

Factors Associated With the Psychological and Behavioral Adjustment of Siblings of Youths With Spina Bifida

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The intensive health management activities associated with spina bifida (SB) often command a pervasive presence in the lives of families and may place some members at risk for psychosocial difficulties. However, research on the risk and protective factors associated with sibling adjustment is limited. Anchored upon a social-ecological theoretical framework, the purpose of this cross-sectional study was to explore the relationships between select individual, family, and peer factors and sibling adjustment. A convenience sample of 224 adolescent siblings and parents of youths with SB

responded to anonymous mailed surveys. Siblings completed measures of attitude toward spina bifida, family satisfaction, warmth and conflict in the sibling relationship, peer support, and three dimensions of adjustment—self-concept, prosocial behavior, and behavior problems. Parents provided SB clinical data and family demographics. The individual, family, and peer factors explained a significant amount of variance in sibling self-concept, prosocial behavior, and behavior difficulties, with R^2 ranging from .27 to .57. Differing patterns of the relationships between the ecological factors and sibling adjustment emerged. Findings highlight multi-level opportunities to intervene and support siblings. Minimizing threats and bolstering protective influences on sibling adjustment is an essential component of family-centered services in SB.

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This work was funded in part by the National Institutes of Health Predoctoral Fellowship for Students with Disabilities No. F31HD047998-01A1; National Association of Social Workers Foundation Jane B. Aron Doctoral Fellowship; Ambulatory Pediatrics Association Region IV; and Virginia Organization of Health care Social Workers. We thank Suzanne Feetham, Patrick Dattalo, Donna Harrington, and Bruce DeForge for their helpful reviews of this article. We also acknowledge the Spina Bifida Association and families of youths with spina bifida for their support of this research.

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Keywords: sibling adjustment, spina bifida, family-centered care, social-ecology theory

Regarded as one of the most complex chronic health conditions affecting children, spina bifida (SB) develops from the failure of the neural tube to close early in pregnancy and is associated with a broad range of health complications, including hydrocephalus, lower extremity weakness or paralysis, neurogenic bladder,

bowel dysfunction, and neurocognitive deficits (Bowman, McLone, Grant, Tomita, & Ito, 2001; Holmbeck, Greenley, Coakley, Greco, & Hagstrom, 2006; Mitchell et al., 2004). The intensive health management activities associated with SB often command a pervasive presence in the lives of families (Bellin, Sawin, Roux, Buran, & Brei, 2007; Sawin, Bellin, et al., 2003) and may place some members at risk for psychosocial difficulties (Vermaes, Janssens, Bosman, & Gerris, 2005). Consequently, a family-centered approach to health care services that supports adaptive functioning of all family members may be helpful when working with this population.

A family-centered model of health care delivery that emphasizes partnership and shared decision-making between providers and the family has been linked to positive outcomes for both the child with the chronic condition (CHC) and parents (American Academy of Pediatrics, 2003). This model further recognizes that the psychosocial health of the child is interrelated with that of surrounding family members (Witt, Riley, & Coiro, 2003). Although the experience of parents of youths with SB is well documented (Kazak et al., 1997; Vermaes et al., 2005), research on the psychological and behavioral adjustment of siblings is limited, particularly with regard to the relationship between risk and protective factors and adjustment outcomes. Yet, because of decreased parent emotional and/or physical availability, insufficient resources supporting siblings, and lack of access to information about the CHC, siblings may be at increased risk for depression, anxiety, and behavioral problems (Sharpe & Rossiter, 2002; Williams et al., 1997). The limited research with siblings of youths with SB has shown mixed findings: some studies endorse a heightened risk for difficulty in this population (Tew & Lawrence, 1973), with other reports fail to support an elevated risk status (Kazak & Clark, 1986). An enhanced understanding of factors which increase vulnerability to

adjustment problems as well as those which promote age-appropriate, normative functioning is needed to advance the repertoire of intervention tools available to use in work with siblings. Anchored upon a social-ecological theoretical framework, the intent of this study was to increase understanding of the relationships between select individual, family, and peer factors and the psychological and behavioral adjustment of siblings of youths with SB—knowledge that may help inform the development of family-centered intervention services.

THEORETICAL FRAMEWORK

Social ecology theory recognizes diverse individual and contextual influences on human behavior (Bronfenbrenner, 1979, 2004). This holistic model places developing youths in the context of larger, interdependent social systems of influence. The child is seen as being “nested” within the family, neighborhood, and broader community settings, each of which may influence the child’s developmental trajectory (Fraser, 2004). Bronfenbrenner (1979) explained the person-environment interdependence as the “progressive, mutual accommodation between an active, growing human being and the changing properties of the immediate settings in which the developing person lives, as this process is affected by relations between these settings, and by the larger contexts in which the settings are embedded” (p. 21).

