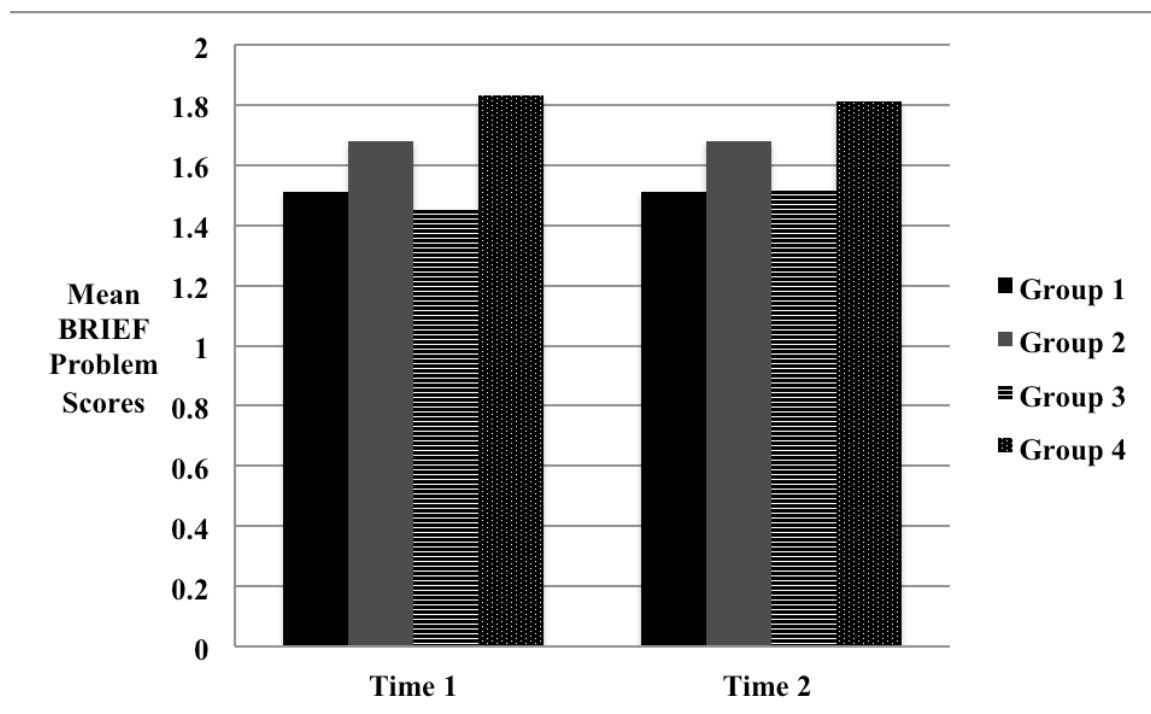
Figure 4). Problems with executive functions (as measured by the BRIEF) was a marginally significant predictor of whether a child was “Adherent, Not Responsible” or “Adherent, Responsible” to bowel programs, $b = 2.85$, Wald $\chi^2(1) = 3.43$, $p = .06$.

Longitudinal, Bowel Management

The overall model of neuropsychological variables predicting group membership at Time 2 (see Figure 1) was significant, $\chi^2(9) = 27.11$, $p < .01$. We found that IQ scores significantly predicted whether a child was “Adherent, Not Responsible” or “Adherent, Responsible” to bowel programs, $b = -.06$, Wald $\chi^2(1) = 6.64$, $p < .05$ (see Table 6). The odds-ratio indicated that as IQ increased by one unit, the odds of being “Adherent, Not Responsible” (rather than “Adherent, Responsible”) decreased by .94 units. Further examination of group IQ means revealed a higher mean IQ for participants in Group 1 ($M = 99.70$) compared to Group 2 ($M = 83.89$; see Figure 5). Thus, among families with high levels of medical adherence, youth with higher intellectual functioning at Time 1 were more likely to be responsible for their medical regimen at Time 2.

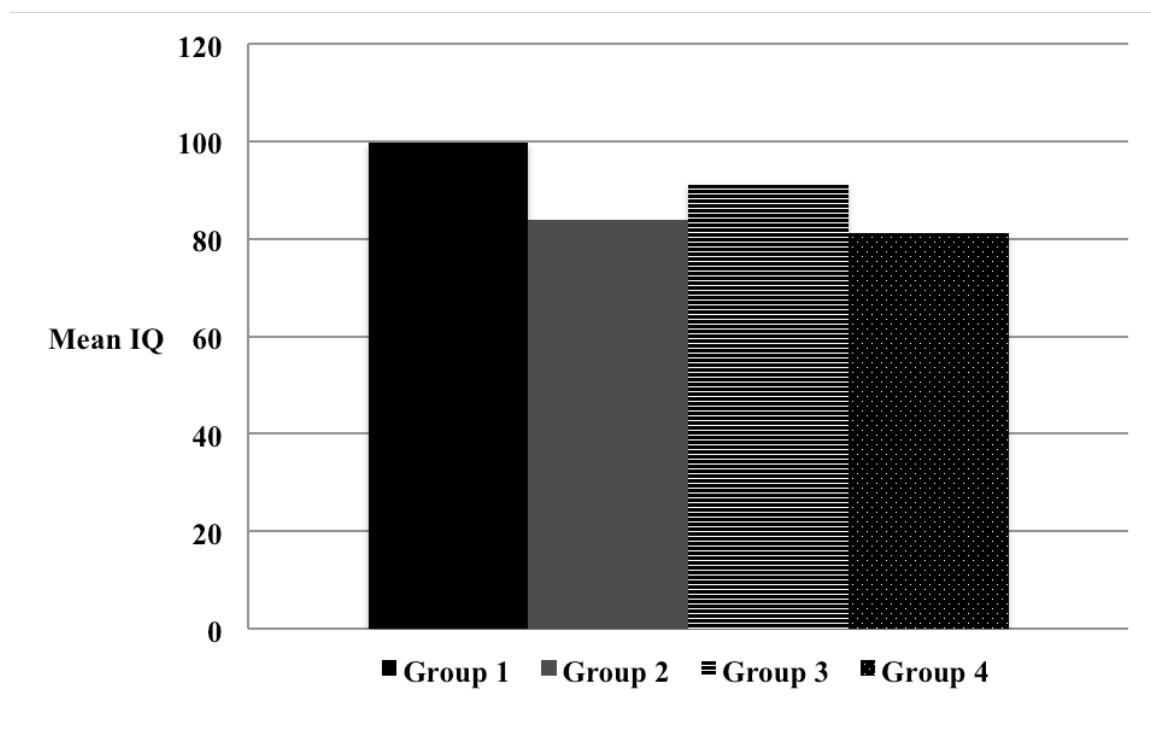
Secondly, we found that BRIEF scores significantly predicted whether a child was “Not Adherent, Not Responsible” or “Adherent, Responsible” with bowel management, $b = 3.14$, Wald $\chi^2(1) = 6.25$, $p < .05$ (see Table 6). The odds-ratio indicated that as total number of problems on the BRIEF increased by one unit, the odds of being “Not Adherent, Not Responsible” (rather than “Adherent, Responsible”) increased by 1.97 units (see Figure 4). This finding suggests that when youth displayed more signs of executive dysfunction at Time 1, bowel adherence and responsibility were compromised at Time 2. Performance-based executive functioning did not predict group membership ($p > .05$).

Figure 4. Mean BRIEF Scores by Bowel Management Adherence and Responsibility Groups at Time 1 and Time 2



Notes. Group 1 = Adherent/Child Responsible; Group 2 = Adherent/Child Not Responsible; Group 3 = Non-Adherent/Child Responsible; Group 4 = Non-Adherent/Child Not Responsible; EF = Executive Function; Higher scores indicate higher executive function problems; $p < .05$ for Group 1 vs. Group 4 at Time 1 and Time 2; $p = .06$ for Group 1 vs. Group 2 at Time 1.

Figure 5. Mean IQ by Bowel Management Adherence and Responsibility Groups at Time 2



Notes. Group 1 = Adherent/Child Responsible; Group 2 = Adherent/Child Not Responsible; Group 3 = Non-Adherent/Child Responsible; Group 4 = Non-Adherent/Child Not Responsible; $p < .05$ for Group 1 vs. Group 2.

Concurrent, Catheterization

The overall model of neuropsychological variables (i.e., IQ, BRIEF, and performance-based executive functions) predicting group membership at Time 1 (see Figure 1) was significant, $\chi^2(9) = 20.68, p < .05$. We found that IQ scores marginally predicted whether a child was “Non-Adherent, Not Responsible” or “Adherent, Responsible” to bowel programs, $b = .08$, Wald $\chi^2(1) = 3.67, p = .06$ (see Table 5).

