

Medical Adherence in Young Adolescents with Spina Bifida: Longitudinal Associations with Family Functioning

Mona A. Stepansky, PhD, Caitlin R. Roache, MA, Grayson N. Holmbeck, PhD, and Karen Schultz, BS
Loyola University

Objective The purpose of this study was 2-fold: (1) to explore the transfer of responsibility of medical tasks from parent to child during the transition to adolescence, and (2) to examine the associations between family functioning and medical adherence in youth with spina bifida. **Methods** Seventy families of children with spina bifida participated in this study. Data were collected during family interaction sessions by using questionnaires completed by mothers, fathers, youth, teachers and health professionals. **Results** Findings suggest that responsibility for medical regimens transfers gradually from parent to child over time. Additionally, family conflict and cohesion were correlated with medical adherence. Finally, family conflict over medical issues was related to a decrease in medical adherence over time. **Conclusions** Results suggest that as youth take more responsibility over their medical regimens, family conflict regarding medical issues becomes a contributor to their adherence behaviors. Interventions that target family conflict may facilitate adherence behaviors.

Key words spina bifida; adherence; adolescence; family conflict.

Medical adherence behaviors of children with acute or chronic medical conditions have been studied extensively by pediatric psychologists (La Greca & Bearman, 2003; Rapoff, 1999). Although substantial attention has been given to treatment regimens in children with chronic illnesses, considerably less attention has been given to adherence in children with physical disabilities, such as spina bifida (Holmbeck et al., 1998). Some children with physical disabilities need to manage complicated and demanding medical adherence regimens that produce substantial physical, mental, and emotional strain on themselves and their families.

Spina bifida occurs during the first month of pregnancy when the development of the spinal cord is interrupted, resulting in the failed closure of one or more vertebrae that surround the developing spinal cord (Liptak, 1997). The degree of motor and sensory disability varies, depending on a number of factors such as the location and severity of the spinal lesion. Most children have numerous medical problems, including neurological, orthopedic, urinary, and bowel difficulties, most of which require adherence to separate regimens (Liptak, 1997).

Although numerous studies have examined medical adherence in pediatric populations, there has been a scarcity of studies that examine the over-time transfer of medical regimen responsibility from parents to their children. In general, research on children with diabetes (Anderson, Auslander, Jung, Miller, & Santiago, 1990) and asthma (McQuaid et al., 2001) has shown that children assume increasing responsibility for their medical regimens with increasing age. While we expected that the transfer of responsibility may follow a similar pattern in youth with spina bifida, there are factors that make this population unique. Specifically, children with spina bifida are often required to adhere to intrusive procedures such as catheterization and specific bowel programs. Additionally, children with spina bifida often have cognitive deficits, which may make it more difficult for them to follow through with their regimens. Thus, on the one hand, parents may be likely to rapidly transfer these medical regimens to their children because of the intrusiveness of the procedures. On the other hand, parents may be less likely to do so because of their children's cognitive delays. Moreover, studies that have examined transfer of

All correspondence should be addressed to Grayson N. Holmbeck, Loyola University Chicago, Department of Psychology, 6525 N. Sheridan Road, Chicago, IL 60626, USA. E-mail: gholmbe@luc.edu

Journal of Pediatric Psychology 35(2) pp. 167–176, 2010

doi:10.1093/jpepsy/jsp054

Advance Access publication June 25, 2009

Journal of Pediatric Psychology vol. 35 no. 2 © The Author 2009. Published by Oxford University Press on behalf of the Society of Pediatric Psychology. All rights reserved. For permissions, please e-mail: journals.permissions@oxfordjournals.org

responsibility in pediatric populations have tended to examine this process cross-sectionally rather than longitudinally. Thus, one goal of this study was to explore the transfer of responsibility of medical regimens in youth with spina bifida from parent to child longitudinally as they approach adolescence.

Treatment adherence is important to study during adolescence, a stage characterized by dramatic physical, and psychological maturation as well as changes in family and peer relationships (Hill, Bromell, Tyson, & Flint, 2007). Ricker, Delamater, and Hsu (1998) discuss several issues regarding medical adherence during adolescence. First, the developmental strivings of adolescence, such as independence and individuation, may result in less optimal adherence to a demanding daily regimen. In addition, other developmental events such as socializing with peers or working at one's first job may interfere with the scheduling of regimen-related activities. Furthermore, the permanent and severe nature of the disease may become more evident and discouraging during adolescence. The realization that one's illness will continue even when one is fully adherent with the treatment regimen may be disappointing and result in nonadherence.

The second goal of this study was to examine familial factors that may predict treatment adherence for youth who are partially or fully responsible for their medical regimens. Family variables have been found to influence medical adherence (Wamboldt & Wamboldt, 2000). In a study of adolescents with spina bifida and cerebral palsy, Blum, Resnick, Nelson, and St Germaine (1991) found that adolescents were highly dependent on their parents. This high level of dependence on their parents, in combination with limited social interactions, makes it likely that the family will play a significant role in the management of the child's illness.