Examining child adjustment without accounting for the family context, or influences found in the surrounding social environment, will therefore yield only a partial picture of the resources and stressors impacting the child’s functioning (Fraser, 2004). Alternatively, because it places the child in the center of surrounding concentric circles of influence, social ecology theory captures the impact of multilevel factors on child adjustment outcomes (Bronfenbrenner, 1979, 2004). A social ecological framework further suggests

that *risk factors* that heighten vulnerability to adjustment problems and *protective factors* that mitigate the effects of the adversity may be found at the individual, family, and environmental levels, and often occur simultaneously (Fraser, Kirby, & Smokowski, 2004).

LITERATURE REVIEW: IDENTIFICATION OF ECOLOGICAL FACTORS INFLUENCING SIBLING ADJUSTMENT

Sibling research has been conducted across a range of health conditions and has evaluated the impact of diverse factors on adjustment outcomes. In particular gender, birth order, and condition severity have been frequently explored as correlates of sibling outcomes. However, in general, no consistent picture of the effects of these factors emerges. For example, some research supports increased risk of poor adjustment in male siblings (Hastings, 2003), but other studies failed to show differences in outcomes across gender (Kaminsky & Dewey, 2002). Research exploring the influence of birth order on psychosocial outcomes has also produced discordant findings, because some studies endorse a greater risk for difficulty in siblings younger than the child with the chronic condition (Gallo, Breitmayer, Knafl, & Zoeller, 1992), and others suggest an elevated risk for older siblings (Labay & Walco, 2004). Mixed findings have likewise been observed with regard to the relationship between condition severity and sibling adjustment (see, e.g., Dyson, 1989; Tew & Laurence, 1973). Identifying demographic and clinical factors associated with sibling adjustment is an important step to screen for siblings who may be at risk for problematic outcomes. However, as illustrated in Figure 1, a social-ecological perspective of sibling adjustment broadens the lens of focus to include other individual, family, and environmental factors possibly underlying the observed diversity in outcomes. A review of the sibling literature identified attitude toward disability, family satisfac-

tion, sibling relationship qualities, and peer support as influential factors among some populations.

Individual-Level Factor: Attitude Toward Disability

Austin and Huberty (1993) proposed an association between a positive attitude about the impact of a CHC and adaptive adjustment outcomes. Although the relationship is well documented among youths with SB (Sawin, Brei, Buran, & Fastenau, 2002; Sawin, Buran, Brei, & Fastenau, 2003), linkages between sibling attitude toward disability and adjustment have been understudied. However, noncategorical research with 62 siblings of youths with a range of chronic conditions found more negative attitudes about the condition were associated with worse adjustment outcomes (Taylor, Fuggle, & Charman, 2001).

Family-Level Factors: Family Satisfaction and Sibling Relationship Qualities

Characteristics of the family environment have also been explored as correlates of sibling adjustment outcomes. Family cohesion, affective expression, shared decision-making, and problem-solving have been showcased as important dimensions of family functioning (Patterson & Garwick, 1994). These qualities were associated with positive outcomes in a sample of 25 siblings of youths with cancer (Horwitz & Kazak, 1990), as well as in research with 22 siblings of youths with a CHC (Williams et al., 1999). Research has also revealed connections between the quality of sibling interactions and adjustment outcomes for some populations. In a study of factors associated with sibling adjustment, sibling relationships characterized by warmth and closeness were associated with positive outcomes for siblings of youths with Down's syndrome ($N = 45$) but not for siblings of youths with a pervasive developmental disorder ($N = 46$) (Fisman et al., 1996).