Longitudinal, Catheterization

The overall model of neuropsychological variables (i.e., IQ, BRIEF, and performance-based executive functions) predicting group membership at Time 2 (see

Figure 1) was non-significant, $\chi^2(6) = 4.05, p > .05$. Measures of IQ and executive functions did not significantly predict group membership (p 's $> .05$).

Hypothesis 3 (Family Dynamics)

Socially, we predicted that youth with more family conflict, less family cohesion, and more family stress would be more likely to fall in the “nonadherent” groups (i.e., “Non-Adherent, Child Responsible” and “Non-Adherent, Child Not Responsible”, Groups 3 and 4 respectively, see Figure 1) compared to Group 1 at Time 1 and Time 2. We did not expect family variables to significantly differentiate Group 2 from Group 1, as we hypothesized high levels of family functioning in both of these groups.

Concurrent, Bowel Management

The overall model of family variables (i.e., family conflict, cohesion, and stress) predicting group membership at Time 1 (see Figure 1) was statistically significant, $\chi^2(18) = 29.79, p < .05$. The questionnaire measure of family cohesion significantly predicted whether a child was “Adherent, Not Responsible” or “Adherent, Responsible” to bowel programs at Time 1, $b = 2.67$, Wald $\chi^2(1) = 3.84, p < .05$ (see Table 5). The odds-ratio indicated that as family cohesion increased by one unit, the odds of being “Adherent, Not Responsible” (rather than “Adherent, Responsible”) increased by 14.48 units. That is, parents who reported high levels of family cohesion were more likely to maintain responsibility for their child’s bowel program and demonstrate high levels of adherence. Indeed, we found higher mean family cohesion for participants in Group 2 ($M = 3.17$) compared to Group 1 ($M = 3.01$).

Interestingly, the questionnaire measure of family cohesion also significantly predicted whether a child was “Not Adherent, Responsible” or “Adherent, Responsible”

to bowel programs, $b = 3.65$, Wald $\chi^2(1) = 4.18$, $p < .05$ (see Table 5). The odds-ratio indicated that as family cohesion increased by one unit, the odds of being “Not Adherent, Responsible” (rather than “Adherent, Responsible”) increased by 38.31 units. Further examination of conflict means revealed higher family cohesion for participants in Group 3 ($M = 3.23$) compared to Group 1 ($M = 3.01$). Counter-intuitively, this finding suggests that parents perceived higher levels of family cohesion when their child was non-adherent, but responsible for their bowel program.

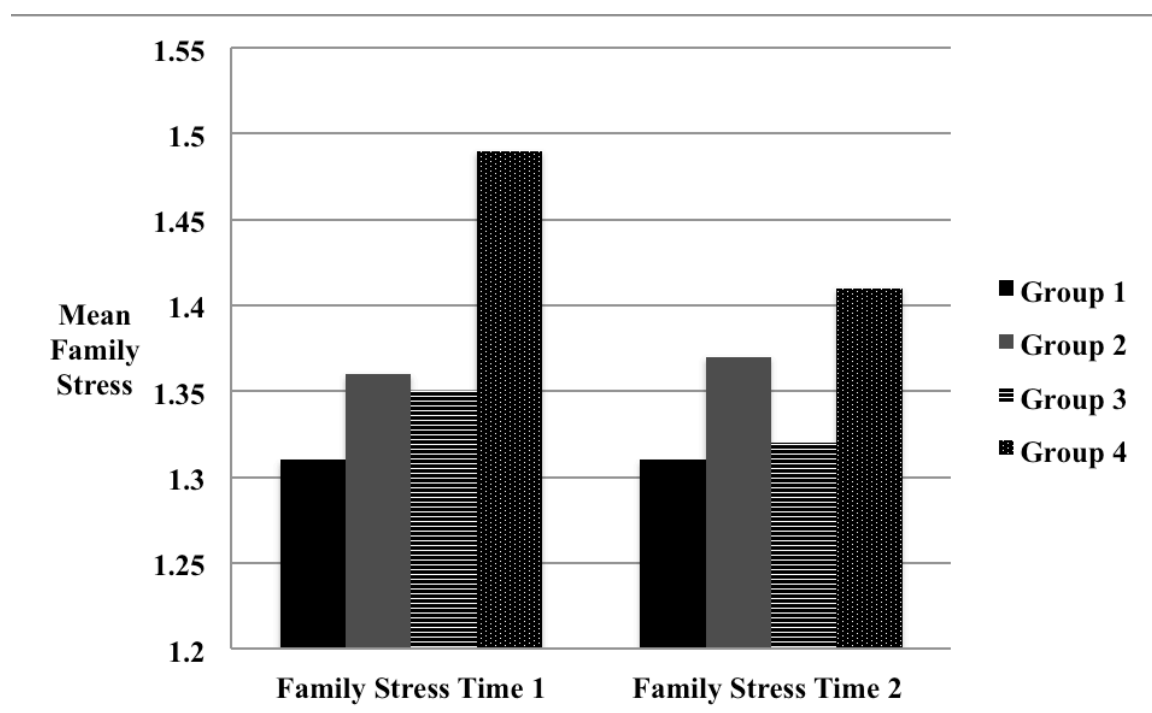
Finally, family stress significantly predicted whether a child was “Not Adherent, Not Responsible” or “Adherent, Responsible” to bowel programs, $b = 4.73$, Wald $\chi^2(1) = 4.38$, $p < .05$. The odds-ratio indicated that as family stress increased by one unit, the odds of being “Not Adherent, Not Responsible” (rather than “Adherent, Responsible”) increased by 113.70 units (with the high odds-ratio reflecting the transformation on the skewed family stress variable). Analysis of family stress means revealed higher family stress for participants in Group 4 ($M = 1.49$) compared to Group 1 ($M = 1.31$; see Figure 6). That is, families who reported higher stress levels also reported concurrent problems with bowel adherence and lower levels of child medical responsibility. Measures of family conflict and the observational measure of family cohesion did not significant predict group membership (p 's $> .05$).

Longitudinal, Bowel Management

The overall model of family variables (i.e., family conflict, cohesion, and stress) predicting group membership (see Figure 1) was not statistically significant, $\chi^2(18) = 26.07$, $p > .05$. Similar to our concurrent finding at Time 1, family stress significant predicted whether a child was “Not Adherent, Not Responsible” or “Adherent,

Responsible” to bowel programs at Time 2, $b = 5.05$, Wald $\chi^2(1) = 4.66$, $p < .05$ (see Table 6). The odds-ratio indicated that as family stress increased by one unit, the odds of being “Not Adherent, Not Responsible” (rather than “Adherent, Responsible”) increased by 155.76 units (with the high odds-ratio reflecting a transformed variable). Analysis of family stress means revealed higher family stress for participants in Group 4 ($M = 1.42$) compared to Group 1 ($M = 1.31$; see Figure 6). This finding suggests that when parents reported high levels of family stress at Time 1, they were more likely to be non-adherent to bowel recommendations and continue to maintain responsibility for their child’s bowel program at Time 2.

Figure 6. Mean Family Stress (Square-Root Transformation) by Bowel Management Adherence and Responsibility Groups at Time 1 and Time 2



Notes. Group 1 = Adherent/Child Responsible; Group 2 = Adherent/Child Not Responsible; Group 3 = Non-Adherent/Child Responsible; Group 4 = Non-Adherent/Child Not Responsible; $p < .05$ for Group 1 vs. Group 4 at Time 1 and Time 2.

Concurrent Catheterization

The overall model of family variables (i.e., family conflict, cohesion, and stress) predicting group membership at Time 1 (see Figure 1) was non-significant, $\chi^2 (18) = 12.85, p > .05$. Measures of family conflict, cohesion, and stress did not significantly relate to group membership (p 's $> .05$).

Longitudinal, Catheterization

The overall model of family variables (i.e., family conflict, cohesion, and stress) predicting group membership (see Figure 1) was non-significant, $\chi^2 (12) = 18.57, p > .05$. Parent-reported family conflict predicted whether a child was “Adherent, Not Responsible” or “Adherent, Responsible” to catheterization, $b = -3.82$, Wald $\chi^2 (1) = 4.20, p < .05$ (see Table 6). The odds-ratio indicated that as family conflict increased by one unit, the odds of being “Adherent, Not Responsible” (rather than “Adherent, Responsible”) decreased by .02 units. Analysis of family conflict means revealed higher family conflict participants in Group 1 ($M = 1.30$) compared to Group 2 ($M = 1.22$). That is, when parents reported higher levels of family conflict at Time 1, children were more likely to be responsible and adherent with their catheterization at Time 2 (as opposed to not responsible and adherent).

