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Condition Self-Management in Pediatric Spina Bifida: A Longitudinal Investigation of Medical Adherence, Responsibility-Sharing, and Independence Skills

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Abstract

Objective This study aimed to evaluate rates of medical adherence, responsibility, and independence skills across late childhood and adolescence in youth with spina bifida (SB) and to explore associations among these disease self-management variables. **Method** 111 youth with SB, their parents, and a health professional participated at two time points. Informants completed questionnaires regarding medical adherence, responsibility-sharing, and child independence skills. **Results** Youth gained more responsibility and independence skills across time, although adherence rates did not follow a similar trajectory. Increased child medical responsibility was related to poorer adherence, and father-reported independence skills were associated with increased child responsibility. **Conclusions** This study highlights medical domains that are the most difficult for families to manage (e.g., skin checks). Although youth appear to gain more autonomy across time, ongoing parental involvement in medical care may be necessary to achieve optimal adherence across adolescence.

Key words: adherence; adolescents; health behavior; spina bifida.

In pediatric populations, prevalence rates of chronic disease are estimated to be 15–20% for children in the United States (e.g., asthma and diabetes; Judson, 2004; Perrin, Bloom, & Gortmaker, 2007). Many of these conditions require multifaceted care, including regular administration of medication, dietary changes, and regular clinic appointments. Across a variety of pediatric populations, research suggests that pediatric noncompliance to medical recommendations, or medical nonadherence, is associated with increased morbidity (e.g., infection and symptomatology), mortality (Kennard et al., 2004), and decreased psychological functioning (Drotar et al., 2007). From a public health perspective, medical nonadherence is also related to higher rates of health-care utilization (Piecoro et al., 2007), and affects the cost-effectiveness of medical care, clinical decisions, and clinical trials (Rapoff, 2011). In fact, the New England Healthcare Institute estimated that $290 billion in avoidable medical spending is generated annually by nonadherence (2009). Thus, the manner in which children and their family members manage chronic illness may have profound individual and societal effects on health and health-care outcomes.

The purpose of this study was to evaluate rates of self-management (i.e., medical adherence, responsibility-sharing, and independence skills) for various spina bifida (SB) medical tasks across late childhood and adolescence. Similar to the Pediatric Self-Management Model proposed by Modi and colleagues (2012), we conceptualize medical self-management as a broad

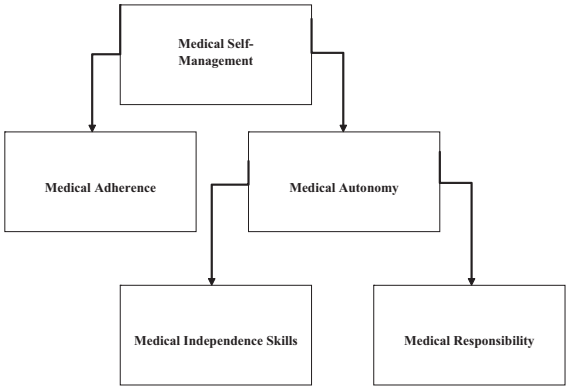

Figure 1. Medical self-management in pediatric SB: Medical adherence, responsibility, and independence skills.

term that encompasses behaviors conducted by a child or family member in relation to a pediatric chronic health concern (see Figure 1).

SB is one of the most common and disabling birth defects in the United States, occurring in roughly 3 of every 10,000 live births per year (CDC, 2011). SB results from the incomplete closure of the neural tube during the early stages of pregnancy, and includes in malformations of the spinal cord and cerebral cortex (i.e., the Arnold-Chiari II malformation). Individuals with SB experience chronic bowel and bladder difficulties, physical disabilities, and cognitive deficits throughout their lifetime. Children with SB are also at-risk for ancillary health complications, including latex allergies, urinary tract infections, tethered cord syndrome (i.e., a neurological syndrome that is characterized by abnormal attachment of spinal tissues to the spinal cord; NINDS, 2012), obesity, sleep difficulties, and secondary CNS insults due to repeated shunt repairs. Patients with SB must execute routine medical treatments to treat this condition, such as intermittent catheterization, administering medications, completing bowel programs, and conducting routine skin checks to prevent pressure sores.

Medical adherence is an aspect of medical self-management that refers to youth's compliance with their prescribed medical regimen (e.g., catheterizing appropriately and on time; Haynes, 1979; see Figure 1). Gaining insight into medical adherence behaviors during the adolescent years is essential, as long-term health-care behaviors, including diet, exercise, and drug/alcohol use, are often established and consolidated during this developmental period (Williams, Holmbeck, & Greenley, 2002). 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, Reynolds, Siminerio, Escobar, & Becker, 2008), the literature on adherence in youth SB remains in its beginning stages (e.g., Holmbeck et al., 1998; Stepansky, Roache, Holmbeck, & Schultz, 2010). Research on the topic consists of family and neurocognitive predictors of nonadherence, including associations between medical nonadherence and increased family conflict (Psihogios & Holmbeck, 2013; Stepansky et al., 2010) and executive dysfunction (O'Hara & Holmbeck, 2013). Although these preliminary studies offer useful information regarding barriers to adherence, descriptive information on condition management in SB is lacking. For example, important questions regarding actual rates of nonadherence and how these rates shift over the course of development have yet to be answered. Such research would provide useful information to medical providers regarding important targets for adherence interventions, and may be particularly important to study during adolescence when adherence rates appear to be at the their lowest levels across a variety of illness populations (i.e., a 50% adherence rate found in some populations; La Greca & Mackey, 2009).

In addition to medical adherence, a second domain related to the development of medical self-management is medical autonomy (see Figure 1). Medical autonomy refers to an interpersonal process in which an adolescent begins to develop shared responsibility (e.g., greater involvement in SB medical tasks in the context of continued parental support) and greater independence skills (e.g., competencies to complete SB medical tasks; see Figure 1; O'Hara & Holmbeck, 2013). In other words, we propose that growth in medical autonomy reflects greater responsibility *and* competency for caring for one's disease independently. Regarding medical responsibility, many youth with chronic health conditions gain increased responsibility for their care during the transition to adolescence. For example, one study found the number of children who were partially or fully responsible for SB medical regimens (e.g., catheterization and bowel programs) increased over time (Stepansky et al., 2010). By the time children were between the ages of 12 and 13 years, most gained responsibility for catheterization, and roughly half gained responsibility for their bowel program.