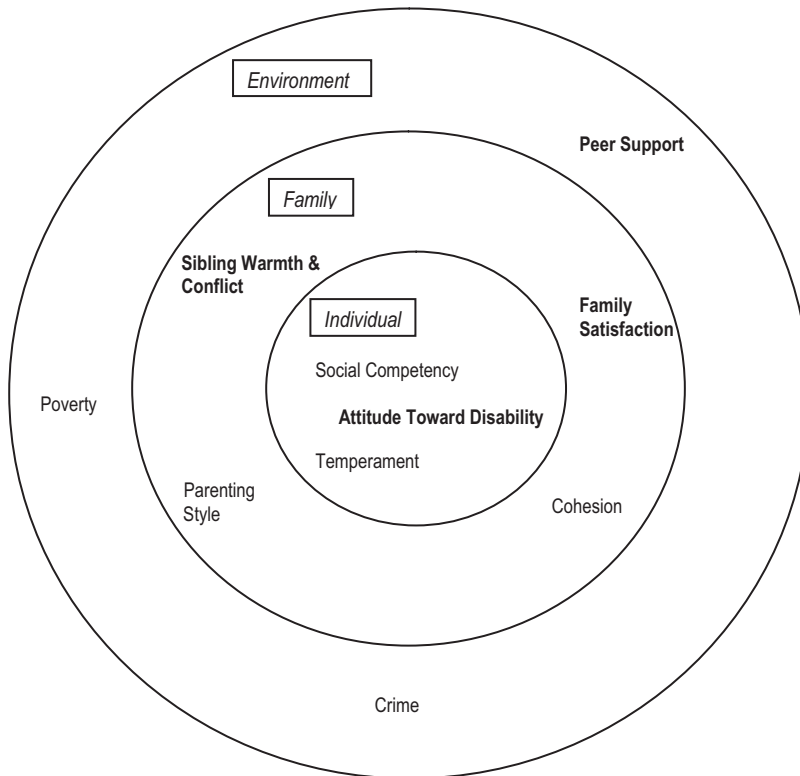


Figure 1. Application of a social-ecological framework to the study of sibling adjustment. The framework is based on the work of Bronfenbrenner (1979) and Fraser (2004). Ecological factors explored in the current study are noted in bold.

Environmental-Level Factor: Peer Support

A third layer of risk and protective factors is found in the siblings' social environments outside of the family. The importance of peer relations on sibling adjustment has been of particular interest. In their work with 30 siblings of youths with autism, Kaminsky and Dewey (2002) observed that higher levels of social support from classmates were related to better academic functioning and lower levels of loneliness. Perceived peer support has also predicted behavioral adjustment in siblings of youths with pervasive developmental disorder ($N = 46$) and Down's syndrome ($N = 45$) (Wolf, Fisman, Ellison, & Freeman, 1998).

In summary, although there is a growing body of research linking specific ecolog-

ical factors to sibling adjustment, the effects of these factors have primarily been evaluated separately. Furthermore, sibling research has generally included modest sample sizes. This study advances family science by simultaneously examining the direct effects of individual (attitude toward SB), family (family satisfaction, sibling warmth, and sibling conflict) and peer factors (close friend support and classmate support) on sibling adjustment. The hypotheses tested in this research were: (1) the overall ecological model inclusive of the individual, family and peer factors would explain a significant amount of variance in sibling self-concept, prosocial behavior, and behavior difficulties; and (2) the individual, family and peer components of the ecological model would each significantly contribute to

the explanation of self-concept, prosocial behavior and behavior difficulties.

METHOD

Participants

The data reported here come from a mixed-method investigation of the psychosocial adjustment of siblings of youths with SB (Bellin, 2006). A convenience sampling strategy was used to recruit a national cross-sectional sample of 254 families for this study. Participants were recruited from the Spina Bifida Association (SBA; <http://www.spinabifidaassociation.org>) and three geographically diverse SB clinic sites over a 15-month period (June 2004 through September 2005).¹ Parent and adolescent participants completed anonymous self-report surveys. All adolescent participants were: (a) between the ages of 11–18 years; (b) English-speaking; (c) had no current major health condition; and, (d) resided in the same household as the brother or sister with SB who was at least 6 months old. Families who failed to meet inclusion criteria ($N = 7$) or were missing the parent ($N = 8$) or adolescent survey ($N = 15$) were excluded. The final sample was comprised of 224 adolescent-parent dyads.

Procedure

The study was reviewed and approved by the university institutional review board, the Professional Advisory Committee of SBA, and the human subjects protections committees of the participating clinic sites. Interested families responded to flyers or newsletter advertisements circulated through the SBA and clinic sites. Mailed study packets included parent consent and youths assent forms, a sibling survey with the instruments described below, and a parent survey that yielded SB clinical data and family demographics. The parent and sibling surveys were assigned an arbitrary matching number and included no identifying information. The packet also included a return postcard for the purpose

of sending the adolescent participant a \$15 gift card and entering each parent and adolescent participant into \$100 gift card raffles. The postcards were destroyed at the completion of the study.