Additionally, the observational measure of family cohesion predicted whether a child was “Non-adherent, Responsible” or “Adherent, Responsible” to catheterization, $b = -3.82$, Wald $\chi^2 (1) = 4.20, p < .05$ (see Table 6). Counter-intuitively, the odds-ratio showed that as observed family cohesion increased by one unit, the odds of being “Non-Adherent, Responsible” also increased by 20.88 units. Further analysis of group means revealed higher levels of family cohesion in the “Non-Adherent, Responsible” group (M

= 3.62) compared to the “Adherent, Responsible” group ($M = 3.39$). Thus, among children who were responsible for catheterization at Time 2, higher observed levels of cohesion when the child was non-adherent to catheterization (as opposed to adherent). Child-reported family conflict, the observational measure of family conflict, and family cohesion did not significant predict group membership (p 's > .05).

Hypothesis 4 (Social Adjustment)

We hypothesized that poorer friendship quality, lower peer emotional support, and higher levels of peer conflict would differentiate individuals in the “nonadherent” groups (i.e., “Non-Adherent, Child Responsible” and “Non-Adherent, Child Not Responsible”, Groups 3 and 4 respectively, see Figure 1) from youth in Group 1, at Time 1 and 2. That is, youth who were responsible and adherent to their medical regimens would display higher levels of social adjustment than youth with poor adherence. In addition, we expected that levels of social adjustment would differentiate children in Group 2 (i.e., “Adherent, Child Not Responsible) compared to Group 1. Although adherence is high in both groups, we expected that children who struggle socially would also demonstrate difficulties with medical autonomy development.

Concurrent, Bowel Management

The overall model of social adjustment variables (i.e., friendship quality, peer conflict, and emotional support from peers) predicting group membership (see Figure 1) was non-significant, $\chi^2 (9) = 13.03, p > .05$. However, peer conflict was marginally predicted whether a child was “Non-Adherent, Not Responsible” or “Adherent, Responsible” to bowel programs, $b = 10.82$, Wald $\chi^2 (1) = 3.64, p = .06$ (see Table 5).

Longitudinal, Bowel Management

The overall model of social adjustment variables predicting group membership (see Figure 1) was non-significant, $\chi^2 (9) = 15.39, p > .05$. However, the observational measure of peer conflict significantly predicted whether a child was “Adherent, Not Responsible” (i.e., Group 2) or “Adherent, Responsible” (i.e., Group 1), $b = 7.48$, Wald $\chi^2 (1) = 4.36, p = < .05$ (see Table 6). The odds-ratio indicated that as peer conflict increased by one unit, the odds of being “Adherent, Not Responsible” (rather than “Adherent, Responsible”) increased by 1778.14 units (with the large odds-ratio reflecting the transformation on the observational peer conflict measure). Further examination of peer conflict means revealed higher mean conflict for participants in Group 2 ($M = .29$) compared to Group 1 ($M = 0.23$; See Figure 6). Thus, among youth with high levels of medical adherence, youth who were not responsible for their bowel program were more likely to report higher levels of peer conflict. Friendship quality and emotional support did not significantly predict group membership (p 's $> .05$).

Concurrent, Catheterization

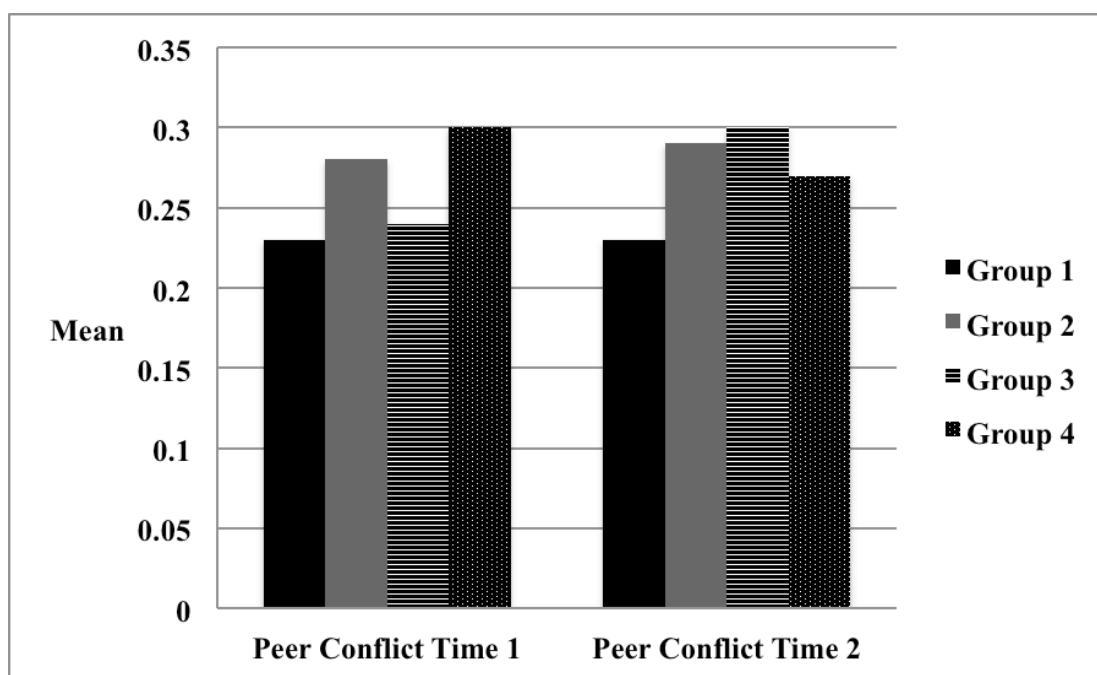
The overall model of social adjustment variables (i.e., friendship quality, peer conflict, and emotional support from peers) predicting group membership at Time 1 (see Figure 1) was non-significant, $\chi^2 (9) = 7.08, p > .05$. Friendship quality, peer conflict, and emotional support from peers did not significantly predict group membership (p 's $> .05$).

Longitudinal, Catheterization

The overall model of social adjustment variables predicting group membership (see Figure 1) was non-significant at Time 2, $\chi^2 (6) = 2.39, p > .05$. Friendship quality,

peer conflict, and emotional support from peers did not significantly predict group membership (p 's > .05).

Figure 7. Mean Peer Conflict (Observational Measure; Log Transformation) by Bowel Management Adherence and Responsibility Groups at Time 1 and Time 2



Notes. Group 1 = Adherent/Child Responsible; Group 2 = Adherent/Child Not Responsible; Group 3 = Non-Adherent/Child Responsible; Group 4 = Non-Adherent/Child Not Responsible; $p = .06$ for Group 1 vs. Group 4 at Time 1; $p < .05$ for Group 1 vs. Group 2 at Time 2.

Hypothesis 5

We hypothesized that each of the four domains (family, peer, neuropsychological, and biological variables) would account for significant variance in concurrent and longitudinal medical responsibility and adherence. However, given the robust relationship between family functioning and condition management in the pediatric psychology literature (e.g., Hanson, Henggeler, & Burghen, 1987; Lewin et al., 2006; Rapoff, 2011), we expected family functioning to emerge as the strongest predictor group. To address this hypothesis, we evaluated the total amount of variance

explained by the model (i.e., the change in explained variance from the baseline to the final model), and then divided the amount of variance accounted for by the number of predictors in the model (i.e., to control for the fact that more predictors would account for more variance).

At Time 1, the final model including neuropsychological variables significantly related to bowel and catheterization responsibility and adherence (see Table 7). The set of family predictors significantly related to bowel management, while the biological variables significantly related to catheterization. Taken together, the domain that accounted for the most variance in bowel management was the neuropsychological group, followed by the family, peer, and biological domains, respectively. For catheterization, the biological group accounted for the most variance, followed by neuropsychological, peer, and family domains, respectively.

Regarding Time 2, the group of neuropsychological predictors (IQ, BRIEF, and performance-based executive functions) was statistically significant for bowel management (see Table 8). For bowel management, the domain that accounted for the most variance was neuropsychological functioning, followed by the social functioning, family functioning, and biological group, respectively. In terms of catheterization, the biological domain was the strongest, followed by the family, neuropsychological, and peer domains, respectively (although, all of these domains were non-significant in the overall model).

Table 7. Total Amount of Variance Accounted for by each Predictor Group (Divided by the Total Number of Predictors): Catheterization and Bowel Program at Time 1

Domains	Predictors	Variance for Bowel Program	Variance for Catheterization
<u>Biological</u>	Lesion level GMF Number of Shunt Revisions	1.28 ($N = 78$)	8.06^{**} ($N = 66$)
<u>Neuropsychological</u>	IQ BRIEF EF Test Data	8.89^{**} ($N = 114$)	6.89[*] ($N = 97$)
<u>Family Functioning</u>	Family Conflict (P, C, O) Family Stress Family Cohesion	5.92[*] ($N = 110$)	2.17 ($N = 94$)
<u>Social Adjustment</u>	Emotional Support Friendship Quality Peer Conflict	4.34 ($N = 100$)	2.36 ($N = 88$)

Notes. GMF = Gross motor functioning; EF = Executive functions; * $p < .05$, ** $p < .01$ for final model; N = Number of participants.