Although parents of youth with chronic health conditions grant increased medical responsibility during adolescence, increased responsibility may not necessarily follow from gains in medical independence skills. A large body of work in pediatric type 1 diabetes demonstrates that if the transfer of medical responsibilities from parent to child occurs when the child does not yet exhibit appropriate medical independence skills, this can have a deleterious effect on subsequent levels of adherence (e.g., Anderson, Ho, Brackett, Finkelstein, & Laffel, 1997; Holmes et al., 2006, Wiebe et al., 2014; Wysocki, Taylor, Hough, Linscheid, & Yeates, 1996). For example, Wiebe and colleagues (2014) found that when youth with

diabetes became more responsible for their medical regimen without simultaneous growth in diabetes self-efficacy, medical adherence decreased rapidly. Given these findings, it is not surprising that several investigations have shown that higher levels of parental involvement in pediatric type 1 diabetes care are associated with higher levels of treatment adherence and glycemic control (e.g., Anderson, Ho, Brackett, & Laffel, 1999, Ellis et al., 2007, & Helgeson et al., 2008). Similarly, among children with SB, higher levels of parental medical responsibility resulted in higher levels of medical adherence, even after controlling for relevant developmental and cognitive factors, such as child age and IQ (Psihogios & Holmbeck, 2013).

In summary, youth with chronic health conditions obtain increased responsibility of their medical regimen during adolescence, but this development may result in poor adherence if it is not accompanied by growth in medical independence skills (e.g., Wiebe et al., 2014; Holmes et al., 2006). These findings highlight the importance of understanding the interplay among issues related to optimal disease self-management: adherence (i.e., is the child/family compliant with medical recommendations?), responsibility (i.e., has the child been granted responsibility for completing medical tasks?), and independence skills (i.e., is the child capable of completing medical tasks autonomously?). That is, to achieve optimal disease self-management over the course of adolescence, progress across each of these three domains is essential.

In this study, we sought to study condition self-management variables simultaneously to better understand how medical tasks are managed in families of youth with SB across adolescence, and, in the process, examine the utility of a framework for condition self-management in these populations (see Figure 1). To inform future research and to guide clinical pursuits (e.g., facilitating better understanding of the medical domains that children and families struggle with most), this study provides clinically relevant, person-centered data regarding medical adherence, shared responsibility, and medical independence skills in an SB population. Specifically, through creation of dichotomous variables, we present data on rates of SB medical adherence, responsibility-sharing, and independence skills across salient medical tasks. We believe that these data will facilitate health provider and parent understanding of the percentage of patients that demonstrates difficulties with medical management across adolescence.

The first aim of this investigation was to study rates of medical adherence, responsibility-sharing, and independence skills in an SB population across late childhood and adolescence, as well as explain the relationships between these three self-management variables (see Figure 1). Though a few studies evaluated barriers to adherence and autonomy in SB populations (e.g., parenting styles and executive functioning; O'Hara & Holmbeck, 2013), no studies to our knowledge have reported descriptive data regarding disease self-management in SB. Thus, this study is the first to provide descriptive information for condition self-management skills across four salient medical tasks in SB populations: (1) catheterization, (2) bowel program (including data on the degree to which one follows dietary restrictions), (3) skin checks, and (4) exercise. In addition to this advancement, this investigation extends a previous, cross-sectional study examining the relationship between medical responsibility-sharing and adherence (Psihogios & Holmbeck, 2013). Psihogios and Holmbeck (2013) found that adherence was maximized when parents were primarily responsible for SB medical tasks and when there were low levels of family conflict. The current study extends this research by investigating the relationship between medical self-management variables across two time points, utilizing mother- and father-report data, and exploring medical independence skills in conjunction with medical adherence and shared responsibility.

Given the literature on rates of nonadherence across other illness groups, it was expected that SB adherence would also be compromised during mid-adolescence. We hypothesized that adherence would be the highest during late childhood, when parents are more involved in their child's medical care (Stepansky et al., 2010). We hypothesized that youth with SB would gradually gain greater independence skills and responsibility with their regimen across time.

A second objective of this study was to explore associations among the three disease self-management variables, including exploring whether independence skills moderate the relationship between SB responsibility-sharing and medical adherence. We expected that independence skills would be a significant moderator of the relationship between child SB responsibility and adherence, such that children with greater levels of responsibility would show the poorest adherence when they also had lower levels of skills. Finally, our third aim was to study the longitudinal relationship between disease independence skills and child-responsibility with medical tasks. We hypothesized that child independence skills at the first time point would predict later child responsibility. That is, children with more competence for managing their regimen would be granted greater responsibility for disease self-management 2 years later.

Method

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, Wasserman, Gershenson, Holmbeck, & Essner, 2011). This report used data regarding health care behaviors at Time 1 and Time 2 (data collected approximately 2 years after Time 1 data collection). 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, or myelocystocele); (2) age 8–15 years at Time 1; (3) ability to speak or read English or Spanish; (4) involvement of at least one primary caregiver; and (5) residence within 300 miles of lab to allow for home-based data collections. Of the original 246 families who met eligibility criteria, 163 families agreed to participate but 21 of those families could not be contacted or later declined, and two families eventually did not meet inclusion criteria (i.e., one child was too young and one had a milder form of SB). The final participants included 140 families of children with SB (53.6% female; *M* age = 11.40). 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 41.1 (*SD* = 15.8), suggesting a generally middle-class sample with some variability. Children of families who declined participation did not differ from those who participated with respect to type of SB (e.g. myelomeningocele or other), χ^2 (1) = .0002, $p > .05$, shunt status, χ^2 (1) = .003, $p > .05$, or occurrence/non-occurrence of shunt infections, χ^2 (1) = 1.08, $p > .05$.

Child medical information regarding physical health status was gathered from their medical chart (medical chart release was obtained during home visit) and questionnaire data. Of the 140 participants, medical chart data indicated the following diagnosis rates: 87.9% myelomeningocele, 8.3% lipomeningocele, and 3.8% other. Additionally, over half of the children had spinal lesions located in the lumbrosacral or lumbar spinal regions (62.9%), 19.0% in the sacral region, and 18.1% thoracic region. Medical chart data showed that 80.3% of the children had a shunt, and mother questionnaire data indicated the average number of shunt surgeries was 3.14 (*SD* = 5.07). According to parent-reported questionnaire data, 81.1% of the children used braces to ambulate and 61.4% used a wheelchair (as some children used both methods of ambulation). Similar to past studies (e.g., Wills, Holmbeck, Dillon, & McLone, 1990), youth with SB demonstrated a low average IQ (*M* Full Scale IQ = 85.68, *SD* = 16.58). Of the 140 children that participated in this study at Time 1, 26 children (19.7%) had an IQ of less than 70. For this study, we provided child-reported descriptive data on SB responsibility-sharing by all participants (regardless of IQ). These child-reported data for those with low IQs should be interpreted with caution owing to the possibility that IQ may have affected these ratings. All other analyses were performed with data provided by parents or health professionals, and IQ was controlled when relevant (see Objectives 2 and 3).