Instrumentation

Attitude Toward SB

The 13-item Child Attitude toward Illness Scale (CATIS) was developed by Austin and Huberty (1993) to capture feelings and attitudes about the health condition from the perspective of the affected child. Higher scores reflect a more positive attitude. The CATIS has solid psychometric properties (Heimlich, Westbrook, Austin, Cramer, & Devinsky, 2000). Under the guidance of the instrument developer (Austin), the CATIS was revised by adding the phrase “brother/sister with spina bifida” to each item.

Family Satisfaction

The five-item Family APGAR, created by Smilkstein (1978) and revised by Austin and Huberty (1989) for use by children, provided an assessment of how satisfied adolescent participants were with family interaction. The instrument measures five aspects of satisfaction with family, adaptation, partnership, growth, affection, and resolve, with higher scores reflecting higher levels of family satisfaction. Moderate test–retest reliability ($r = .73$) and internal consistency ($\alpha = .71$) have been reported (Austin & Huberty, 1989). The measure has established reliability and validity for use with adolescents who have SB (Sawin et al., 2002; Sawin, Buran et al., 2003).

Warmth and Conflict in the Sibling Relationship

The Sibling Relationship Questionnaire-Brief Version (SRQ) developed by Furman and Buhrmester (1985) was used

¹ Data were combined because of low participation rate ($N = 12$, 5.4%) from clinic sites.

to assess perceptions of sibling warmth and conflict. The complete SRQ includes 15 subscales that form four factors: warmth/closeness; relative power/status; conflict; and rivalry. Youths rate the characteristics of their sibling relationship across a 5-point Likert-type scale, with higher scores reflecting higher levels of the concept being measured (Furman & Buhrmester, 1985). The Warmth Factor-Brief Version is derived from six two-item subscales (intimacy, companionship, similarity, admiration by sibling, admiration of sibling, and affection) and one three-item subscale (prosocial behavior). Three two-item subscales (quarreling, antagonism, and competition) form the Conflict Factor-Brief Version. The SRQ has established psychometrics for use in the general sibling population (Furman & Buhrmester, 1985) and in sibling relationships when one member has a CHC (Fisman, Wolf, Ellison, & Freeman, 2000).

Peer Social Support

Participants completed two components of Harter's (1985) Social Support Scale for Children (SSSC), the 6-item close friend scale and the 6-item classmate scale. Higher scores reflect greater perceived support. Internal consistency reliabilities for the classmate and close friend subscales range from .74 to .79 and .72 to .83, respectively (Harter, 1985). The reliability of this instrument is also supported in more recent research with siblings of youths with diverse health conditions (Williams et al., 2002).

Self-Concept

The Children's Self-Concept Scale 2 (CSCS) (Piers & Herzberg, 2002) was administered to measure global self-concept. A dichotomous "yes" or "no" response format is used for the 60-item CSCS. Total scale scores range from 0 to 60. This psychometrically strong instrument has been widely used as an indicator of child and adolescent psychological adjustment in di-

verse populations (Piers & Herzberg, 2002), including youths with and without disabilities (Gans, Kenny, & Ghany, 2003).

Behavioral Adjustment

Two measures of behavioral adjustment, prosocial behavior and behavior difficulties were assessed by the 25-item Strengths and Difficulties Questionnaire (SDQ), a brief behavioral screening tool (Goodman, Meltzer, & Bailey, 1998). The five-item prosocial behavior scale ranges from 0 to 10, with higher scores indicating greater prosocial behavior. Four 5-item subscales, emotional symptoms, conduct problems, hyperactivity-inattention, and peer relationship problems, are summed to form the difficulties score (range = 0 to 40), with higher scores suggesting more behavior problems. Strong psychometric properties have been documented (Goodman, 2001).

SB Severity Index

The selection of factors forming the SB severity index was based on previous research and clinical experience and included the following variables reported by the parent: (a) level of lesion (e.g., sacral, lumbosacral, lumbar, thoracic); (b) presence of hydrocephalus; (c) shunt status; (d) number of shunt revisions; (e) perception of SB severity; and (f) perception of a learning disability in the child with SB. The latter two indicators were based on a subjective rating from parents. Specifically, these items of interest were measured on a 5-point Likert-type scale (1 = *strongly disagree* to 5 = *strongly agree*). Standardized *z* scores on the variables were computed and summed to generate a total SB severity index score, an approach consistent with previous research with this population (Macias, Saylor, Kreh, Clifford, & Romanczuk, 2000).