Table 8. Total Amount of Variance Accounted for by each Predictor Group (Divided by the Total Number of Predictors): Catheterization and Bowel Program at Time 2

Domains	Predictors	Variance for Bowel Program	Variance for Catheterization
<u>Biological</u>	Lesion level GMF	4.33 ($N = 82$)	4.26 ($N = 67$)
<u>Neuropsychological</u>	IQ BRIEF EF Test Data	9.04 ** ($N = 92$)	1.35 ($N = 72$)
<u>Family Functioning</u>	Family Conflict (P, C, O) Family Stress Family Cohesion	4.68 ($N = 91$)	1.62 ($N = 70$)
<u>Social Adjustment</u>	Emotional Support Friendship Quality Peer Conflict	5.13 ($N = 82$)	0.80 ($N = 65$)

Notes. GMF = Gross motor functioning; EF = Executive functions; ** $p < .01$ for final model; N = Number of participants.

Exploratory Analyses

After examining the main hypotheses of this study, we conducted exploratory multinomial regression analyses to determine if certain predictors uniquely related to overall medical adherence or responsibility (across all medical domains). Based on our categorical analyses, we speculated that the biological predictors would be especially salient predictors of medical responsibility, whereas neuropsychological and family variables would relate to both medical adherence and responsibility, and social adjustment would relate to medical adherence only. We utilized continuous biological, neuropsychological, family functioning, or social adjustment variables as predictors, and overall, parent-reported medical adherence or responsibility (i.e., the mean of mother-

and father-reported medical adherence and the mean of mother- and father-reported child responsibility) at Time 1 or Time 2 as outcomes (for a total of 16 multinomial regression analyses). For analyses involving medical responsibility or adherence at Time 2, we controlled for medical responsibility or adherence at Time 1.

Biological

We found that lesion level significantly predicted overall medical adherence at Time 1, $B = .15$, $\beta = .23$, $t [79] = 2.12$, $p < .05$, suggesting that higher level lesions (and thus, higher SB severity) was associated with higher concurrent adherence. After controlling for adherence at Time 1, none of the biological variables (i.e., gross motor functioning problems, lesion level, and number of shunt revisions) were significant predictors of adherence at Time 2 (p 's $> .05$).

In terms of child medical responsibility, regression analyses revealed that lesion level significantly predicted child responsibility with their medical regimen, $B = -.15$, $\beta = -.24$, $t [79] = -2.18$, $p < .05$ at Time 1. This finding suggests that higher spinal lesions were associated with less concurrent child medical responsibility. After controlling for child medical responsibility at Time 1, gross motor functioning classification, lesion level, and number of shunt revisions were not significant predictors of medical responsibility at Time 2 (p 's $> .05$).

Neuropsychological

Regarding medical adherence at Time 1, regression analyses revealed that BRIEF scores predicted overall medical adherence, $B = -.51$, $\beta = -.34$, $t [119] = -3.89$, $p < .001$, suggesting that higher scores on the BRIEF (i.e., higher levels of executive dysfunction) related to poorer adherence. We also found that IQ significantly related to medical

adherence at Time 1, $B = .001$, $\beta = .01$, $t [119] = .05$, $p < .05$. This finding indicates that higher child intellectual functioning was associated with higher levels of medical adherence. Notably, after controlling for adherence at Time 1, regression analyses showed higher scores on the BRIEF (i.e., higher levels of executive dysfunction) also related to poorer levels of medical adherence at Time 2, $B = -.38$, $\beta = -.27$, $t [94] = -2.71$, $p < .01$. Taken together, executive dysfunction (as measured by the BRIEF) was a robust predictor of overall medical adherence at Time 1 and Time 2 (even after controlling for adherence at Time 1).

In terms of medical responsibility, regression analyses showed that higher IQ scores related to higher child responsibility at Time 1, $B = .01$, $\beta = -.27$, $t [119] = -2.7$, $p < .01$. Similarly, IQ remained a significant predictor of child responsibility at Time 2, $B = .003$, $\beta = .16$, $t [94] = 2.22$, $p < .05$. That is, after controlling for medical responsibility at Time 1, higher IQ scores related to higher child responsibility at Time 2. Executive functioning (as measured by the BRIEF and the performance-based measure) did not relate to medical responsibility at Time 1 or Time 2 (p 's $> .05$).

Family Functioning

The parent-reported measure of family conflict significantly related to medical adherence at Time 1, $B = -.58$, $\beta = -.23$, $t [116] = -2.59$, $p < .05$. This finding suggests that higher family conflict related to poorer medical adherence. The remaining family functioning variables (the observational measure of family conflict, child-reported family conflict, the observational and questionnaire measure of family cohesion, and family stress) did not significantly relate to medical adherence at Time 1 (p 's $> .05$). Regarding medical adherence at Time 2, the parent-reported measure of family conflict remained

significant, $B = -.59$, $\beta = -.25$, $t [94] = -2.50$, $p < .01$. Thus, after controlling for medical adherence at Time 1, high levels of family conflict related to poorer medical adherence. We also found that the observational measure of family cohesion significantly related to medical adherence at Time 2, $B = -.25$, $\beta = -.24$, $t [94] = -2.45$, $p < .05$. Interestingly, this finding suggests that high levels of observed family cohesion at Time 1 were associated with poorer adherence at Time 2.

In terms of medical responsibility, none of the family predictors were significantly related to child medical responsibility at Time 1 (p 's $> .05$). Similarly, we did not find significant relationship between family functioning and child medical responsibility at Time 2 (p 's $> .05$).

Social Adjustment

Regarding medical adherence at Time 1, all social adjustment predictors (i.e., the observational measure of peer conflict, friendship quality, and emotional support from peers) were non-significant (p 's $> .05$). Interestingly, after controlling for Time 1 adherence, child emotional support from peers significantly related to medical adherence at Time 2, $B = .18$, $\beta = .27$, $t [82] = 2.69$, $p < .01$. This finding suggests that youth with higher emotional support at Time 1 had higher levels of medical adherence at Time 2. All other predictors were non-significant (p 's $> .05$).

For medical responsibility, we found a significant relationship between friendship quality and medical responsibility, $B = -.13$, $\beta = -.19$, $t [103] = -1.99$, $p < .05$, suggesting that higher friendship quality related to less child medical responsibility. All other predictors were non-significant (p 's $> .05$). Furthermore, none of the social adjustment

variables emerged as significant predictors of child medical responsibility at Time 2 (p 's $> .05$).

CHAPTER FIVE

DISCUSSION

The purpose of this multi-source, multi-method, longitudinal study was to examine biological, neuropsychological, and social (i.e., family dynamics and social adjustment) predictors of medical adherence and responsibility in a pediatric SB sample. This study extended the current literature by testing the utility of a bio-neuropsychosocial model of adjustment (Holmbeck & Devine, 2010) to evaluate the predictive value of four different domains of functioning on the development of medical responsibility and adherence in youth with SB, an underrepresented population in the pediatric self-management literature (Sawin, Betz, & Linroth, 2010). Specifically, this investigation tested the following predictive domains: 1) biological (gross motor functioning, number of shunt revisions, and lesion level), 2) neuropsychological (IQ and executive functioning), 3) family (conflict, cohesion, and stress), and 4) social adjustment (friendship quality, emotional support from peers, and peer conflict). We expected that each of these domains would relate to medical adherence and responsibility in unique ways (see Figure 2) and together, these domains would account for significant variability in concurrent and longitudinal health care behaviors. For all of our main study analyses, we utilized the most optimal level of child functioning (i.e., “Adherent, Child Responsible”, see Figure 1) as the reference group to evaluate differences in bio-neuropsychosocial functioning across medical adherence/responsibility groups (i.e., “Adherent, Child Not Responsible”, “Nonadherent, Child Responsible”, and

“Nonadherent, Child Not Responsible”) at two, biennial study time points. Strengths of this investigation included the use of clinically relevant cut-points for medical responsibility and adherence variables, the inclusion of mother, father, and teacher-reported data, the use of observational measures of family dynamics, and analyzing data across time.

Preliminary analyses showed that the majority of children were characterized as “Adherent, Not Responsible” for their bowel program at Time 1 (i.e., 50.0% of sample) and Time 2 (i.e., 41.0% of the sample; see Table 3), suggesting that many parents continue to be involved in their child’s bowel management across late childhood and early adolescence. Rates of bowel program nonadherence were slightly higher at Time 1 (35.8% of the sample) compared to Time 2 (26.8% of the sample). Interestingly, children appeared to possess much greater responsibility with their catheterization, with the majority of youth characterized as “Adherent, Responsible” at Time 1 (43.0% of the sample) and Time 2 (46.9% of the sample). Compared to bowel program nonadherence, rates of catheterization nonadherence appeared lower (with 14.1% of the sample were characterized as nonadherent at Time 1 and 23.5% of the sample were characterized as nonadherent at Time 2). Further, analyses of descriptive information showed that participants who fell in the “Not Responsible” groups were more likely to be younger than participants who fell in the “Adherent, Responsible” group.