Past research suggests that family cohesion influences treatment adherence by offering support and supervision to the child (La Greca & Bearman, 2003). Other studies suggest that cohesive families affect treatment adherence in children because parents in highly cohesive homes may be more likely to establish family routines that incorporate the planning of disease management (Kell, Kliewer, Erickson, & Ohrene-Frempong, 1998), and these parents are more likely to establish adaptive rules and belief systems about health and illness in the family (Kazak, Rourke, & Crump, 2003). Finally, research with adolescents who have type 1 diabetes indicates that when youth appraise their parents as "collaborators" in resolving diabetes-related problems, they are more likely to exhibit successful adherence during adolescence (Wiebe et al., 2005).

Conversely, research has consistently linked family conflict to poor regimen adherence (Jacobson et al., 1994; Miller & Drotar, 2003). Such findings underscore the salience of parent-child tensions in disease management. The strain of the increased responsibilities and demands of the treatment regimen provide fertile ground for parent-child disagreements to arise. When conflict does develop, it may place a child at increased risk for poor adherence and potentially poor health outcomes.

In this study, reports from mothers, fathers, youth, teachers, and health professionals were utilized to measure the variables examined. Moreover, both observational and self-report measures were used to assess family cohesion and conflict. Finally, longitudinal data were used to assess whether family predictors at Time 1 were related to subsequent change in medical adherence from Time 1 (ages 8–9 years) to Time 2 (ages 10–11 years) and whether family predictors at Time 2 were related to subsequent change in medical adherence from Time 2 to Time 3 (ages 12–13 years). It was hypothesized that responsibility for medical regimens would transfer gradually from parent to child over time. Additionally, it was hypothesized that family cohesion would be positively associated with medical adherence at each time point (cross-sectionally), and that family conflict would be negatively associated with medical adherence at each time point (cross-sectionally). Similarly, higher levels of family cohesion and lower levels of family conflict were hypothesized to predict better medical adherence over time.

Methods

Participants

Participants in this study were part of a larger study supported by grants from the March of Dimes on the transition to adolescence in families of children with spina bifida (e.g., Friedman, Holmbeck, Jandasek, Zukerman, & Abad, 2004; Holmbeck et al., 1998, 2002). During the first data collection procedure, 70 families having children with spina bifida between 8 and 9 years of age were interviewed [39 males, 31 females; $M(\text{age}) = 8.33$]. Families were then interviewed every 2 years thereafter. Most of the child participants were White (82%; a rate that paralleled the prevalence rate for spina bifida in the general population at the time this sample was recruited, Hynd and Willis, 1988). Mothers from all families participated in the study; however, only 55 father/stepfathers participated. A wide range of family incomes was represented (9% had annual incomes of <\$20,000 a year; 58% had incomes between \$20,000 and \$60,000; and 28% reported incomes >\$60,000; 5% missing). Information

on a number of physical status variables was collected from maternal report or from the child's medical chart. Most of the participating children had spina bifida myelomeningocele ($n = 58$; lipomeningocele, $n = 8$; occulta and meningocele, $n = 4$). With respect to lesion level, 32% had sacral level lesions, 54% had lumbar level lesions, and 13% had thoracic level lesions. Most of the children had a shunt (71%), participated in some form of catheterization program (64%), and used braces (63%) or a wheelchair (18%) for ambulation (19% unassisted). Less than half of the mothers reported that their children participated in a bowel program (43%).

Sample Recruitment

Participants in this study were recruited from four sources: (1) a children's hospital, (2) a children's hospital that cares exclusively for youth with physical disabilities, (3) a university-based medical center, and (4) a statewide spina bifida association. Letters that requested the participation of the families were sent by mail. Out of 310 names received from the above sources, 72 families lived too far away to be contacted (more than 120 miles from the laboratory), 56 could not be contacted due to incorrect addresses and/or phone numbers, 64 declined to participate, 11 had children who did not have spina bifida, 14 had children who turned 10-years old before they could be scheduled for a family visit, 16 had parents and/or children who did not speak English, and 7 were eliminated for miscellaneous reasons, leaving 70 families. A comparison of the children in the participating families with the children from families that declined to participate revealed no differences with respect to lesion level [$\chi^2(2) = 0.62, p > .05$], or type of spina bifida [$\chi^2(1) = 1.63, p > .05$].

Procedure

At each time point, assessment of the parent and child participants involved a 3-hr home visit conducted by graduate- and undergraduate-level research assistants. Families were interviewed at Time 1 when the children were 8–9 years old and then again at Time 2 and Time 3, 2 and 4 years, respectively, after the first assessment session. Prior to conducting the visits, all research assistants were trained to administer the research protocol. At Time 1, data were collected from 70 families with spina bifida. At the second wave of data collection, Time 2, the retention rate was 99%, with one family declining to participate ($n = 69$). At Time 3, the retention rate was 96% with three additional families declining to participate ($n = 66$).