Data Analysis

Regression imputation was used to impute values for randomly missing data

dispersed throughout the variables and cases. Data on two items of the CSCS were missing for 6.3% of the sample ($N = 14$), and 15 participants (6.7% of sample) incorrectly completed the SSSC scales. To ascertain whether these data were missing in a nonrandom fashion, a dummy variable (0 = *nonmissing*, 1 = *missing*) was constructed, and a test of mean differences on the outcome variables was performed (Tabachnick & Fidell, 2001). Because no significant differences emerged between the groups on self-concept, prosocial behavior, and behavior difficulties, cases with the imputed values were retained in the sample.

Three hierarchical multiple regression analyses were performed using SPSS 15.0 to test the association between select individual, family and peer factors and sibling (1) self-concept, (2) prosocial behavior, and (3) behavior difficulties. To control for the effects of the clinical (SB severity) and demographic (birth order and gender) factors, these variables were entered on step one of each model. Next, the individual level factor (attitude toward SB) was entered on step two, the family level factors (family satisfaction, sibling warmth and conflict) on step three, and the peer level factors (classmate support and close friend support) on step four. For each model, the total variance accounted for by the ecological factors (R^2) and the change in explained variance associated with each step of the model (R^2 change) were examined. Finally, following the guidelines of Dattalo (2008) and Smithson (2003), confidence intervals (CIs) around R^2 were constructed to provide additional perspective on the utility of the models.

RESULTS

Sibling participants were primarily Caucasian ($N = 195$, 87.1%) and older than the child with SB ($N = 160$, 71.4%). A majority was female ($N = 122$, 54.5%). Participants reported a mean age of 13.81 years ($SD = 2.30$) and had lived with their

brother or sister with SB for an average of 9.79 years ($SD = 4.17$). The participating parent was most often married or partnered ($N = 197$, 87.9%) and the mother of the adolescent ($N = 188$, 83.9%). Over half of the parents reported an annual family income above \$56,000 ($N = 123$, 54.9%).

In terms of the child with SB (mean age = 10.93, $SD = 5.61$), the vast majority had a shunt ($N = 193$, 86.2%), with an average of 2.20 revisions ($SD = 5.50$) to the shunt. The level of lesion spanned all levels of the spinal cord, though a lumbar lesion level was most frequently indicated ($N = 89$, 39.7%). The characteristics of the children with SB in this sample are similar to other larger studies of children with SB (Bowman et al., 2001; Sawin, Buran, et al., 2003). A slight majority of the parent participants agreed that their child with SB had learning difficulties ($N = 117$, 52.2%; scale $M = 3.13$, $SD = 1.40$) and nearly half considered the SB to be severe in nature ($N = 109$, 48.7%; scale $M = 3.10$, $SD = 1.22$). The computed z scores on the severity index ranged from -10.29 to 11.69 ($M = .020$, $SD = 3.31$).

Descriptive Analyses

Instrument analyses, including measures of central tendency, dispersion, and internal consistency, are reported in Table 1. In each case, a higher score reflects higher levels of that concept being measured, and in the case of the Attitude Toward Illness scale, a higher score indicates a more positive attitude about the impact of SB on the participant's life. A correlation matrix (see Table 2) was also constructed to provide a preliminary picture of the relationships between the explanatory and outcome variables. As no indication of multicollinearity among the explanatory variables emerged—Pearson's correlations ranged from $r = -.04$ (association between sibling conflict and close friend support) to $r = .54$ (association between classmate support and close friend support)—all were re-

Table 1
Descriptive Statistics of Predictor and Outcome Variables (N = 224)

	<i>M</i>	<i>SD</i>	Minimum	Maximum	Scale range	α	<i>N</i> of items
Ecological factors							
Attitude ^a	48.88	8.72	19	65	13–65	.86	13
Family satisfaction ^b	18.08	4.54	6	25	5–25	.86	5
Sibling warmth ^c	51.70	10.10	23	75	15–75	.91	15
Sibling conflict ^d	14.43	5.07	6	30	6–30	.81	6
Classmate support ^e	20.01	3.27	7	24	6–24	.79	6
Close-friend support ^f	20.57	3.90	8	24	6–24	.89	6
Outcomes							
Self-concept ^g	47.88	8.58	12	59	0–60	.91	60
Prosocial behavior ^h	8.07	1.66	3	10	0–10	.62	5
Behavior difficulties ⁱ	10.76	5.60	1	32	0–40	.78	20