Descriptive data showed that families struggled to adhere to bowel recommendations, even when parents were responsible for disease management. This finding is noteworthy, as ongoing non-adherence to a child’s bowel program may result in bowel incontinence, constipation, pain, and bladder/kidney diseases. On the other

hand, higher rates of child responsibility for catheterization and lower rates of nonadherence likely reflect an earlier transfer of medical responsibility from parent to child (Stepansky et al., 2010). Possibly, higher rates of nonadherence to bowel programs indicate children's early difficulties with managing this aspect of their medical regimen for the first time. Further research should evaluate whether rates of bowel program adherence improve after families have navigated early issues related to transferring these medical responsibilities to children.

Regarding correlational data, we found several significant relationships between continuous medical outcome variables, study predictors, and demographic data (see Table 2). In general, higher levels of medical adherence were associated with more severe SB (e.g., more limitations in gross motor functioning, higher lesion level, and more shunt revisions), which likely reflected a main study finding that increased parental involvement in the regimen for youth was associated with more severe SB. Additionally, higher levels of medical adherence were correlated with higher executive functioning skills, lower family conflict, lower family stress, and higher child friendship quality.

In terms of medical responsibility, we discovered that higher levels of child responsibility with the SB medical regimen were associated with older age, higher executive functioning skills, higher IQ, lower family stress, lower peer conflict, and higher emotional support from peers. Of note, we found counter-intuitive correlations between older age and less peer conflict, but lower friendship quality. Given research on social functioning deficits in youth with SB (e.g., Devine, Holmbeck, Gayes, & Purnell, 2012), we suspect that these findings reflect difficulties with maintaining friendships during adolescence (i.e., adolescents with fewer close friendships would rate lower

friendship quality) and related challenges with social functioning (e.g., more passivity in social interactions; Holmbeck et al., 2003, Blum, Resnick, Nelson & St Germaine, 1991).

Hypothesis 1 (Biological)

We found support for our hypothesis that SB severity would relate to less child responsibility with bowel management and catheterization. Regarding bowel management, results suggested that youth with more limitations in gross motor functioning at Time 1 were less likely to be responsible for their bowel program at Time 2 (compared to the reference group, “Adherent, Child Responsible”), but parents were adherent to bowel recommendations. Additionally, we found that youth with higher spinal lesions were also less likely to be responsible for their catheterization at Time 1 (compared to the reference group), but had similarly high levels of parent-facilitated adherence. Gross motor functioning also significantly differentiated whether a child was “Adherent, Not Responsible” or “Adherent, Responsible” to catheterization at Time 2. That is, youth with more gross motor limitations were more likely to be dependent on parents for catheterization management, but such families demonstrated high levels of adherence.

These findings suggest that the development of medical responsibility for bowel management and catheterization is more challenging for a child with more profound gross motor limitations and higher lesion levels. Youth with more severe SB appeared to rely on family members more for assistance with their medical regimen. Considering that parents showed high levels of adherence in the face of more severe SB, limited child responsibility with catheterization and bowel programs might reflect appropriate decision-making from parents and healthcare teams, who recognize important motor

limitations that may undermine a medical self-management. Contrary to our hypothesis that some families may struggle to manage more complex SB, characteristics of more severe SB did not relate to poor adherence. This finding is encouraging, as treatment complexity is considered an important risk factor for poor adherence (Quittner et al., 2000), and it is likely that children with more severe SB will have more complex treatments to follow (e.g., children with more severe SB may require a more intensive bowel program). This finding aligns with research that suggests that parents may be more diligent about monitoring adherence behaviors for more severe pediatric diseases (Drotar 2000; Reed-Knight, Blount, & Gilleland, 2014). Notably, parents of youth with more severe SB demonstrated resilience in their ability to manage more severe disease factors and subsequent medical demands.

Hypothesis 2 (Neuropsychological)

Regarding bowel management, we found support for our hypothesis that youth with neuropsychological challenges were less likely to be responsible for their medical regimen, but parents demonstrated high levels of adherence. Compared to the reference group at Time 1, youth with higher executive dysfunction (as measured higher parent- and teacher-reported problems on the BRIEF) were less likely to be responsible for their bowel program, but parents of such youth tended to be adherent to bowel recommendations. We found further support for this hypothesis for medical outcomes at Time 2. Specifically, youth with lower intellectual functioning at Time 1 were less likely to be responsible for their bowel program at Time 2, but parents in these families tended to demonstrate high levels of bowel program adherence.

This study also found partial support for our hypothesis that neuropsychological deficits at Time 1 would negatively impact bowel program adherence. Higher levels of parent- and teacher-reported executive dysfunction at Time 1 (indexed by more problems on the BRIEF) significantly related to whether a child was “Not Adherent, Not Responsible” or “Adherent, Responsible” with bowel management at Time 2. That is, when youth displayed more signs of executive dysfunction at Time 1, bowel adherence and responsibility were compromised at Time 2. Interestingly, no significant findings emerged for catheterization.

Similar to past research (e.g., Tarazi, Zabel, & Mahone, 2008; O’Hara and Holmbeck, 2013; Friedman et al., 2009), we discovered that the neuropsychological impairments associated with SB (e.g., executive dysfunction and intellectual difficulties) negatively impacted a child’s ability to obtain medical responsibility, particularly with the completion of a bowel program. Study findings showed that executive dysfunction (as measured by the BRIEF) related to less, concurrent child responsibility with bowel management, as well as responsibility with bowel management two years later. Lower IQ at Time 1 also related to less child responsibility with bowel management at Time 2. Thus, our study extends the current body of literature by demonstrating that IQ and executive functioning challenges during late childhood/early adolescence tended to undermine the development of medical responsibility over a two-year period.

Importantly, no significant findings emerged for catheterization at Time 1 or Time 2, which may have reflected lower statistical power in this domain. Another possible consideration is that parents of youth with SB transfer catheterization responsibilities at an earlier age than other medical regimen tasks (Stepansky et al., 2010; Psihogios,

Kolbuck, & Holmbeck 2015). Thus, parents and healthcare professionals may emphasize earlier responsibility with catheterization, but other medical activities (including bowel programs) may be transferred more gradually across adolescence. The early emphasis on transfer of catheterization responsibilities is adaptive to some extent, as children with SB are required to catheterize multiple times throughout the day and independence with catheterization may provide benefits at school and in the community (e.g., spending the night at a friend's house). For medical management tasks that occur less frequently than catheterization (e.g., completion of bowel program once per day), parents may be less likely to transfer responsibilities because of fewer incentives (e.g., a child can wait till he or she gets home from a friend's house to complete the medical task). Other factors may moderate the link between neuropsychological challenges and less child responsibility for bowel management, including disease factors (e.g., limited child mobility) and parent perceptions (e.g., a child is not capable of completing their bowel program).

For a subset of families, child executive dysfunction (measured by the BRIEF) related to suboptimal adherence to bowel programs, even when parents maintained full responsibility for the completion of bowel management tasks. This finding suggests that parents of children with more severe executive dysfunction experienced difficulties managing their child's bowel program. Potentially, a child's symptoms of executive dysfunction (e.g., difficulties with inhibition, shifting, and emotional control) may undermine parent's ability to care for their child (e.g., a child may be oppositional to their bowel program). Another explanation is that parents of youth with more profound executive dysfunction may also have problems in this cognitive domain. Clearly, interventions are needed to support parents around bowel management adherence and the

transfer of bowel management responsibilities to children, particularly when executive functioning and IQ challenges are present.

Hypothesis 3 (Family Dynamics)

In general, our findings did not support our hypothesis that levels of family functioning would be similar in Group 1 and Group 2. In fact, we found that parents who reported high levels of family cohesion were more likely to maintain responsibility for their child's bowel program and demonstrate high levels of adherence compared to the reference group (i.e., "Adherent, Child Responsible"). Interestingly, our hypothesis that family cohesion would relate to better medical adherence was not confirmed. Specifically (and counterintuitively), parents of children who fell in the "Not Adherent, Child Responsible" group reported higher family cohesion than parents of children who fell in the "Adherent, Child Responsible" for bowel program management at Time 1. Similarly, we found that high observed family cohesion at Time 1 related to child non-adherence with catheterization at Time 2. These finding suggests that higher levels of family cohesion coincided with nonadherence to bowel and catheterization recommendations when children were granted primary responsibility for medical tasks.

On the other hand, when parents maintained responsibility for the medical regimen, higher levels of family cohesion related to high levels of adherence. This finding suggests that positive family interactions and involvement in medical tasks is important for bowel program adherence during early adolescence. Ongoing parental support and involvement in disease management appears to alleviate some of the challenges that undermine adherence when children are granted primary responsibility for their bowel program.