After signing consent and assent forms, family members were asked to complete packets of questionnaires independently. The families were then asked to participate in a series of videotaped interaction tasks. Two simple warm-up tasks were initially presented to the family, which were not coded for this study. The counterbalanced interaction tasks that were coded for the current study were as follows: an unfamiliar board game, the structured family interaction (SFIT) task (Ferreira, 1963), and a family conflict task (Smetana, Yau, Restrepo, & Braeges, 1991). Families were paid \$50, \$75, and \$100, respectively, for each time point. Teachers were paid \$5 for Times 1–2 and \$10 for Time 3. Health care professionals were paid \$2 at each data collection period.

Unfamiliar Board Game Task

For this task, families were asked to engage in a novel game. The game varied across T1, T2, and T3 and was unavailable for retail purchase. The interaction task required that families play the game with their own rules for 10 min.

Structured Family Interaction Task

Prior to the videotaped family interaction tasks, family members were asked to independently select their first and second choices from a list of issues commonly discussed by families (i.e. choosing a restaurant, deciding upon a television show, etc.). Identical questions were then presented during the family interaction tasks and each participant was asked to consider the perspective of other family members when coming to a single family decision.

Family Conflict Task

Prior to participating in the interaction task, the Parent-Adolescent Conflict (PAC) scale was completed by mother, father, and child. This 20-item questionnaire is a brief version of the Issues Checklist (Robin & Foster, 1989) and includes items that tap potential conflicts. Some items focus on conflict surrounding medical-related issues (five items, such as how s/he does her/his catheterization), and others focus on general issues that were nonmedical in nature (15 items, such as whether my child does chores around the house). First, the individual decides whether or not that particular issue was brought up in the last 2 weeks, and marks "yes" or "no" accordingly. Then, the individual indicates the frequency with which the issue was discussed. For each "yes" response, the respondent rates the intensity of affect associated with the discussions on a Likert scale from one (calm) to five (angry). After the respondents completed the PAC scale,

scores were computed for each item by multiplying conflict frequency by intensity. Items with the five highest scores across respondents were selected for the conflict task. The family was asked to choose three of the five items and then discuss the issues, incorporating different family members' points of view, for a total of 10 min.

Demographic Data

The following data were obtained from responses by the parents: gender of the child, ethnicity of family members, socio-economic status (i.e. occupation, educational attainment, and yearly income of adults in the household), family structure, and prior contacts with mental health, medical and special education services.

Questionnaire Measures: Family Predictors

Family Conflict

The intensity of parent-child conflicts was assessed using the PAC (as discussed earlier; Robin & Foster, 1989). For the current study, the mean intensity rating across family members was employed as a measure of familial conflict. Because intensity ratings were only required for items where discussions were reported, a reliability α coefficient could not be computed for this variable because reliability software requires that the same items be completed by all the participants. Similarly, correlations between respondents are not reported (and were not expected to be significant) because a total score for a given respondent may be based on different items than the total score for another respondent (even when the other respondent is from the same family; e.g., mother and child may disagree on what issues have caused conflict in the family in the last 2 weeks, which would cause their conflict intensity scores to be based on different conflict issues).

Family Cohesion

The Family Environment Scale (FES; Moos & Moos, 1986) is a 90-item self-report measure of perceived family social environment that yields 10 subscales; the cohesion subscale was composed of nine items. At Time 1, parents were asked whether each statement described their family by replying "true" or "false". Prior to collecting the Time 2 data, we converted the response option scale for the FES from a true-false scale to a 4-point Likert scale for two reasons. First, the Cronbach α for some of the FES scales were low (<0.60) at Time 1, which is common when using true-false scales. Second, the literature on the FES has found similar low α for some scales when using the true-false response strategy (e.g. Roosa & Beals, 1990; Sanford, Bingham, & Zucker, 1999). With the Likert scale, response options ranged from

strongly disagree (1) to strongly agree (4). In support of changing to a Likert scale version, correlations across time were high: for mothers and fathers, the across-time correlations involving Time 1 (T1 with T2 and T1 with T3) ranged from .41, $p < .01$, to .52, $p < .01$. The Cohesion subscale also demonstrated adequate internal consistency (α ranged from 0.68 to 0.77 across respondents and time points).

Observational Measures: Family Predictors

The videotaped tasks were coded using a macrocoding scheme developed for this project by Holmbeck, Belvedere, Gorey-Ferguson, and Schneider (1995), which was based on a system developed by Smetana et al. (1991). Coders were trained for approximately 10 hours and were required to achieve at least 90% reliability with an expert coder during training. Coders viewed one interaction task at a time and then rated it on a variety of family interaction variables using a 5-point Likert scale. Each score was based on the mean of the scores provided by two separate coders. Inter-rater reliability was assessed at Time 1 with intraclass correlations and were found to be satisfactory (0.86 for family cohesion and 0.74 for family conflict).