^a Attitude Toward Illness (CATIS) (Austin & Huberty, 1993). ^b Family APGAR (Austin & Huberty, 1989). ^c Sibling Relationship Questionnaire (SRQ)-Warmth/closeness scale (Furman & Buhrmester, 1985). ^d Sibling Relationship Questionnaire (SRQ)-Conflict scale (Furman & Buhrmester, 1985). ^e Social Support Scale for Children (SSSC)-Classmate scale (Harter, 1985). ^f Social Support Scale for Children (SSSC)-Close friend scale (Harter, 1985). ^g Self-Concept Scale for Children (SCSC) (Piers & Herzberg, 2002). ^h Strengths and Difficulties Questionnaire (SDQ)-Prosocial behavior scale (Goodman et al., 1998). ⁱ Strengths and Difficulties Questionnaire (SDQ)-Behavior difficulties scale (Goodman et al., 1998).

tained in the model (Tabachnick & Fidell, 2001).

Self-Concept Model

As reported in Table 3, the overall model inclusive of individual, family, and peer factors significantly predicted self-concept, $R^2 = .57$, 95% CI = 0.49 to 0.65, $F(13, 215) = 22.26$, $p < .001$. In this model, the hypothesized relationships between the individual, family, and peer factors and sibling self-concept were supported. After controlling for the clinical and demographic factors, the ad-

dition of the individual level factor on step 2 explained a significant amount of variance in self-concept, R^2 change = .189, $F(1, 215) = 50.88$, $p < .001$. The addition of the family level factors on step 3, R^2 change = .182, $F(3, 212) = 20.83$, $p < .001$, and the peer level factors on step 4, R^2 change = .184, $F(2, 210) = 44.74$, $p < .001$, likewise explained variation in self-concept scores above and beyond that accounted for in previous steps of the model. A main effect for several ecological factors was found (see Table 3). Specifically, self-concept was associated with at-

Table 2
Intercorrelations Among Predictor and Criterion Variables (N = 224)

Variable	1	2	3	4	5	6	7	8	9
1. Attitude toward spina bifida	—	.32**	.15*	-.21**	.23**	.20**	.43**	.11	-.27**
2. Family satisfaction		—	.48**	-.19**	.40**	.36**	.54**	.35**	-.41**
3. Sibling warmth			—	-.31**	.19**	.18*	.31**	.42**	-.21**
4. Sibling conflict				—	-.12	-.04	-.22**	-.16	.27**
5. Classmate support					—	.54**	.63**	.24**	-.45**
6. Close-friend support						—	.42**	.20**	-.27**
7. Self-concept							—	.28**	-.64**
8. Prosocial behavior								—	-.22**
9. Behavior difficulties									—

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

Table 3

Self-Concept Model (N = 220)

	Step 1	Step 2	Step 3	Step 4
SB severity index	-.10	-.05	-.02	-.05
Birth order ^a	.03	-.04	-.02	-.01
Gender ^b	.04	.01	.04	.12*
Attitude toward spina bifida		.46**	.29**	.22**
Family satisfaction			.40**	.20*
Sibling warmth			.06	.08
Sibling conflict			-.08	-.07
Classmate support				.46**
Close-friend support				.07
<i>R</i> ² change	.01	.19	.18	.18
<i>df</i>	3,216	1,215	3,212	2,210
<i>F</i>	.94	50.88**	20.83**	44.74**
Total <i>R</i> ²	.01	.20	.38	.57
Total <i>R</i> ² (adjusted)	.00	.19	.36	.55
<i>df</i>	3,219	4,219	7,219	9,219
<i>F</i>	.94	13.58**	18.84**	30.64**

Note. Standardized regression coefficients reported in upper half of table.

^a Coding for birth order: 0 = *younger*, 1 = *older*. ^b Coding for gender: 0 = *female*, 1 = *male*.

* $p < .05$. ** $p < .001$.

titude toward SB ($\beta = .22, p < .001$), family satisfaction ($\beta = .20, p = .001$), and classmate social support ($\beta = .46, p < .001$). Participants who reported more positive feelings about the impact of SB on their lives, described higher satisfaction with family, and greater social support from classmates also endorsed a higher self-concept. Lastly, sibling gender was the only significant covariate in the final model ($\beta = .12, p = .01$), with higher self-concept linked to male gender.