The relationship between family cohesion and medical outcomes appeared to be more nuanced than expected, as it did not perfectly coincide with research investigating medical self-management and compliance in other disease populations (e.g., parenting a child with diabetes). Indeed, past research suggests that positive family communication may promote adherence to medical treatment by facilitating effective medical problem-solving and improving an adolescents' mood (Iskander, Rohan, Pendley, Delamater, & Drotar, 2015). This appeared to be true in families where parents retained responsibilities for bowel management, but the opposite was found when children were fully responsible for their bowel management and catheterization (i.e., when children were responsible for their medical regimen, and families were highly cohesive, adherence was poor).

Past research shows that when parents of youth with SB balance emotional support, affection, and approval with age-appropriate expectations and consequences, adherence is maximized (O'Hara & Holmbeck, 2013). Similarly, research in the pediatric diabetes literature suggests that an authoritative parenting style (which involves parental warmth and engagement, as well as limit-setting) is related to higher levels of child adherence with diabetes medical tasks (Anderson, 2004). Possibly, families of youth with SB who report high levels of family cohesion and child responsibility, but non-adherence to treatments, provide appropriate levels of support, but are challenged by setting age-appropriate limitations on behavior. Providing limits to a child with SB may be especially challenging, as the child may already be reluctant to take on a more active role with his or her medical care.

Regarding family conflict, we did not confirm our hypothesis that family conflict would unequivocally relate to non-adherence. Regarding our findings for catheterization,

our analyses revealed that when parents reported higher levels of family conflict at Time 1, children were more likely to be responsible and adherent with their catheterization at Time 2 (as opposed to not responsible and adherent). This finding was surprising given the robust link between high family conflict and poor medical adherence in pediatric populations, including in studies of youth with SB (Psihogios & Holmbeck, 2013; Stepansky et al., 2010). In contrast, our study found that family conflict differentiated child who responsible vs. not responsible for their catheterization (with higher conflict in families of youth who were responsible), but did not relate to adherence. This finding lends support to the point that to some extent, family conflict may promote realignments in medical decision-making responsibilities within the family system. In our sample, family conflict may also be a proxy for continued parental engagement in the medical regimen, even when children are independent with executing medical tasks.

Although a significant body of literature research suggests that highly conflict family environments are detrimental to pediatric medical adherence (e.g., Hanson, Henggeler, & Burghen, 1987; Lewin et al., 2006; Rapoff, 2011), it is important to note that the intensity of conflict levels in our sample was generally in the low range ($M = 1.71$, Range 1.00 – 4.00). That is, “higher” levels of family conflict did not mean “high” levels of family conflict. Furthermore, as this study utilized clinical cut-points for determining “adherent” to catheterization and “nonadherent” to catheterization, it is possible that our study methodology did not capture the more subtle implications of family conflict on medical adherence (which, was significant when looking at adherence as a continuous variable). Finally, despite the longitudinal nature of our statistical method (i.e., investigating Time 1 predictors of Time 2 medical adherence and responsibility), we

were unable to control medical variables at Time 1 due to limited statistical power. Thus, it is likely that higher family conflict did not cause increased medical responsibility and adherence, but rather, higher conflict in families may reflect parental efforts to maintain adherence in the face of increased child responsibility with catheterization.

As predicted, we found support for our hypothesis that parents who reported high levels of family stress were more likely to report low concurrent and longitudinal parental adherence to their child's bowel program, in comparison to families of children who were responsible and adherent for bowel management. Parenting a child with a chronic health condition is a psychologically, socially, and financially taxing experience. Similar to other pediatric populations (e.g., youth who underwent organ transplantation; Fredericks et al., 2007), our study found that family stress was significantly related to poor adherence to bowel programs. Interestingly, family stress also related to less child responsibility with bowel management. The relationship between family stress and self-management behaviors and adherence for youth with SB is not well understood. We suspect that these variables are transactional, with family stress negatively impacting medical adherence and stalling the transfer of medical responsibilities to children, and poor medical adherence and limited child responsibility leading to higher levels of family stress. Evidently, more longitudinal research is needed to understand these relationships, as well as to identify common factors that relate to high stress levels in families of youth with SB (e.g., health disparities, family/marital discord, financial burden, and medical complexity).

These results highlight the unique challenges of parenting a child with SB and the potential for unexpected problems in the transfer of medical responsibilities from parents

to children. Past research suggests that youth with SB tend to be less self-reliant, less likely to make independent decisions, and more passive in family interactions (Holmbeck et al., 2003, Blum, Resnick, Nelson & St Germaine, 1991). Compared to typically developing youth, children with SB lag in autonomy development by approximately two years, and autonomy development is even further delayed in youth from lower SES backgrounds (Devine et al., 2011). These difficulties, combined with the potential impact of parents' perceptions of high levels of child vulnerability (e.g., Holmbeck et al., 2002), may make conversations about adherence and independence more difficult. Research following youth with SB into emerging adulthood is needed to understand whether greater involvement of parents in the medical regimen and other aspects of autonomy development are adaptive for young adults with SB.

Hypothesis 4 (Social Adjustment)

Our research found no support for our hypotheses for this domain, with the exception of one finding that showed that the observational measure of peer conflict at Time 1 significantly related to whether a child was “Adherent, Not Responsible” or “Adherent, Responsible” to bowel management at Time 2. That is, among youth with high levels of medical adherence, youth who were not responsible for their bowel program were more likely to report higher levels of peer conflict. This finding is notable, as most of the existing body of literature on the relationships between adolescent social functioning and medical care has focused exclusively on adherence, but not on the development of medical responsibilities. Our research suggests that youth who struggle to obtain responsibility for their bowel programs also struggle with navigating peer relationships. Importantly, youth who do not obtain responsibility for their medical

management in adolescence may be at-risk for further peer exclusion, rejection, and bullying due to an inability to “keep up” with peers’ growing independence. Quite possibly, the physical and neuropsychological challenges that undermine social functioning in pediatric SB (Lennon et al., 2015) also may affect the obtainment of medical responsibility. For example, youth who struggle with executive dysfunction may be more likely to demonstrate peer conflict in social interactions (e.g., due to difficulties with problem-solving), which may also impact their engagement in developmentally appropriate activities, such as seeking more independence with their medical regimen. Our preliminary finding suggests added importance for more research on social functioning in youth with SB, as social difficulties may have a direct implication for medical outcomes.

Hypothesis 5

To address this hypothesis, we evaluated the total amount of variance explained by the model (i.e., the change in explained variance from the baseline to the final model), and then divided the amount of variance accounted for by the number of predictors in the model (i.e., to control for the fact that more predictors would account for more variance). We found that the neuropsychological domain was the most salient predictor of bowel management at Time 1 and Time 2, which suggests that a child’s neuropsychological functioning most strongly differentiates children who are adherent/non-adherent and responsible/not responsible for their bowel management. As hypothesized, the family functioning domain was also a significant predictor of bowel responsibility and adherence at Time 1 (though, this domain accounted for less variance than neuropsychological predictors). For catheterization, a different pattern emerged. The

biological domain was the strongest predictor of medical management of catheterization at Time 1 and Time 2 (although, the overall effect was non-significant at Time 2). The neuropsychological domain also accounted for significant variability in catheterization management at Time 1, but accounted for less variance than the group of biological variables.

These findings suggests that the biological, neuropsychological, and family domains play unique and influential roles on adherence behaviors and the development of medical responsibilities in youth with SB. Interestingly, different predictor groups appeared to relate more strongly to the development of medical responsibility (e.g., the biological domain), whereas other groups related to both responsibility and adherence (e.g., neuropsychological and family functioning). Additionally, the findings for catheterization and bowel management were not identical.

Importantly, the neuropsychological and family domains emerged as the strongest predictor groups for bowel management. These findings suggest that the neuropsychological profiles of youth with SB (e.g., difficulties with executive functioning and low average intellectual functioning), as well as family dynamics (e.g., family stress, cohesion, and conflict), are important considerations when evaluating families' management of bowel programs during late childhood and adolescence. Difficulties in each of these domains undermined the development of medical responsibility, as well as negatively impacted adherence. These findings suggest that healthcare providers should consider neuropsychological challenges and family functioning as potential barriers to the successful transfer of medical responsibilities to children and adherence during late childhood/early adolescence. In other pediatric

populations with neurological complications, preliminary intervention research demonstrated that a collaborative, family problem-solving intervention was effective for promoting adherence in children with new-onset epilepsy (Modi, Guilfoyle, & Rausch, 2013). Possibly, an intervention such as this, that addresses several barriers to adherence including neuropsychological difficulties (e.g., problem-solving) and family dynamics, may be useful for facilitating optimal medical management of pediatric SB.