Procedure

At the first time point, trained undergraduate and graduate student research assistants collected data during two home visits that each lasted approximately 3 hours. Families who completed all parts of the study received monetary compensation ($150 for families) and gifts (e.g., t-shirts and pens). Informed consent from parents and assent from children were obtained before the start of the first home visit. Parents of participating children 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. As needed, research assistants read the questionnaires out loud to the child to ensure that the child understood the questions. Families also participated in audio- and video-taped structured interaction tasks. Additionally, neuropsychological testing of the child was completed to assess IQ and other neuropsychological indices.

The second time point of the study took place approximately 2 years after participants had completed their first home visit. One hundred eleven of the original 140 participants completed Time 2 (i.e., 80% of the sample). At this time point, five families withdrew and one participant passed away. Nine participants declined participation but agreed to be re-contacted for Time 3. Eleven families had invalid contact information and three families consented to do the study via mail, but failed to return the questionnaires. At Time 2, data were collected during one visit to the family's home. Similar to Time 1, families who completed all parts of the study (i.e., questionnaires, audio-taped interviews, neuropsychological testing, and structured video-taped interactions) received monetary compensation ($150 for families).

Measures

Medical Adherence (Time 1 and Time 2)

The SB Self-Management Profile (SBSMP; Wysocki & Gavin, 2006) measured parent-reported adherence to SB medical treatments. For the purposes of this study, only the diet recommendations, catheterization, bowel program, skin checks, and exercise subscales were used. These scales were chosen because they represent some of the most essential SB medical tasks and also appear on other self-management measures used in

this study, thus, allowing a comparison of specific SB tasks across measures.

The SBSMP is a 14-item, structured interview that addresses different aspects of SB medical regimen. This scale addresses whether a task is being completed, but it does not specify who is responsible for completing specific medical tasks (e.g., parent or child). This investigation focused on items related to adherence to catheterization (e.g., catheterizing on time, following steps of catheterization), bowel program (e.g., taking medications to reduce constipation, following dietary recommendations), skin checks, and exercise. In this study, this measure was administered to mothers and fathers as a questionnaire rather than as an interview. Owing to the number of participants who completed each item (i.e., parents can endorse "not applicable" for certain items), scale reliability could not be computed for this sample. Total scores were transformed into z-scores because the Likert scale response range varied across items (i.e., the measures includes 4-, 5-, and 6-point Likert scales).

In this study, we formed categorical variables from items assessing adherence to catheterization, bowel program, skin checks, and exercise. Items were dichotomized based on clinically relevant cut-points. For instance, based on clinical recommendations that children with SB catheterize every 3–4 hr, a score of "1" or "adherent" was given to families who indicated that they miss catheterizing —four to five times per week or less (i.e., less than once per day). A score of "0" or "nonadherent" was given to families who indicated that they missed catheterization one time per day or more (i.e., failing to catheterize within a 6–8 hr window or more). Using bowel program as a second example, families who indicated that they take medications to reduce constipation (i.e., suppositories, enemas, or stool-softening medications) 50% of the time or greater were assigned a score of "1" or "adherent" to bowel program. On the other hand, a score of "0" or "nonadherent" was assigned to families who indicated that the child takes bowel medications less than 50% of the time. For any medical domain, if the family indicated that their child was not required to complete the medical task, they were dropped from the analysis for that specific domain.

Health-professional report on a medical adherence screener was also used to assess rates of compliance to specific medical tasks (e.g., catheterization, bowel program, skin checks, and exercise). Adherence rates were again calculated by dichotomizing items that included the following response options: "never," "occasionally," "sometimes," "usually," and "always." If the health-professional rated the child as "sometimes," "usually," or "always" following treatment recommendations, a score of "1" or "adherent" was assigned to that task. If the health-professional rated that child as "occasionally" or "never" following medical recommendations, the child received a score of "0" or "nonadherent" for that task.

Although dichotomizing a continuous variable reduced variability in responses, cut-points were established to create meaningful clinically relevant categorical variables (i.e., actual rates of medical adherence across salient SB medical tasks). The creation of categorical variables enabled computation of frequencies of children who were adherent with specific SB medical tasks versus frequencies of children who were not adherent (i.e., we sought to investigate rates of medical adherence). This approach offers more specific information for health-care providers who are interested in the percentage of patients who manage SB tasks successfully across adolescence.

Medical Responsibility Sharing (Time 1 and Time 2)

The Sharing of SB Management Responsibilities Scale (SOSBMR), which is an adaptation of the Diabetes Family Responsibility Questionnaire (Anderson, Auslander, Jung, Miller, & Santiago, 1990), was used to assess mother, father, and child perceptions of the division of SB responsibilities within the family. The SOSBMR consists of 34 items that describe SB and health-related issues relevant to children with SB (e.g., remembering to catheterize regularly, every 2–4 hr). Parents and their children independently rated who was primarily responsible for each task (e.g., parent, child, equal, or not applicable). These items are grouped into several subscales: health-care appointments, communication about SB, medications, general needs and self-care, ambulation, skin care, catheterization, bowel management, exercise, and diet. Owing to the possibility of "not applicable responses," internal consistency at the total-score level could not be computed, as reliability software uses listwise deletion when computing alpha coefficients. Rather, analyses of internal consistency at the subscale levels were conducted. At Time 1 and Time 2, acceptable alphas were found across all subscales for mother, father, and child data (i.e., $\alpha > .60$).

For items on the four subscales (catheterization, bowel program, skin checks, and exercise), a score of "1" was assigned to tasks where the parent is primarily responsible, "2" was assigned to tasks that shared equally between the parent and child, and "3" pertained to tasks the child was primarily responsible. Total subscale mean scores ranged from 1 to 3. For this project, any subscale mean above or equal to 2.1 (i.e., slightly above "shared responsibility") was assigned a "1" or "child responsible" for that particular medical domain. Any score below 2.1 (i.e., scores ranging from "shared responsibility" to "parent responsibility") was assigned a score of "0" or "parent responsible" for that particular medical domain.