Cohesion and conflict were examined using the same codes as McKernon and colleagues (2001). Cohesion was measured using the following codes: (1) requests input from others; (2) comfort level during interaction; (3) involvement in task; (4) parents present a united front; (5) parental promotion of dialogue and collaboration; and (6) four measures of general family atmosphere [disengaged (reverse scored), openness, depression (reverse scored), and able to reach an agreement]. Conflict was measured using the following codes: (1) level of conflict within dyads for mother-child and father-child; (2) frequently disagrees with others; and (3) attempted resolution of issues (reverse scored). Scale α for cohesion and conflict were 0.73 and 0.85, respectively, for Time 1, 0.79 and 0.87 for Time 2, and 0.69 and 0.90 for Time 3.

Questionnaire Measures: Adherence Outcomes

Responsibility for Medical Regimens

Mothers were asked to indicate whether their child had a catheterization schedule or a bowel program and whether their child did these tasks: (1) independently without reminding, (2) independently with reminding, (3) with partial assistance, or (4) with complete assistance.

Medical Adherence: Parent Report

The parent report of medical adherence in spina bifida scale (PROMASB, Holmbeck et al., 1998) includes

39-Likert scale items focused on spina bifida related tasks (i.e. catheterization, bowel care, skin care, medication, and ambulation). The PROMASB has demonstrated adequate psychometric properties as well as adequate variability for all scales (Holmbeck et al., 1998). For this study, adherence to the child's catheterization program and the overall level of adherence were assessed across all medical tasks. Questions focused on several dimensions of catheterization: accuracy, regularity, difficulty in achieving adherence, parental reminding, etc. (Holmbeck et al., 1998). A general adherence item was also included (e.g. "Overall, my child is compliant with his/her medical regimen"). All items were keyed in the direction of greater adherence. Internal consistency for the catheterization subscale was adequately demonstrated for mother report (Cronbach's α Time 1 = 0.76, Time 2 = 0.80, Time 3 = 0.82) and father report (Cronbach's α Time 1 = 0.73, Time 2 = 0.78, Time 3 = 0.85).

Medical Adherence: Teacher Report

A 13-item version of the PROMASB was administered to teachers (TROMASB, Holmbeck et al., 1998). For this study, adherence to catheterization and their overall compliance was examined. Questions focused on the following four dimensions of catheterization: accuracy, difficulty in achieving adherence in the child, teacher reminding, and the frequency of teacher help for the child. In addition to these four task-specific items, an overall compliance item was included in this scale (e.g. "Overall, this child is compliant with his/her medical regimen"). To establish adequate internal consistency, one item was removed ("I often have to help this child with his/her catheterization"). Cronbach's α were as follows: Time 1 = 0.88, Time 2 = 0.57, Time 3 = 0.69. Given the low α at Time 2, teacher-report data from this time point were dropped and not included in the analyses.

Medical Adherence: Health Professional Report

Health professionals completed a 5-item Likert scale adherence measure, which included an assessment of the child's adherence to their catheterization program (one item) and their overall compliance (one item). Responses ranged from never (1) to always (5).

Results

Preliminary Analyses

To decrease the number of analyses, responses for medical adherence that were highly correlated were combined. In general, correlations between mother and father reports of medical adherence were consistently high ($M = 0.36$;

range = 0.10–0.57), as were correlations between teacher and health professional reports ($M = 0.35$; range = 0.28–0.42). Thus, scores for these respondent dyads were combined for the analyses. Additionally, mother and father reports on the cohesion questionnaire were consistently high and, therefore, were combined (range = 0.38–0.50). Finally, mother, father, and child reports on the conflict questionnaire were also combined (as discussed earlier concerning associations between respondents for this scale). Since the scale metric was identical across respondents, all within-respondent scale totals were computed as item means and then the mean across all available respondents was computed. In other words, if a family had conflict scores for mother and child report only, the composite score represented the mean of the conflict scores for mother and child report. In this way, the across-respondent mean represented the mean of all available data for a given family at that data collection point. Because of the small sample size, we did not include any corrections for Type I errors (e.g. reducing the criterion for statistical significance from 0.05 to 0.01). On the other hand, we present effect sizes for the correlational and regression analyses as reported further (Cohen, 1992).

Transfer of Responsibility over Time

As expected, the number of child participants who were partially or fully responsible for their medical regimens increased over time (see Table I). The transfer of responsibility was measured for their catheterization and bowel programs. The transfer of responsibility was more evident for the catheterization programs with only two participants having their parents fully responsible for their care by Time 3 when the children were 12- and 13-years old. In contrast, about half of the participants still had their parents fully responsible for their bowel programs by Time 3. Thus, subsequent cross-sectional and longitudinal analyses were based on youth partially or fully responsible for

Table I. Examining Transfer of Responsibility over Time^a

	Time 1	Time 2	Time 3
Catheterization parent fully responsible	14	2	2
Catheterization child partially responsible	26	31	25
Catheterization child fully responsible	0	6	14
Total	40	39	41
Bowel program parent fully responsible	23	16	13
Bowel program child partially responsible	7	15	8
Bowel program child fully responsible	0	1	4
Total	30	32	25

^aTable reflective only of individuals on a catheterization or bowel program. Those not on a program were excluded.

their catheterization program, given that the number of participants was low for those partially or fully adherent to their bowel program.