For catheterization, biological variables (e.g., higher spinal lesions and greater gross motor deficits) were robust predictors of medical management, particularly in relation the transfer of catheterization responsibilities from parents to children. This finding is notable, as these are non-modifiable barriers to the development of medical responsibility. Regarding general autonomy development, research suggests that young adults with SB are less likely to live independently, go to college, and obtain part- or full-time employment (Bowman, McLone, Grant, Tomita, & Ito, 2001; Cohen, Kasen, Chen, Hartmark, & Gordon, 2003; Zukerman, Devine, & Holmbeck, 2007). Although less research has been devoted to investigating biological variables that relate to the obtainment of emerging adulthood milestones, it is likely that the disease severity is an important predictor of autonomy development in medical and broad independence domains. Clearly, youth with SB who have more severe disease markers remain in clear need of additional, ongoing healthcare interventions and supports (e.g., occupational therapy and assistive technology) to promote independence skills in medical and non-medical domains.

Exploratory Analyses

After examining the main hypotheses of this study, we conducted exploratory multinomial linear regression analyses to determine if certain predictors uniquely related to overall medical adherence or responsibility (across all medical domains). Specifically, the continuous medical adherence variable addressed seven dimensions of the SB medical regimen, including appointment keeping, bowel control program, skin and wound care, exercise, medications, clean intermittent catheterization, and dealing with urinary tract infections. The continuous medical responsibility variable also consisted of several domains: health appointments, communication about SB, medications, general needs and self-care, ambulation, skin care, catheterization, bowel management, and exercise and diet. Exploratory analyses were conducted to determine whether some predictor variables were stronger predictors of medical responsibility, whereas other predictors would more closely related to medical adherence. In testing our primary hypotheses, the categorical analyses could not address this issue because the adherence and responsibility outcomes were evaluated jointly (see Figure 1).

Regarding biological predictors, we found partial support for our hypothesis that biological factors would relate to child responsibility. Specifically, children with higher spinal lesions at Time 1 had less concurrent responsibility with their medical regimen. Contrary to our hypothesis, youth with higher spinal lesions at Time 1 also had high levels of concurrent medical adherence (though, the adherence measure did not assess who was responsible for completing medical tasks). This finding is similar to our results from Hypothesis 1, which showed that ongoing parental involvement in the face of higher disease severity facilitates adherence. However, after controlling for medical

responsibility or adherence at Time 1, none of the biological variables significantly predicted medical outcomes at Time 2.

In the neuropsychological domain, we found that neuropsychological variables were important predictors of medical responsibility and adherence (thus, supporting our hypothesis that neuropsychological factors would relate to both medical domains). Our exploratory linear regression analyses revealed similar findings as our categorical model. Specifically, higher executive dysfunction at Time 1 (as measured by the BRIEF) related to poorer medical adherence at Time 1 and Time 2 (even after controlling for adherence at Time 1). This finding is important, as child executive dysfunction during late childhood/early adolescence appeared to negatively impact concurrent medical adherence, as well as predicted poorer medical adherence two years later. Interestingly, child executive dysfunction did not relate to child responsibility with medical regimen. Given the link between executive dysfunction and non-adherence, it is likely that attention to a child's skills in executive functioning (e.g., planning/organization, working memory, and cognitive flexibility) play important roles in health outcomes during adolescence.

In addition to this finding, exploratory analyses showed that higher IQ scores related to higher medical adherence and responsibility with SB management. That is, youth with higher intellectual skills were more likely to be given responsibility for their medical regimen and demonstrate higher levels of adherence. As expected, it appeared that children with higher intellectual functioning had fewer challenges with achieving responsibility with medical tasks and demonstrating higher levels of medical adherence.

Regarding family functioning, we found support for our hypothesis that family predictors would relate to medical adherence. However, we did not find significance for medical responsibility. We replicated a cross-sectional finding (Psihogios & Holmbeck, 2013) that higher family conflict related to poorer overall medical adherence at Time 1. Importantly, our research extended this literature by demonstrating that family conflict at Time 1 also predicted poorer adherence at Time 2 (even after controlling for adherence at Time 1). We also found that higher levels of observed family cohesion at Time 1 were associated with poorer adherence at Time 2.

Finally, we found modest support for our hypothesis that social adjustment would relate to medical adherence, but not medical responsibilities. Specifically, child-reported emotional support from peers at Time 1 related to higher medical adherence at Time 2. This finding suggests that when youth perceived support from their friendships during early childhood/late adolescence, they tended to have higher medical adherence two years later. For medical responsibility, we found a significant relationship between friendship quality and medical responsibility, suggesting that higher friendship quality related to less child medical responsibility. As older child age related to more medical responsibility (see Table 2), it is likely that children who have more responsibility for their disease are also older and more aware of the differences between themselves and typically developing peers. This new awareness of differences may lead to lowered friendship quality, especially if youth with SB are reluctant to share information about their disease and/or peers are disapproving of perceived differences. All other predictors were non-significant.

Limitations and Future Directions

Although this study included a number of strengths (such as utilizing clinically-relevant cut-points for medical responsibility and adherence variables, the inclusion of mother, father, and teacher-reported data, the use of observational measures of family dynamics, and analyzing data across time), there are several limitations of the current study that should be addressed in future work. First, as is common in pediatric samples, the sample size was small, particularly when evaluating the categorical catheterization variable. This limited the statistical power of the analyses and the likelihood of detected larger effects, as well as impeded our ability to control for relevant confounds (such as child age and family SES).

Second, there were limitations with the characteristics of this sample. Notably, the majority of this sample was Caucasian. Given the higher rates of SB within a Hispanic population (Lary & Edmonds, 1996), there was an increased effort to include Hispanic, Spanish-speaking youth with SB in this study. For instance, recruitment procedures, questionnaires, tasks, and letters to families were translated to Spanish, and Spanish-speaking research assistants recruited and collected data from Spanish-speaking families. These accommodations allowed for higher rates of Hispanics in this study (28%) compared to other studies investigating youth with SB (e.g., Holmbeck et al., 2003). However, 54% of the sample was Caucasian, which limits the generalizability of study findings to other ethnic groups. Additionally, the sample of this study was limited to one illness group. Although there are several advantages to conducting research with a single illness group (e.g., children with different illnesses may not demonstrate the same difficulties; Holmbeck et al., 2003), this methodology limits the degree to which we can

generalize our findings to other chronic illness groups. Finally, this sample included large age range of youth (ages 8-15 at Time 1 and youth ages 10-18 at Time 2), who were at diverse stages of development. While some younger study participants may have just begun to take on medical responsibilities, older participants may have attained medical responsibilities years ago. To partially address this limitation, we provided correlational data (see Table 2), descriptive information about age by group (see Tables 3 and 4), and evaluated whether age differed by adherence/responsibility groups. However, further research is needed to better understand how youth and families manage spina bifida care during specific developmental periods, such as the transition to adolescence. As barriers and facilitators to spina bifida adherence and responsibility may vary by developmental stage, future work should evaluate whether developmental status moderates relationships between salient predictors (e.g., executive functioning) and medical management outcomes.

Third, when investigating longitudinal data with the categorical medical outcomes, we were unable to control for baseline medical adherence and responsibility due to limited statistical power. This limitation was somewhat remediated by exploratory analyses with continuous medical outcomes, which controlled for baseline adherence and responsibility variables. For the categorical analyses, influence of biological, neuropsychological, and social factors on medical adherence and responsibility across time could not be directly determined, as the management of SB medical tasks at Time 1 related to the management of SB medical tasks at Time 2. Without a true, longitudinal model, we cannot rule out the possibility that medical adherence and responsibility may directly influence modifiable variables, such as family dynamics, social functioning, and

impulsivity. For example, families might attempt to be more cohesive and supportive in response to a child's difficulty to independently adhere to their medical regimen.

There were also several limitations regarding the parent-report questionnaire measure of medical adherence. While self-report measures possess key advantages, including being low-cost, minimally burdensome to families, and easy to administer, self-report measures may inflate adherence rates (e.g., Bender et al., 2000) due to social desirability and other factors (Stirratt et al., 2015). Further, this study focused on SB medical tasks that are common for individuals with SB, the measure did not take into account the child's prescribed regimen. As such, families who responded "N/A" for particular medical tasks were removed from the analyses, reducing the sample sizes across analyses. Additionally, this adherence measure does not measure who is responsible for completing medical tasks (i.e., parent, child, or shared responsibility), though, we attempted to lessen the impact of this limitation by simultaneously evaluating medical adherence and responsibility based on data from two questionnaires (see Figure 1). Other methodologies, such as the daily diary method, have been shown to be more precise for evaluating medical adherence (Quittner et al., 2008). Though this methodology has yet to be employed for youth with SB and their families, this strategy may yield a more accurate depiction of medical adherence in this population.