Medical Independence Skills (Time 1 and Time 2)

The Spina Bifida Independence Survey (SBIS), adapted from the Diabetes Independence Survey (Wysocki, Meinhold, et al., 1996), was used to measure parent's evaluation of the child's attainment of SB skills and knowledge. The SBIS is composed of 50 SB-specific skill questions to which participants respond "yes," "no," "not sure," or "not applicable" for each item that assessed whether the child was able to correctly perform each skill independently (e.g., move in and out of his/her wheelchair at home, perform each catheterization step correctly). The items on the SBIS are organized into different medical task categories, including: (1) recognizing symptoms, (2) communication, (3) medications, (4) diet, (5) exercise, (6) ambulation, (7) basic needs, (8) skin, (9) urinary tract infection, (10) catheterization, and (11) bowel. Similar to the other measures, a total score internal consistency value could not be calculated owing to missing or "not applicable" responses. Acceptable internal consistency (i.e., $\alpha > .60$) was found for all continuous subscales, with the exception of mother-reported exercise at Time 1 ($\alpha = .57$). Thus, analyses involving mother-reported child independence with exercise at Time 1 were not evaluated.

Similar to how the other scales were scored, this study formed categorical variables from items assessing independence skills in the areas of catheterization, bowel program, skin checks, and exercise. On individual items on the SBIS, responses of "no" to an item were given value of "1" (indicating that child had not mastered the skill), whereas a response of "yes" was assigned a value of "2" (indicating the child has mastered the skill). Data from parents who endorsed "not sure" or "not applicable" were removed from relevant analyses. To dichotomize continuous scale means, medical domains in which the mean across items was equal to 2.00 were assigned a score of "1" or "child possessing independence skills." Medical domains in which the mean across items was ≤ 1.99 were assigned a score of "0" or "child not possessing independence skills."

Moderation Analyses

We hypothesized that concurrent child independence skills (across all medical domains) would moderate associations between child responsibility with medical tasks and medical adherence, such that children with increased responsibility would show the poorest adherence when they also had lower levels of skills. That is, the relationship between child responsibility and medical adherence would depend on the presence of low or high medical independence skills. To test this hypothesis, procedures outlined by Aiken and West (1991) were followed for testing interactions using multiple regression. Specifically, the independent variables were centered (by subtracting the appropriate sample means) and centered predictors were used in the analyses. The predictors for the regressions were child age, gender, IQ, and socioeconomic status (SES) (covariates), child independence skills (mother and father reports at Time 1 and Time 2), child responsibility for medical regimen (mother and father reports at Time 1 and Time 2) and the interaction term: Responsibility × Independence Skills. The dependent variable was mother- or father-reported medical adherence at Time 1 or Time 2. All regressions were run separately for each time point and reporter, yielding a total of eight regressions (e.g., mother report predicting mother-reported adherence at Time 1 and Time 2, father report predicting father-reported adherence at Time 1 and Time 2, mother report predicting father-reported adherence at Time 1 and Time 2, and father report predicting mother-reported adherence at Time 1 and Time 2).

Results

Objective 1

The first objective of this study was to analyze rates of medical adherence, child-responsibility, and independence skills across the four key medical domains: catheterization, bowel program, skin checks, and exercise. Rates of self-management behaviors were determined by evaluating the percentage of participants that fell in "nonadherent," "child responsible for medical tasks," and "child possessing independence skills" categories.

Adherence

Collapsing across age-groups, mothers endorsed the highest level of nonadherence for skin checks, dietary recommendations, and taking medications to reduce constipation at Time 1 and Time 2 (see Table I). Similarly, fathers also endorsed the highest level of nonadherence to skin checks, dietary recommendations, and taking medications to reduce constipation at Time 1 and Time 2. In contrast, overall rates of parent-reported nonadherence for catheterizing appropriately and on time were significantly lower (<20% nonadherent across reporters). Health professionals rated adherence to catheterization as the most problematic among other domains (i.e., 13.9% and 10.4% at Time 1 and Time 2, respectively).

Given the large age range at Time 1 (i.e., 8–15 year olds) and Time 2 (i.e., 10–18 year olds), descriptive data using age cohorts are also reported (i.e., analyzing rates of nonadherence among 8–9 year olds, 10–11 year olds, 12–13 year olds, 14–15 year olds, and 16–17 year olds; see Table I). In this way, this study possessed the ability to analyze trends across developmental periods. Results suggested variability in adherence rates across tasks, age-groups, and reporters. Interestingly, rates of nonadherence appeared to

Table I. Mother- and Father-Reported Rates of Nonadherence (% Nonadherent) at Time 1 and Time 2: Overall and by Age Cohorts

Time point	Medical task	8–9 year olds	10–11 year olds	12–13 year olds	14–15 year olds	16–17 year olds	Overall
1	Diet	34.4%(29)	44.4%(26)	40.0%(25)	33.3%(24)	–	**34.0%(104)**
		33.3%(24)	21.7%(23)	25.0%(16)	29.2%(24)	–	**27.3%(87)**
2		–	47.8%(23)	20.0%(15)	45.0%(25)	37.5%(16)	**33.3%(78)**
		–	31.8%(22)	38.4%(13)	28.5%(14)	26.7%(15)	**31.3%(64)**
1	Bowel Medications	25.%9(27)	31.2%(16)	23.0%(13)	33.3%(15)	–	**28.2%(71)**
		22.2%(18)	7.7%(13)	18.2%(11)	29.4%(17)	–	**20.3%(59)**
2		–	25.0%(16)	12.5%(8)	45.5%(11)	36.3%(11)	**30.4%(46)**
		–	38.9%(18)	40.0%(5)	28.6%(7)	33.3%(9)	**35.9%(39)**
1	Skin checks	38.2%(34)	29.6%(27)	24.1%(29)	25.9%(27)	–	**30.3%(117)**
		47.8%(23)	29.2%(24)	26.3%(19)	24.0%(25)	–	**33.3%(91)**
2		–	53.8%(26)	47.1%(16)	29.6%(28)	42.1%(19)	**42.7%(89)**
		–	35.0%(20)	50.0%(14)	37.5%(16)	28.6%(14)	**37.5%(64)**
1	Exercise	12.1%(33)	14.8%(27)	21.4%(28)	25.0%(24)	–	**17.5%(112)**
		8.3%(24)	29.2%(24)	31.6%(19)	28.0%(25)	–	**24.4%(92)**
2		–	7.4%(27)	16.6%(18)	29.6%(27)	21.1%(19)	**18.7%(91)**
		–	18.2%(22)	14.3%(14)	25.0%(16)	20.0%(15)	**19.4%(67)**
1	Catheterize on Schedule	23.0%(26)	14.3%(21)	12.5%(24)	9.1%(22)	–	**14.9%(93)**
		4.8%(21)	5.0%(20)	0.00%(15)	9.5%(21)	–	**5.1%(77)**
2		–	13.0%(23)	15.4%(13)	16.7%(18)	20.0%(15)	**15.9%(69)**
		–	15.7%(19)	9.1%(11)	8.3%(12)	16.7%(12)	**13.0%(54)**
1	Follow steps of Catheterization	11.1%(27)	4.4%(23)	0.00%(23)	4.4%(23)	–	**5.2%(96)**
		13.0%(23)	5.3%(19)	0.00%(16)	8.7%(23)	–	**7.3%(81)**
		–	0.00%(24)	0.00%(24)	0.00%(19)	6.7%(15)	**1.4%(71)**
		–	5.0%(20)	0.00%(11)	0.00%(13)	0.00%(12)	**1.9%(56)**