Cross-Sectional Correlational Analyses

Cross-sectional analyses were conducted to examine associations between the family variables and medical adherence for catheterization. Because we sought to focus on the child's level of adherence, these correlational analyses were conducted only for families where the child was reported to be partially or fully responsible for their medical regimen. The cross-sectional correlations between the family variables and medical adherence are presented in Table II. At Time 1, significant positive correlations were found between the FES and parent report of catheterization adherence ($r = .48, p < .05$) and between observed family cohesion and parent report of general adherence ($r = .43, p < .05$). Additionally, significant negative correlations were found between the PAC for nonmedical issues and parent report of general adherence ($r = -.41, p < .05$) and between observed family conflict and parent report of general adherence ($r = -.046, p < .05$).

At Time 2, only the FES was significantly correlated with parent report of general adherence ($r = .37, p < .05$; see Table III). Finally, at Time 3, positive correlations

were found between the FES and parent report of general adherence ($r = .35, p < .05$) and between observed family cohesion and parent report of general adherence ($r = .44, p = .01$; see Table II). Significant negative correlations were found between the PAC for medical issues and parent report of general adherence ($r = -.46, p < .01$) as well as between the PAC for nonmedical issues and parent report of general adherence ($r = -.58, p < .01$). Thus, the highest correlations were found between family functioning and adherence at Time 3, when most of the children were fully or partially adherent to their regimen and when they were transitioning into the early adolescent period (i.e. 12–13 years of age). With respect to effect sizes, the significant correlations in Table II ranged from .365 to .576, which represent medium (.30) to large (.50) effect sizes, according to Cohen (1992).

Longitudinal Regression Analyses

A series of longitudinal regression analyses were conducted to determine if family functioning was related to subsequent change in medical adherence over time. Similar to the correlational analyses, longitudinal analyses were conducted only for families where the child was reported to be partially or fully responsible for their medical regimen. To control for medical adherence at the previous

Table II. Cross-sectional Analyses of Time 1 to Time 3

Independent variables	Dependent variables	Time 1 <i>r</i> (<i>n</i>)	Time 2 <i>r</i> (<i>n</i>)	Time 3 <i>r</i> (<i>n</i>)
1. Conflict medical questionnaire (M-F-C)	M-F catheterization	.178 (25)	-.111 (37)	-.036 (36)
	T-HP catheterization	-.135 (25)		-.308 (35)
	M-F general	-.381 (25)	.012 (37)	-.456** (35)
	T-HP general	-.016 (25)		-.010 (36)
2. Conflict nonmedical questionnaire (M-F-C)	M-F catheterization	.025 (26)	-.075 (37)	-.250 (39)
	T-HP catheterization	-.182 (26)		-.290 (38)
	M-F general	-.409* (26)	-.059 (37)	-.576** (37)
	T-HP general	-.026 (26)		-.059 (39)
3. Family cohesion questionnaire (M-F)	M-F catheterization	.479* (26)	.188 (36)	-.041 (39)
	T-HP catheterization	-.210 (26)		.005 (38)
	M-F general	.089 (26)	.365* (36)	.353* (37)
	T-HP general	-.065 (26)		-.065 (39)
4. Family conflict observational	M-F catheterization	.009 (26)	-.118 (33)	.086 (35)
	T-HP catheterization	-.057 (26)		-.130 (34)
	M-F general	-.461* (26)	-.282 (33)	-.224 (33)
	T-HP general	-.269 (26)		.153 (35)
5. Family cohesion observational	M-F catheterization	-.248 (26)	-.081 (33)	-.071 (35)
	T-HP catheterization	.095 (26)		.132 (34)
	M-F general	.434* (26)	.004 (33)	.441* (33)
	T-HP general	.191 (26)		.203 (35)

Note. Time 2 Teacher reports were dropped due to low alphas. M-F-C, mother, father, child report composite; M-F, mother and father report composite; T-HP, teacher and health professional report composite.

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

Table III. Longitudinal Analyses: Family Variables Predicting Adherence

Independent variables	Dependent variables	T1→T2		T2→T3	
		β	$R^2\Delta$	β	$R^2\Delta$
1. Conflict medical questionnaire (M-F-C)	M-F catheterization	0.119	.014	-.337	.113*
	T-HP catheterization			-.305	.091*
	M-F general	.293	.083	-.403	.168*
	T-HP general			-.032	.001
2. Conflict nonmedical questionnaire (M-F-C)	M-F catheterization	.023	.001	-.111	.012
	T-HP catheterization			-.016	.000
	M-F general	.113	.011	-.252	.059
	T-HP general			.110	.012
3. Family cohesion questionnaire (M-F)	M-F catheterization	-.042	.002	-.085	.007
	T-HP catheterization			-.163	.026
	M-F general	.126	.016	-.148	.019
	T-HP general			-.177	.030
4. Family conflict observational	M-F catheterization	-.012	.000	-.064	.004
	T-HP catheterization			.054	.003
	M-F general	-.100	.009	-.267	.060
	T-HP general			-.180	.029
5. Family cohesion observational	M-F catheterization	-.096	.009	.116	.013
	T-HP catheterization			.051	.002
	M-F general	-.184	.034	.008	.000
	T-HP general			.124	.014

Note. Time 2 Teacher reports were dropped due to low alphas. M-F-C, mother, father, child report composite; M-F, mother and father report composite; T-HP, teacher and health professional report composite.