Prosocial Behavior Model

The overall model (see Table 4) comprised of the individual, family, and peer factors significantly predicted prosocial behavior, $R^2 = .27$, 95% CI = 0.17 to 0.36, $F(9, 219) = 8.50, p < .001$. In contrast to the self-concept model, however, the addition of the individual level ecological factor on step 2, R^2 change = .013, $F(1, 215) = 3.17, p = .076$, and the peer level factors on step 4, R^2 change = .008, $F(2, 210) = 1.14, p = .322$, failed to explain variance in prosocial behavior above and beyond that accounted for in the previous steps. However, the addition of family level factors on step 3 was significant,

R^2 change = .154, $F(3, 212) = 14.73, p < .001$. In the final model, family satisfaction ($\beta = .18, p = .019$) and sibling warmth ($\beta = .27, p < .001$) achieved significance, with higher levels of family satisfaction and sibling closeness associated with greater prosocial behavior among participants. Gender again emerged as the only significant covariate in the final model, although in this case, female gender was associated with greater prosocial behavior ($\beta = -.23, p < .001$).

Behavior Difficulties Model

Finally, as shown in Table 5, the third model was also supported, with the set of ecological factors explaining a significant amount of variance in behavior difficulties ($R^2 = .30$, 95% CI = 0.21 to 0.40, $F(9, 219) = 10.16, p < .001$). In this model, the hypothesized relationships between the individual, family, and peer factors and sibling behavior difficulties were also supported. Specifically, the addition of the individual level factor on step 2 explained a modest but significant amount of variance in behavior difficulties, R^2 change = .075, $F(1, 215) = 17.54, p < .001$. The entry of

Table 4

Prosocial Model (N = 220)

	Step 1	Step 2	Step 3	Step 4
SB severity index	-.10	-.08	-.04	-.05
Birth order ^a	.03	.02	.01	.02
Gender ^b	-.29**	-.30**	-.24**	-.23**
Attitude toward spina bifida		.12	.01	.001
Family satisfaction			.22*	.18*
Sibling warmth			.27**	.27**
Sibling conflict			.04	.04
Classmate support				.10
Close-friend support				-.01
<i>R</i> ² change	.091	.013	.154	.008
<i>df</i>	3,216	1,215	3,212	2,210
<i>F</i>	7.24**	3.17	14.73**	1.14
Total <i>R</i> ²	.091	.105	.259	.267
Total <i>R</i> ² (adjusted)	.079	.088	.235	.236
<i>df</i>	3,219	4,219	7,219	9,219
<i>F</i>	7.24**	6.28**	10.59**	8.50**

Note. Standardized regression coefficients reported in upper half of table.

^a Coding for birth order: 0 = *younger*, 1 = *older*. ^b Coding for gender: 0 = *female*, 1 = *male*.

* $p < .05$. ** $p < .001$.

the family factors on step 3 explained an additional 14.8% of the variance in behavior difficulties, $F(3, 212) = 13.47, p < .001$ and the peer factors an additional 7.8% of the variance, $F(2, 210) = 11.73, p < .001$. The demographic and clinical covariates

failed to achieve significance in the final model. However, a main effect was found for family satisfaction ($\beta = -.23, p = .002$), sibling conflict ($\beta = .18, p = .005$), and classmate support ($\beta = -.31, p < .001$), with lower levels of family satisfaction and

Table 5

Behavior Difficulties Model (N = 220)

	Step 1	Step 2	Step 3	Step 4
SB severity index	-.004	-.03	-.03	-.02
Birth order ^a	-.04	.01	-.03	-.02
Gender ^b	-.04	-.02	-.004	-.08
Attitude toward spina bifida		-.28**	-.13*	-.09
Family satisfaction			-.35**	-.23*
Sibling warmth			.03	.02
Sibling conflict			.19*	.18*
Classmate support				-.31**
Close-friend support				.004
<i>R</i> ² change	.003	.075	.148	.078
<i>df</i>	3,216	1,215	3,212	2,210
<i>F</i>	.185	17.54**	13.47**	11.73**
Total <i>R</i> ²	.003	.078	.225	.303
Total <i>R</i> ² (adjusted)	.011	.061	.200	.273
<i>df</i>	3,219	4,219	7,219	9,219
<i>F</i>	.185	4.54*	8.82**	10.16**

Note. Standardized regression coefficients reported in upper half of table.

^a Coding for birth order: 0 = *younger*, 1 = *older*. ^b Coding for gender: 0 = *female*, 1 = *male*.

* $p < .05$. ** $p < .001$.

support from peers and greater conflict in the sibling relationship associated with increased behavior difficulty.