Another major limitation was the assessment of bowel program management in this study. Both the adherence and responsibility questionnaires contained broad questions regarding bowel management, such as whether the child follows diet recommendations and takes enemas, suppositories, and/or stool softeners. Our measurement tools precluded us from assessing adherence and responsibility to more

specific, individualized bowel program recommendations. For example, it is possible that some participants may be asked to deliver enemas via rectum, whereas other participants follow procedures for Malone Antegrade Continence Enemas (MACE) which involves connecting a tube through the umbilicus once per day for enema administration.

Furthermore, despite assessing child responsibility and adherence to diet recommendations, some children may have little control over this aspect of their bowel program (e.g., diet may be based on the food that is available in the household). Future work should examine whether medical outcomes vary based on individualized bowel treatments, including how adolescents and parents discuss and manage specific dietary regimens at home.

Conclusions

Despite study limitations, the results of this study have important implications. Characteristics of more severe SB (e.g., higher spinal lesions and limitations in gross motor functioning) were salient predictors of lower medical responsibility. This suggests that youth with more profound physical disabilities tend to be less autonomous with their medical regimen. Notably, parents appeared to manage more severe SB very well, as parent-facilitated adherence to their child's bowel and catheterization recommendations was high. Despite high levels of adherence, youth who struggle to become autonomous with their medical care are likely the same individuals who will struggle to meet other medical (e.g., successfully transitioning to adult-centered care) and non-medical (e.g., obtaining employment) independence goals. Undoubtedly, youth with SB who have more severe disease markers remain in need of additional, ongoing healthcare interventions and supports to promote independence skills in medical and non-medical domains. Later in

development, these individuals will likely benefit from added support with the transition to adult medical care, as well as access to patient-advocate partnerships (e.g., to help young adults understand social security benefits and insurance) and community-based resources (e.g., centers that educate on disability rights and provide opportunities for skill-development in independent living).

A child's neuropsychological functioning emerged as one of the strongest predictors of medical adherence and responsibility, particularly with bowel management. For some families, a child's neuropsychological deficits were important considerations for the decision to transfer medical responsibilities to youth. Indeed, parents of youth with problems in intellectual and executive functioning skills were less likely to grant responsibility for bowel management to their children. Notably, our exploratory analyses did not find a link between neuropsychological variables and broad medical responsibility (i.e., responsibility across all SB management tasks), which suggests that parents may only consider executive functioning before transferring specific SB medical tasks (e.g., the transfer of bowel responsibilities).

Although some parents were successful in managing medical responsibilities, other parents struggled to adhere to medical recommendations while caring for their neurocognitively complex child. Potentially, a child's symptoms of executive dysfunction (e.g., difficulties with inhibition, shifting, and emotional control) may undermine parent's ability to care for their child (e.g., a child may be oppositional to their bowel program). Another explanation is that caring for the developmental needs of a child with more profound neuropsychological deficits may cause added stress for parents, which we identified as another important risk factor for nonadherence. Clearly, healthcare providers

should attend to the relationship between a child's neuropsychological profile and medical activities in families, as these youth may be at higher risk for non-adherence and slower autonomy development.

In the family domain, we discovered a complex relationship between medical management and family dynamics. Unexpectedly, we found high levels of family cohesion (measured by parent-report and the observational measure) when children were responsible, but non-adherent to treatments. Past research shows that when parents of youth with SB balance emotional support, affection, and approval with age-appropriate expectations and consequences, adherence was maximized (O'Hara & Holmbeck, 2013). We speculate that families of youth with SB, who reported high levels of family cohesion and child responsibility, but non-adherence to treatments, struggled with setting age-appropriate expectations and limitations with the medical regimen. Indeed, although the mean level of parent-child conflict in our sample was in the low range, higher family conflict related to better adherence to catheterization when children were responsible for this task. Thus, family conflict may be a proxy for continued parental involvement in the medical regimen once youth have been granted independence with the execution of medical tasks.

These complex and nuanced findings demonstrate the fine balance that parents must strike between supporting their child, while also setting age-appropriate limits, and engaging in problem-solving discussions about medical responsibilities. Parents of youth with SB may benefit from psychosocial support in developing an authoritative communication style (i.e., balancing warmth and nurturance while encouraging independence and setting age-appropriate limits and expectations on behavior) with their

child around medical adherence and responsibility, as well as receiving training in techniques to support their child's medical autonomy in a developmentally-sensitive manner (e.g., through the use of scaffolding and collaborative problem-solving). S

Importantly, we found support for our hypothesis that parents who reported high levels of family stress were more likely to report low parental adherence to their child's bowel program. Similar to other pediatric populations (e.g., youth who underwent organ transplantation; Fredericks et al., 2007), our study found that family stress was a significant barrier to medical adherence. Notably, we also found that high levels of family stress related to limited child engagement in their bowel regimen. This finding highlights the need for psychological services for parents of youth with SB (e.g., interventions in stress-management and coping). More research is needed to identify common factors that relate to high stress levels in families of youth with SB, such as health disparities, family/marital discord, financial burden, and a child's medical complexity.

Among our main study analyses, we found limited support for the relationship between social adjustment variables and medical adherence and autonomy. In general, our findings were in line with the existing and mixed body of research on the link between social functioning variables and adherence behaviors (see Palladino & Helgeson, 2012, for a review). Among our main study analyses, one finding suggested a positive relation between peer support and adherence, while the other analyses were non-significant. Among youth with high levels of medical adherence, youth who were not responsible for their bowel program were more likely to report higher levels of peer conflict. This finding suggests that youth who struggled to obtain responsibility for their

bowel programs also struggled with navigating peer relationships. Youth who do not obtain responsibility for their medical management in adolescence may be at-risk for further difficulties with peers (e.g., peer rejection) due to an inability to keep pace with peers' growing independence.

Consistent bio-neuropsychosocial model of adjustment for pediatric SB (Holmbeck & Devine, 2010), several risk factors emerged as important considerations for healthcare providers who work with children with SB, including disease severity as barrier to independence with self-management (e.g., more limitations in gross motor functioning and higher spinal lesions) and family functioning (e.g., high levels of family stress) and neuropsychological deficits (e.g., executive dysfunction) as barriers to adherence and responsibility for bowel and catheterization self-management. Based on study findings, we observed three broad patterns of medical family/self-management and adherence: 1) when children had marked physical and/or neuropsychological limitations, parents typically maintained medical responsibilities and were adherent, 2) when children were high functioning, with few physical and/or neuropsychological limitations, children successfully took on medical responsibilities and were adherent, and 3) when children were not particularly high or low functioning (i.e., "middle of the road" functioning), the most difficulties with adherence occurred. Possibly, for children who are not extremely high or low functioning, other factors (e.g., family functioning) are more salient. This study is one of the first to offer targeted foci for medical adherence and family- and self-management interventions for youth with poorly managed SB, including parent stress-management and collaborative family-based, problem-solving around medical management. Attention to these risk factors is crucial for improving clinical care and the

selection and adaption of evidence-based adherence interventions that match targets for youth with SB.

Future research should consider the interactions amongst these variables across time (e.g., the transactional relationship between child neuropsychological factors and family dynamics) in relation to SB medical adherence and the allocation of treatment responsibilities. Another exciting area of research involves assessing the temporal stability of medical self-management and adherence constructs over time. A past study that utilized the same dataset showed that parents of youth with SB tended to rate similar levels of adherence across a two-year period (Psihogios, Kolbuck, & Holmbeck, 2015). However, research has yet to evaluate movement between responsibility/adherence groups and the factors that contribute to positive (or negative) movement between groups, across time. Finally, as high levels of parental involvement in the medical regimen may or may not be adaptive in the long-term, evaluating the development of medical self-management skills into emerging adulthood, when young adults seek a successful transition to adult healthcare, is of the utmost importance in this population.

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VITA

Alexandra Psihogios was born on December 19, 1988 in Damascus, Maryland to Petros and Michelle Psihogios. She graduated Cum Laude in Psychology from the University of Maryland, Baltimore County in December 2009. In December 2012, she graduated with a Masters of Arts in Clinical Psychology at Loyola University Chicago. As a graduate student, Ms. Psihogios was involved in research led by Dr. Grayson Holmbeck, which investigated psychosocial development of youth with spina bifida during the transition to adolescence. She completed her graduate and doctoral research under the mentorship of Dr. Holmbeck.

Ms. Psihogios' research work during her graduate education involved the study of medical adherence and family disease management among children and adolescents with spina bifida. Her clinical work involved consultation, intervention, and assessment with children, adolescents, young adults, and their families, with an emphasis on youth with cancer. Ms. Psihogios completed her clinical psychology internship at The Children's Hospital of Philadelphia. She is currently a postdoctoral research fellow in behavioral oncology at The Children's Hospital of Philadelphia, under the direction of Drs. Lamia Barakat and Lisa Schwartz. Ms. Psihogios' hopes to develop efficacious and cost-effective interventions for improving medical adherence among adolescents and young adults with chronic health conditions.