* $p < .05$.

time point, this variable (either medical adherence for catheterization or general medical adherence) was entered at the first step. Thus, the analysis assessed the effect of family functioning on change in medical adherence over time. When Time 1 family functioning measures were used to predict medical adherence at Time 2, no significant effects were found (see Table III). However, when Time 2 family functioning measures were used to predict medical adherence at Time 3, the PAC for medical issues consistently predicted medical adherence, such that higher levels of family conflict predicted a decrease in medical adherence over time (parent report of catheterization, $\beta = -0.337$, $p < .05$; parent report of general adherence, $\beta = -0.403$, $p < .05$; and teacher/health professional report of catheterization, $\beta = -0.305$, $p < .05$). Effect sizes for these R^2 -change findings (range = .091–.168) suggested that the significant longitudinal effects in Table III ranged from small to medium (Cohen, 1992).

Discussion

The purpose of this study was twofold (1) to explore the transfer of responsibility for medical tasks from parent to child over time, and (2) to examine cross-sectional and

longitudinal associations between family functioning variables and medical adherence in youth with spina bifida who were either partially or fully responsible for their medical regimens. The current study addressed a number of weaknesses in past research by including father data in addition to mother data, observational assessments of family functioning in addition to questionnaire measures, and using multiple informants to assess medical adherence.

This study showed that children with spina bifida achieve increased responsibility for their medical regimens with increasing age. Studies of other illness populations have noted that the transfer of responsibility from parent to child when the child is not yet developmentally ready to take on these tasks may prevent the development of appropriate self-care (Wysocki et al., 1996). Alternatively, not allowing children to be responsible for their self-care when they are older and developmentally able to do so may stifle their autonomy development and encourage an excessive dependence on their parents. The latter scenario may be particularly likely for children with spina bifida, given that their autonomy may often be compromised by higher levels of intrusive parenting (Holmbeck et al., 2002). Thus, it will be important for future studies to

examine how ready the child is to assume adherence responsibilities.

Support was also found for the hypothesis that family functioning would be significantly associated with medical adherence, with all results being in the directions predicted. That is, family cohesion was found to be correlated positively with medical adherence at each time point, while family conflict was negatively correlated with medical adherence at Time 1 and Time 3. Additionally, longitudinal results showed that conflict surrounding medical issues at Time 2 was associated with a decrease in adherence from Time 2 to Time 3, across reporters and type of adherence assessed.

These cross-sectional and longitudinal results for associations between family conflict and adherence are consistent with a developmental perspective on adherence in children with spina bifida. At Time 3, children are entering early adolescence (ages 12 and 13 years), and parent and child views of autonomy may begin to diverge (Holmbeck, 1996). To assert their autonomy, these adolescents may begin to rethink the legitimacy of their parents' authority over various medical regimen tasks, which may lead to an increase in conflict and non-adherence (Holmbeck, 1996). There were more significant associations between conflict and adherence at Time 3 than at Time 1 or Time 2, and these associations emerged across type of conflict (i.e. medical and nonmedical). In other words, conflict and adherence appear to have become increasingly intertwined during early adolescence.

In this study, relatively few findings emerged for the observational predictors. We offer several reasons for this relative lack of significant effects. First, families were only observed for a short period of time. Second, while the interaction tasks were designed to elicit discussions that children and parents typically have regarding spina bifida related behaviors, there was no guarantee that such topics were discussed. As such, it is possible that the interactions were not relevant to adherence and were instead focused on conflict over topics such as homework or television watching. Future studies will benefit from the inclusion of observational measures that are based on more extended family interactions, or on interactions centered around adherence-related behaviors.

In addition to family functioning influencing medical adherence, other variables such as illness severity and cognitive functioning may influence adherence behaviors in children with spina bifida. Such variables should receive more attention in future research. Specifically, neuropsychological factors such as executive functioning or memory skills may have direct implications for adherence behaviors, as these would relate to remembering and organizing

details surrounding adherence regimens. Interestingly, we were unable to find any studies that examined associations between cognitive functioning and adherence behaviors in youth with spina bifida, despite a seemingly apparent relationship.