Note. Mother report listed in first row, father report listed in second row, *n* for each group in parentheses.

decline for catheterization (e.g., catheterizing on time and following steps of catheterization), but generally peaked during late childhood for other medical domains (e.g., bowel, diet, exercise, and skin checks). For example, children who were 8–9 years old at Time 1 displayed higher rates of mother-reported nonadherence to diet recommendations 2 years later, when they were now between the ages of 10–11 years (see Table I). Similarly, children who were 8–9 years old at Time 1 displayed significantly higher rates of father-reported nonadherence to skin recommendations at Time 2.

Responsibility Sharing

Overall rates of child responsibility indicated that across reporters and medical tasks, children gained responsibility over their medical care over time (see Table II). Among the medical domains, mothers, fathers, and children reported the highest levels of child responsibility for catheterization at both Time 1 and Time 2. Age cohort data showed a fairly clear relationship between age and increased responsibility, and this finding was consistent across the three reporters. However, children typically rated themselves as more responsible than did mothers and fathers (e.g., children were more likely than mothers and fathers to report that they were responsible for bowel programs across both time points and all age cohorts).

Independence Skills

In terms of independence skills to execute the various SB medical tasks, mothers rated children as showing the highest level of independence skills with bowel programs at Time 1, whereas fathers rated children as showing the highest level of skill with catheterization at Time 1 (see Table III). At Time 2, fathers continued to rate children as most competent with catheterization, whereas mother rated children as most competent with exercise. For the most part, with increasing age, parents rated children as having increasing ability to take care of a variety of medical tasks (e.g., exercise, skin checks, and catheterization). A notable exception to this trend was that mothers rated children as having lower levels of competence for managing bowel programs at Time 2 compared with Time 1.

Age cohort data revealed a similar trend; namely, as children got older, they appeared to gain more skill across most SB medical tasks (see Table III). For example, mothers and fathers consistently rated older children as having more skill in catheterization over time, such that 81.3–90.0% of children in the 16–17 year old cohort possessed appropriate catheterization abilities, depending on the reporter. Although this trend was observed for diet, catheterization, skin checks, and exercise, it was not consistently true for child independence skills with bowel programs (i.e., mothers and fathers typically rated the lowest level of competence for bowel programs for the 10–11 year old cohort).

Table II. Mother-, Father-, and Child-Reported Rates of Child Responsibility (% Responsible) at Time 1 and Time 2: Overall and by Age Cohorts

Time point	Medical task	8–9 year olds	10–11 year olds	12–13 year olds	14–15 year olds	16–17 year olds	Overall
1	Bowel	3.0%(33)	14.8%(27)	24.1%(29)	34.6%(26)	–	**17.9%(115)**
	Program	4.0%(25)	0.0%(22)	30.0%(20)	40.0%(25)	–	**18.1%(92)**
		15.2%(33)	46.2%(26)	51.5%(33)	62.0%(29)		**43.1%(121)**
2		–	25.9%(27)	17.6%(17)	42.9%(28)	52.6%(19)	**36.1%(91)**
		–	9.5%(21)	13.3%(15)	35.3%(17)	56.3%(16)	**26.7%(69)**
		–	31.1%(28)	36.8%(19)	70.0%(30)	71.4%(21)	**54.7%(98)**
1	Skin check	5.9%(34)	26.9%(26)	41.4%(29)	30.4%(23)	–	**25.7%(112)**
		0.0%(24)	8.7% (23)	25.0%(20)	28.0%(25)	–	**10.5%(92)**
		15.2%(33)	28.0%(25)	28.1%(32)	48.1%(27)	–	**29.4(117)**
2		–	25.0%(24)	29.4%(17)	40.7%(27)	56.3%(16)	**37.1%(84)**
		–	22.2%(18)	26.7%(15)	35.3%(17)	50.0%(16)	**31.0%(66)**
		–	29.2%(24)	36.8%(19)	58.0%(31)	61.9%(21)	**51.0%(95)**
1	Exercise	16.0%(25)	17.4%(23)	32.0%(25)	33.3%(21)	–	**25.0%(94)**
		10.0%(20)	0.0% (16)	35.7%(14)	23.8%(21)	–	**9.0%(71)**
		26.1%(23)	36.8%(19)	40.7%(27)	52.2%(24)	–	**40.4%(93)**
2		–	4.0%(25)	13.3%(24)	50.0%(26)	53.3%(15)	**31.0%(81)**
		–	0.0%(13)	0.0%(11)	33.3%(15)	37.5%(16)	**20.0%(55)**
		–	34.8%(23)	66.7%(15)	69.2%(26)	60.0%(20)	**56.0%(84)**
1	Catheterization	40.7%(27)	52.4%(21)	66.7%(24)	72.7%(22)	–	**57.9%(94)**
		36.4%(22)	30.4%(23)	52.6%(19)	58.3%(24)	–	**30.1%(88)**
		28.1%(32)	45.8%(24)	66.7%(30)	75.0%(28)	–	**54.3%(114)**
2		–	38.4%(26)	62.5%(16)	72.0%(25)	66.7%(18)	**59.6%(85)**
		–	42.1%(19)	64.3%(14)	76.5%(17)	80.0%(15)	**65.2%(65)**
		–	45.8%(24)	70.6%(17)	88.5%(26)	75.0%(20)	**69.9%(87)**

Note. Mother report listed in first row, father report in second row, child report in third row, *n* for each group in parentheses.