This study had several limitations that should be addressed in future studies focused on associations between family functioning and medical adherence in this population. First, the current study only examined the transfer of responsibility from the mother's perspective. Other studies have noted that there are discrepancies in the degree to which child and parent are in agreement regarding who is in charge of adherence tasks (Anderson et al., 1990). Second, it may be necessary to examine specific components of cohesion that have an impact on medical adherence. For example, Fiese and Wamboldt (2000) have maintained that there are at least two aspects of family cohesion that may affect medical adherence (i.e. routine practices, ritual importance, etc.). Third, adherence was assessed only with self-report. Although the inclusion of teacher and health professional report was a strength of this study, their reports are still partly based on information that parents provide (Rapoff, 1999). An alternative to questionnaire reports of adherence would be to include observational methods such as self-monitoring or daily logs (La Greca & Bearman, 2003). Fourth, because we conducted numerous analyses on a relatively small sample, the results should be interpreted cautiously. Fifth, the sample was homogenous and included primarily White, English-speaking participants. Future studies would benefit from a more heterogeneous sample, particularly one that includes more Latino families, given the relatively high rate of spina bifida in this population (Lary & Edmonds, 1996). Finally, there is a lack of normative and psychometric data for the medical adherence measure used in this study (i.e. PROMASB; Holmbeck, et al., 1998).

From a clinical perspective, the findings of this study have implications for potential interventions targeted at how families manage the transition into adolescence and the transfer of medical regimen responsibilities from parent to child. It may be useful for healthcare workers to be cognizant of the quality of a child's family functioning and to include specific questions about family conflict and cohesion as part of a comprehensive evaluation of the patient. Although it is important for physicians and healthcare providers to support parents in facilitating self-reliance in their children, it is equally important for medical professionals to ensure that this transfer of responsibility is completed in an atmosphere that will elicit effective adherence on the part of the child.

Clinicians should be aware that conflict manifests itself in different ways during different stages of youth development. Although conflict may impede successful adherence behaviors on the part of the child/adolescent, it is also important to recognize that these sometimes stressful parent–child interactions are part of the typical developmental transition from childhood to adolescence.

Acknowledgements

The authors wish to thank Ann Walsh Johnson, Joy Ito, Pat McGovern, Pat Braun, Caroline Anderson, David McLone, John Lubicky, the Spina Bifida Association of Illinois, the staff of the spina bifida clinics at Children's Memorial Hospital, Shriners Hospitals for Children-Chicago, and Loyola University of Chicago Medical Center. We also thank numerous undergraduate and graduate research assistants for their help in data collection and data entry. Most importantly, we gratefully acknowledge the contributions to this study by the parents, children, teachers, and health professionals who participated over many years.

Funding

March of Dimes Birth Defects Foundation (12-FY01-0098, partial); the National Institute of Child Health and Human Development (RO1 HD048629).

Conflict of interest: None declared.

Received October 16, 2008; revisions received May 20, 2009; accepted May 31, 2009

References

- Anderson, B. J., Auslander, W. F., Jung, K. C., Miller, J. P., & Santiago, J. V. (1990). Assessing family sharing of diabetes responsibilities. *Journal of Pediatric Psychology, 15*, 477–492.
- Blum, R. W., Resnick, M. D., Nelson, R., & St. Germaine, A. (1991). Family and peer issues among adolescents with spina bifida and cerebral palsy. *Pediatrics, 88*, 280–285.
- Cohen, J. (1992). A power primer. *Psychological Bulletin, 112*, 155–159.
- Ferreira, A. J. (1963). Decision making in normal and pathological families. *Archives of General Psychiatry, 8*, 68–73.
- Fiese, B. H., & Wamboldt, F. S. (2000). Family routines, rituals and asthma management: A proposal for family-based strategies to increase treatment adherence. *Families, Systems & Health, 18*, 405–418.
- Friedman, D., Holmbeck, G. N., Jandasek, B., Zukerman, J., & Abad, M. (2004). Parent functioning in families of preadolescents with spina bifida: Longitudinal implications for child adjustment. *Journal of Family Psychology, 18*, 609–619.
- Hill, N.E., Bromell, L., Tyson, D. F., & Flint, R. (2007). Developmental commentary: Ecological perspectives on parental influences during adolescence. *Journal of Clinical Child and Adolescent Psychology, 36*, 367–377.
- Holmbeck, G. N. (1996). A model of family relational transformation during the transition to adolescence: Parent-adolescent conflict and adaptation. In J. A. Graber, J. Brooks-Gunn, & A. C. Petersen (Eds.), *Transitions through adolescence: Interpersonal domains and context* (pp. 167–199). Hillsdale, NJ: Lawrence Erlbaum Associates, Inc.
- Holmbeck, G. N., Belvedere, M. C., Christensen, M., Czerwinski, A. M., Hommeyer, J. S., Johnson, S. Z., et al. (1998). Assessment of adherence with multiple informants in pre-adolescents with spina bifida: Initial development of a multidimensional, multitask parent-report questionnaire. *Journal of Personality Assessment, 70*, 427–440.
- Holmbeck, G. N., Belvedere, M., Gorey-Ferguson, L., & Schneider, J. (1995). *Family macro-coding manual—March of Dimes triadic version*. Unpublished Manual, Loyola University of Chicago, Department of Psychology.
- Holmbeck, G. N., Johnson, S. Z., Wills, K. E., McKernon, W., Rose, B., Erkin, S., et al. (2002). Observed and perceived parental overprotection in relation to psychosocial adjustment in preadolescents with a physical disability: The mediational role of behavioral autonomy. *Journal of Consulting and Clinical Psychology, 70*, 96–110.
- Hynd, G. W., & Willis, W. G. (1988). *Pediatric Neuropsychology*. New York, NY: Gune & Stratton.
- Jacobson, A. M., Hauser, S. T., Lavori, P., Willett, J. B., Cole, C. F., Wolfsdorf, J. I., et al. (1994). Family environment and glycemic control: A four-year prospective study of children and adolescents with insulin-dependent diabetes mellitus. *Psychosomatic Medicine, 56*, 401–409.
- Kazak, A. E., Rourke, M. T., & Crump, T. A. (2003). Families and other systems in pediatric psychology. In M. C. Roberts (Ed.), *The handbook of pediatric psychology* (3rd ed., pp. 159–175). New York, NY: Guilford Press.