Table III. Mother- and Father-Reported Rates of Child Independence Skills With Medical Regimen (% Competent) at Time 1 and Time 2: Overall and by Age Cohorts

Time point	Medical task	8–9 year olds	10–11 year olds	12–13 year olds	14–15 year olds	16–17 year olds	Overall
1	Diet	54.1%(34)	55.6%(27)	44.4%(27)	76.2%(21)	–	**57.3%(109)**
		70.3%(27)	62.5%(24)	75.0%(20)	83.3%(24)	–	**73.2%(95)**
2		–	67.9%(28)	77.8%(18)	92.8%(28)	85.0%(20)	**80.2%(94)**
		–	60.9%(23)	66.7%(15)	83.3%(18)	62.5%(16)	**67.9%(72)**
1	Bowel program	100.0%(34)	85.2%(27)	96.7%(30)	95.8%(24)	–	**94.8%(115)**
		51.9%(27)	45.8%(24)	55.0%(20)	64.0%(25)	–	**54.6%(96)**
2		–	60.1%(28)	83.3%(18)	82.1%(28)	80.0%(20)	**74.3%(94)**
		–	56.5%(23)	40.0%(15)	61.1%(18)	75.0%(16)	**59.0%(72)**
1	Skin checks	45.5%(33)	57.1%(28)	62.1%(29)	58.3%(24)	–	**54.8%(114)**
		66.7%(27)	66.7%(24)	75.0%(20)	72.0%(25)	–	**69.4% (96)**
2		–	67.9%(28)	77.8%(18)	75.0%(28)	80.0%(20)	**74.3%(94)**
		–	69.6%(23)	66.7%(15)	83.3%(18)	75.0%(16)	**74.4%(72)**
1	Exercise	–	–	–	–	–	–
		66.7%(27)	66.7%(24)	65.0%(20)	79.1%(24)	–	**70.1%(95)**
2		–	82.1%(28)	77.8%(18)	85.7%(28)	80.0%(20)	**82.2%(94)**
		–	69.6%(23)	73.3%(15)	88.9%(18)	68.8%(16)	**76.9%(72)**
1	Catheterization	46.4%(28)	52.3%(21)	78.2%(23)	71.4%(21)	–	**61.7%(93)**
		74.0%(27)	75.0%(24)	75.0%(20)	80.0%(25)	–	**76.3%(96)**
2		–	75.0%(28)	72.2%(18)	75.0%(28)	90.0%(20)	**78.2%(94)**
		–	82.6%(23)	100.0%(15)	94.4%(18)	81.3%(16)	**88.5%(72)**

Note. Mother report listed in first row, father report listed in second row, *n* for each group in parentheses, data for mother-report exercise at Time 1 dropped owing to low alpha score.

Objective 2

After exploring self-management skills at the subscale level, a second objective of this study was to explore associations between disease self-management variables (using continuous total scores), including exploring the moderating role of independence skills on the relationship between SB responsibility sharing and medical adherence. Preliminary analyses of potential confounds (age, gender, IQ, and SES; see Table IV) revealed that child age at Time 1 was positively associated with mother- and father-reported medical responsibilities and independence skills at Time 1 and

Table IV. Correlation Coefficients of Parent-Reported Overall Adherence, Independence Skills, and Responsibility Sharing and Child Demographics at Time 1 and Time 2

Medical/Demographic variables	T1 Adh Mo	T1 Adh Fath	T1 Ind Mo	T1 Ind Fath	T1 Resp Mo	T1 Resp Fath	T2 Adh Mo	T2 Adh Fath	T2 Ind Mo	T2 Ind Fath	T2 Resp Mo	T2 Resp Fath	Age	SES	IQ
T1 Adh Mo	1														
T1 Adh Fath	.48**	1													
T1 Ind Mo	.03	−.10	1												
T1 Ind Fath	−.08	−.00	.66**	1											
T1 Resp Mo	−.20*	−.08	.52**	.61**	1										
T1 Resp Fath	−.26*	−.06	.50**	.61**	.76**	1									
T2 AdhMo	.51**	.07	.13	−.14	−.07	−.15	1								
T2 Adh Fath	.21	.18	.08	.16	.05	.12	.35**	1							
T2 Ind Mo	.08	−.02	.72**	.69**	.62**	.49**	.07	.22	1						
T2 Ind Fath	−.18	−.09	.60**	.83**	.62**	.60**	−.22	.08	.65**	1					
T2 Resp Mo	−.05*	.01	.48**	.66**	.81**	.71**	−.08	.04	.61**	.62**	1				
T2 Resp Fath	−.17	.06	.41**	.64**	.64**	.67**	−.20	−.15	.51**	.65**	.75**	1			
Age	.06	.06	.23**	.33**	.36**	.28**	.05	.11	.39**	.24*	.41**	.46**	1		
SES	−.11	.00	−.03	.06	.10	.23*	−.10	−.20	.00	.09	.23*	.24*	−.09	1	
IQ	−.10	−.15	.14	.37**	.25**	.32**	−.10	−.08	.10	.41**	.32**	.31**	−.24**	.47**	1

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

Time 2. There was also a significant positive relationship between SES and father- and mother-reported child responsibility at Time 2. Child IQ was positively associated with father-reported independence skills at Time 1 and Time 2, mother- and father-reported child responsibility at Time 1, and mother- and father-reported child responsibility at Time 2.

Paired-samples *t* tests showed significant differences across the two time points for mother- and father-reported independence skills and child responsibility ($p < .05$), but not adherence ($p > .05$). Several significant correlations were found among the self-management variables (see Table IV). In general, child independence skills were positively associated with increased responsibility. Mother-reported adherence at Time 1 was positively associated with mother-reported adherence at Time 2, and father-reported adherence at Time 2 was positively associated with mother-reported adherence at Time 2.

Regarding the stability of adherence across Time 1 and Time 2, 57.8% of mothers and 73.1% of fathers rated families as "adherent" at both time points or "nonadherent" at both time points for diet recommendations. For bowel programs, 58.7% of mothers and 61.5% of fathers rated families as "adherent" or "nonadherent" at both time points. Regarding skin checks, 66.3% of mothers and 50.0% of fathers rated the same level of adherence to skin checks across the two time points. For exercise, 76.9% of mothers and 74.6% of fathers rated the same level of adherence to exercise regimens at both time points. In terms of catheterization, 73.9% of mothers and 77.8% of fathers endorsed the same level of adherence to catheterizing on schedule, across both time points. Finally, 85.9% of mothers and 87.5% of fathers rated the same level of adherence to following the steps of catheterization at Time 1 and Time 2. In general, parents tended to rate lower adherence at Time 2 for skin checks and catheterizing to schedule (see Table I).