- Kell, R. S., Kliever, W., Erickson, M. T., & Ohrene-Frempong, K. (1998). Psychological adjustment of adolescents with sickle cell disease: Relations with demographic, medical, and family competence variables. *Journal of Pediatric Psychology, 23*, 301–312.
- La Greca, A. M., & Bearman, K. J. (2003). Adherence to pediatric treatment regimens. In M. C. Roberts (Ed.), *Handbook of pediatric psychology* (3rd ed., pp. 119–140). New York: Guilford Press.
- Lary, J. M., & Edmonds, L. D. (1996). Prevalence of spina bifida at birth—United States, 1983–1990: A comparison of two surveillance systems. *Morbidity and Mortality Weekly Report, 45*, 15–26.
- Liptack, G. S. (1997). Neural tube defects. In M. L. Batshaw, & Y. M. Perret (Eds.), *Children with disabilities: A medical primer* (pp. 429–552). Baltimore, MD: Brookes.
- McKernon, W. L., Holmbeck, G. N., Colder, C. R., Hommeyer, J. S., Shapera, W., & Westhoven, V. (2001). Longitudinal study of observed and perceived family influences on problem-focused coping behaviors of preadolescents with spina bifida. *Journal of Pediatric Psychology, 26*, 41–54.
- McQuaid, E. L., Penza-Clyve, S. M., Nassau, J. H., Fritz, G. K., Klein, R., O'Connor, S., et al. (2001). The asthma responsibility questionnaire: Patterns of family responsibility for asthma management. *Children's Health Care, 30*, 183–199.
- Miller, V. A., & Drotar, D. (2003). Discrepancies between mother and adolescent perceptions of diabetes-related decision-making autonomy and their relationship to diabetes-related conflict and adherence to treatment. *Journal of Pediatric Psychology, 28*, 265–274.
- Moos, R. H., & Moos, B. S. (1986). edn.). *Family Environment Scale Manual* (2nd ed.). Palo Alto, CA: Consulting Psychologists Press, Inc.
- Rapoff, M.A. (1999). *Adherence to pediatric medical regimens*. New York: Kluwer Academic.
- Ricker, J. H., Delamater, A. M., & Hsu, J. (1998). Correlates of regimen adherence in cystic fibrosis. *Journal of Clinical Psychology in Medical Settings, 5*, 159–172.
- Robin, A. L., & Foster, S. L. (1989). *Negotiating parent-adolescent conflict: A belief-family systems approach*. New York: Guilford.
- Roosa, M. W., & Beals, J. (1990). Measurement issues in family assessment: The case of the Family Environment Scale. *Family Process, 29*, 191–198.
- Sanford, K., Bingham, C. R., & Zucker, R. A. (1999). Validity issues with the Family Environment Scale: Psychometric resolution and research application with alcoholic families. *Psychological Assessment, 11*, 315–325.
- Smetana, J. G., Yau, J., Restrepo, A., & Braegas, J. L. (1991). Adolescent-parent conflict in married and divorced families. *Developmental Psychology, 27*, 1000–1010.
- Wamboldt, M. Z., & Wamboldt, F. S. (2000). Role of the family in the onset and outcome of childhood disorders: Selected research findings. *Journal of the American Academy of Child & Adolescent Psychiatry, 39*, 1212–1219.
- Wiebe, D. J., Berg, C. A., Korbel, C., Palmer, D. L., Beveridge, R. M., Upchurch, R., et al. (2005). Children's appraisals of maternal involvement in coping with diabetes: Enhancing our understanding of adherence, metabolic control, and quality of life across adolescence. *Journal of Pediatric Psychology, 30*, 167–178.
- Wyssocki, T., Taylor, A., Hough, B. S., Linscheid, T. R., Yeates, K. O., & Naglieri, J. A. (1996). Deviation from developmentally appropriate self-care autonomy: Association with diabetes outcomes. *Diabetes Care, 19*, 119–125.