Regression analyses examining the Child Independence Skills (across all medical tasks) × Child Responsibility (across all medical tasks) interaction for predicting concurrent adherence were nonsignificant across all analyses and reporters. However, the main effect for increased mother-reported child responsibility predicting poorer mother-reported adherence was significant at Time 1, suggesting that mothers perceived poorer adherence in the face of increased child responsibility at both time points (see Table V). Father-reported child responsibility was also associated with poorer mother-reported adherence at Time 1.

Objective 3

Regression analyses exploring the longitudinal relationship between parent perception of overall independence skills for SB medical tasks and later SB responsibilities at Time 2 were conducted. Across the two analyses, mother- or father-reported child responsibility for medical tasks at Time 1 and relevant covariates (age, gender, SES, and IQ) were controlled. Results of the analyses showed a significant relationship between father-reported child independence skills at Time 1 and later father-reported child responsibility ($B = .54$, $\beta = .12$, $t(64) = 2.33$, $p < .05$). This finding suggests that father report of child skills with medical tasks at Time 1 was associated with increases in their responsibility 2 years later. In contrast, this relationship was nonsignificant when using mother-report data.

Table V. Significant Regression Analyses for Child Responsibility × Child Independence Skills Predicting Spina Bifida Medical Adherence

Predictor	*b*	*β*	*t*	*p*
DV = Mother-reported adherence at Time 1				
Covariate: Age	.01	.06	.57	.57
Covariate: Gender	−.05	−.04	−.45	.65
Covariate: SES	−.00	−.11	−.99	.32
Covariate: IQ	−.00	−.03	−.22	.83
Mother-Reported Child Responsibility (Time 1)	−.30	−.24	−2.24	.02*
Mother-Reported Child Independence Skills (Time 1)	.34	.17	1.53	.13
Child Responsibility × Child Independence Skills (Time 1)	.01	.00	.02	.99
DV = Mother-reported adherence at Time 1				
Covariate: Age	.01	.06	.56	.58
Covariate: Gender	.04	.04	.37	.71
Covariate: SES	.00	.02	.15	.89
Covariate: IQ	−.00	−.07	−.55	.59
Father-Reported Child Responsibility (Time 1)	−.41	−.34	−2.76	.01*
Father-Reported Child Independence Skills (Time 1)	.27	.14	.99	.36
Child Responsibility × Child Independence Skills (Time 1)	−.33	−.07	−.59	.59

Discussion

The primary objective of this study was to present a person-centered, clinically relevant profile of rates of medical nonadherence, responsibility sharing, and independence skills in families of youth with SB. We also sought to test relevant hypotheses regarding the moderating role of child independence skills on the relationship between child medical responsibility and adherence, as well as the longitudinal link between child medical independence skills and gains in medical responsibilities. Given the relatively small body of literature on adherence in pediatric SB (e.g., Holmbeck et al., 1998; Psihogios & Holmbeck, 2013; Stepansky et al., 2010), we formulated these aims to provide a solid foundation of knowledge regarding the development of SB self-management behaviors across the course of childhood and adolescence.

Given the literature on rates of nonadherence across other illness groups, we hypothesized that adherence would be compromised during adolescence. Overall rates of parent-reported medical nonadherence appeared to peak during late childhood for many medical domains (e.g., bowel, diet, exercise, and skin checks). Rates of nonadherence across medical subdomains (when collapsing across age-groups) ranged from 1.3% (e.g., mother-reported nonadherence to following steps of catheterization at Time 1) to 42.7% (e.g., mother-reported nonadherence for completing skin checks at Time 2), which were below the expected 50% nonadherence rate. However, with regards to particular tasks and age cohorts, rates of nonadherence approached 50%. For example, mothers and fathers rated 24.1–50.0% of 12–13 year olds as nonadherent to skin checks. Thus, rates of nonadherence may be particularly high within specific medical tasks, during specific developmental periods (e.g., late childhood). Given that the present adherence measure does not assess who is responsible for completing medical tasks, the decreased adherence rate in early adolescence likely reflected increased child responsibility (as evidenced by descriptive responsibility data). This is consistent with past research, which has shown that during adolescence, parental responsibility decreases at the same time that medical self-management deteriorates (Helgeson, Siminerio, Escobar, & Becker, 2009; King, Berg, Butner, Butler, & Wiebe, 2014).

Importantly, health professionals rated adherence to catheterization as the most problematic. Perhaps, differences in parent and health professional reports reflect the fact that the consequences of nonadherence to catheterization may require direct medical intervention, whereas nonadherence to other medical tasks may not require health-care provider involvement. Another explanation is that health professionals may overestimate medical adherence, which has been reported in other illness populations (e.g., adults with hypertension; Steinman et al., 2004).

In terms of the temporal stability of adherence, parents tended to rate similar levels of adherence across the two time points (i.e., 50% of parents or more rated stabile levels of adherence across the medical domains). However, parents tended to rate lower adherence for skin checks and catheterizing on time at Time 2 compared with Time 1 (see Table I; although rates of nonadherence for catheterization were generally low). Interestingly, one study of a separate sample of youth with SB found that parents of youth with SB seemed to transfer responsibility for catheterization during childhood (Stepansky et al., 2010), which also seemed to be the case in this study (e.g., between 28.8 and 40.7% of children were fully responsible for catheterization at ages 8–9 years, depending on the reporter). Perhaps, lower rates of nonadherence for catheterization reflect a successful and earlier transfer of medical responsibilities from parent to child,

whereas varying rates of nonadherence across the other medical domains (with typically lower levels of adherence during late childhood and early adolescence) capture the child's difficulties with managing disease tasks independently for the first time. Clearly, families struggled with adherence to diet, bowel programs, and skin checks, which offers important targets for adherence interventions. Without intervention, ongoing adherence issues in these domains may result in bowel incontinence, pressure sores, and obesity.

We hypothesized that youth with SB would generally gain greater responsibility and independence skills with their regimen across time, such that older adolescents would possess the highest level of responsibility and independence skills for their medical regimen. Descriptive data supported this hypothesis, with age cohort data generally following a trend of increased responsibility and independence skills with age. Correlational data provided supplementary evidence that youth gain greater self-management with time, as child age was consistently, and positively, correlated with increased medical responsibility and independence across reporters (child, mother, and father) and time points (Time 1 and Time 2).

These findings provide encouraging evidence to suggest that medical responsibility and independence skills increase with age in this population. However, reporters did not indicate "100%" child responsibility or independence skills in any medical domain or age-group, suggesting that youth are continuing to develop these skills in late adolescence and early adulthood. Interestingly, descriptive data revealed that youth viewed themselves as more responsible than did parents for bowel programs, skin checks, and exercise. This finding was consistent with child development literature in general (i.e., adolescents typically rate themselves as more competent and independent than parents do; e.g., Dekovic, Noom, & Meeus, 1997), as well as a past study that found that youth with SB rated themselves as more independent with their medical regimen than mothers rated their children (Psihogios & Holmbeck, 2013). A camp-based psychosocial intervention showed that children and adults with SB showed gains in independence skills and responsibility after a weeklong intervention, which suggests that these two domains can be targets for meaningful interventions (Holbein et al., 2013; O'Mahar, Holmbeck, Jandasek, & Zuckerman, 2010).

Regarding the second objective, we expected that independence skills would moderate the relationship between child SB responsibility and adherence, such that children with increased responsibility would show the poorest adherence when they also had lower levels of skills. Although the interaction term was nonsignificant, mother-reported and father-reported child responsibility were negatively associated with concurrent adherence at Time 1, suggesting that parents viewed their children as less adherent when the child was primarily responsible for his/her medical regimen. This finding is similar to the literature on the sharing of children's diabetes responsibilities, which has consistently demonstrated that lower parent involvement over the span of adolescence is associated with less favorable diabetes-related outcomes (e.g., Helgeson et al., 2008; Wiebe et al., 2005). Similarly, one cross-sectional study that used the same data set as the current study showed that high parent–child agreement that the responsibility belonged to the child was directly associated with poor medical adherence, and adherence was maximized when parents were primarily responsible for SB medical tasks and when there were low levels of family conflict (Psihogios & Holmbeck, 2013).

Finally, as part of our third objective, we suspected that child independence skills at Time 1 would predict later child responsibility. That is, children with more competence for managing their regimen would be granted greater responsibility 2 years later. After controlling for relevant confounds in both regressions (including parent-reported child responsibility at Time 1), we found a significant relationship between father-reported child independence skills at Time 1 predicting increased child responsibility at Time 2. In contrast, this relationship was nonsignificant when using mother-report data. The results suggest that when fathers perceive their children to have a high level of skill with SB medical regimens, they also observe high levels of child medical responsibility 2 years later. However, mothers' perceptions of child competence with medical regimen were not associated with increased medical responsibility, suggesting that other factors may influence responsibility sharing (e.g., maternal overprotectiveness). Given that mothers of children with SB have been found to be more overprotective than fathers of children with SB and mothers of typically developing children (Holmbeck et al., 2002), mothers may be less likely to relinquish medical responsibilities to their children (even in the face of high levels of child competence). Put another way, mothers may be more responsible for the caretaking demands of raising a chronically ill child and may be more prone to "miscarried helping" because they perceive their children to be more vulnerable because of their medical condition (Anderson & Coyne, 1991; Thomasgard & Metz, 1997), resulting in medical caretaking even when the child is developmentally capable of taking on additional responsibilities.

Although this study included a number of strengths (such as developing clinically relevant cut-points for disease self-management variables, the inclusion of mother and father data, and analyzing self-

management variables across time), there are several limitations of the current study that should be addressed in future work. First, as is typical in studies of pediatric populations, the sample size in this study was relatively small and predominantly Caucasian. Future research should continue to strive for a more representative sample of Spanish-speaking families, as well as other ethnic groups. Secondly, to explore the association between child independence skills and later child responsibility, we relied on one method (e.g., questionnaire data) and one reporter (e.g., mother-reported child independence predicting mother-reported responsibility). Although our interest was in understanding whether one parent's perception of child independence skills predicted less perceived involvement in medical tasks over time, we recognize that these findings should be interpreted with caution owing to the presence of common-method variance. Third, when assessing the age cohort data, age cohorts possessed unequal sample sizes, which may have resulted in spurious findings for the smaller subgroups. For example, for cohorts with lower n's, a few nonadherent participants could potentially have a stronger effect on non-adherence rates. Another important limitation of the study was that longitudinal analyses were limited to two measurement points, which limited our ability to examine more complex hypotheses (e.g., mediation models) and statistical analyses (e.g., latent growth curve modeling). Furthermore, reliance on parent report of child independence skills was less than ideal, as parents' perceptions of children's abilities to complete medical tasks may be contingent on child responsibility (e.g., if a child is not responsible for medical task, a parent does not have the opportunity to observe the child performing the skill).

There were also several limitations regarding the measurement of medical adherence in this study. The reliance on self-report questionnaires to assess adherence, which has consistently yielded inflated rates of adherence across a variety of pediatric populations and respondents (e.g., Bender et al., 2000), may not be adequate to fully capture the complexity of adherence behaviors in chronically ill populations (including who is taking responsibility for completing medical tasks). Although 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 (which reduced the sample size). 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 evaluation of medical adherence in this population.

The results of this study have important clinical implications. This study suggested that families struggle with adherence to many SB self-management tasks (e.g., bowel program, exercise, skin checks, and diet); such information may be useful for both medical and behavioral health providers working with families of children with SB. Although child independence skills and responsibility for the medical regimen increased over time, families continued to play a large role in SB management (even in the 16–17 year old cohort). Additionally, similar to a past study (Psihogios & Holmbeck, 2013), this study found evidence to suggest that parental involvement in SB medical care appears to be essential for optimal adherence. That is, ongoing monitoring and teaching by parents may be necessary across adolescence and into emerging adulthood.

Although it appears that family involvement has an effect on the management of pediatric conditions (including SB), the existing body of literature has yet to evaluate whether family functioning and other relevant variables (such as the child's neuropsychological functioning, socialization, and disease severity) relate to an adolescent's ability to be independent *and* adherent. In past research studies, adherence has been evaluated broadly, without consideration of 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). Future research should evaluate medical adherence and autonomy simultaneously to provide further understanding of the variables that prevent or promote the successful transfer of medical responsibilities from parent to child. Evaluating the development of self-management skills into emerging adulthood, when young adults seek a successful transition to adult health care, may be especially important in this population.

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