DISCUSSION

The purpose of this investigation was to advance family science by testing a multi-level model of adjustment for siblings of youths with SB. The components of the model, attitude toward SB, family satisfaction, sibling warmth and conflict, and classmate and close friend support, were strongly associated with self-concept, and to a lesser degree, prosocial behavior and behavior difficulties. Increased perspective on the utility of these models is found in Cohen's (1988) f^2 , the effect size measure used in the context of multiple regression, where f^2 of 0.02 = small, 0.15 = medium, and 0.35 = large. These benchmarks place the self-concept model ($f^2 = 1.31$), prosocial behavior model ($f^2 = 0.43$), and the behavior difficulties model ($f^2 = 0.36$) all within the parameters of a large effect size.

A significant contribution of this work was the use of a social-ecological framework to heighten understanding of the diverse risk and protective factors underlying variation in sibling adjustment. Consistent with social-ecology theory, the hypotheses that the individual, family, and peer components of the model would uniquely contribute to the explanation of the adjustment outcomes were largely supported. The value of a social-ecological perspective on sibling functioning was further indicated by findings of divergent factors associated with psychological and behavioral adjustment. For example, attitude toward SB was related to self-concept, whereas qualities of the sibling relationship were associated with behavioral outcomes. Specifically, sibling warmth was a correlate of prosocial behavior, while sibling conflict was a factor associated with behavior difficulties.

The observed relationships between sibling interactions and adjustment outcomes are generally consistent with prior research (Fisman et al., 1996; Gamble &

McHale, 1989). However, contrary to the findings in siblings of youths with cancer where high levels of sibling warmth were related to greater adjustment difficulty (Labay & Walco, 2004), for siblings of youths with SB, higher warmth is related to positive outcomes. The different outcomes associated with sibling warmth may perhaps be explained through Rolland's (1984; Rolland & Williams, 2005) psychosocial typology of illness that explains how the impact of health conditions on the child and surrounding family members may vary according to onset, course, outcome, incapacitation, and degree of uncertainty of the condition. It is possible that siblings of youths with cancer who have an established, intimate relationship and are suddenly confronted with the onset and consequences of a catastrophic illness have more difficulty with adjustment due to their fear of the sibling possibly dying. In siblings where the congenital condition has always been present, the response might be very different. Even though stressors from intermittent health crises may occur, the development of warmth and compassion may enhance positive adjustment outcomes. Though it is not uncommon for sibling relationships to have elements of stress and discord, these data suggest that pronounced conflict may present a threat to adaptive outcomes. Consequently, clarifying the factors underlying problematic sibling interaction seems to be a key task for family-centered practitioners to help families steer their children back toward positive relations.

The impact of the broader family environment on sibling functioning was particularly strong in these data. In fact, family satisfaction was the only ecological factor associated with all dimensions of sibling adjustment. It is not surprising that the manner in which the family encourages growth, the expression of feelings and shared problem-sharing, aspects of family functioning measured by the Family APGAR (Austin & Huberty, 1989), influ-

ences the developing adolescent's self-concept and behavior. Associations between positive family climates and sibling adjustment are well documented, including in research with siblings of youths with autism (Verte, Roeyers, & Buysse, 2003) and diabetes (Gallo & Szychliński, 2003).

Another contribution of this research is our enhanced understanding of the protective role peer relations may play in adjustment outcomes. The complex management of SB in the home environment, frequent visits with diverse health care professionals, and, for some youths with SB, periodic hospitalizations for shunt revisions and orthopedic surgeries, may place significant psychosocial demands on parents. Consequently, parents may not be as physically or emotionally available for siblings. Furthermore, the complex health (i.e., bowel and bladder problems), neurocognitive (i.e., executive functioning), and physical sequelae (i.e., lower extremity weakness or paralysis) associated with SB create challenges for the affected siblings to engage in the full range of socialization activities as typically developing youths. Siblings may therefore seek support and role models outside of the immediate family environment. Indeed, the qualitative data from the larger study indicated that most siblings have developed solid social networks comprised of mature peers who appreciated the unique challenges and opportunities spawned by the experience of having a sibling with SB (Bellin, Kovacs, & Sawin, 2008).

Relationships observed among the demographic and clinical covariates and sibling adjustment outcomes also have notable implications for clinical intervention. While not the primary protective factor for this sample, it appears that gender does have some influence on select outcomes. Whereas a higher self-concept had a significant but small association with male gender, female gender had a larger impact on prosocial behavior. These data appear consistent with theories of development sug-

gesting that women are more focused on relationships (Gilligan, 1982). A final noteworthy result was the lack of a significant effect of condition severity. Instead, the health of the surrounding family environment consistently emerged as a more salient influence on sibling adjustment. Characteristics of the family milieu have similarly been found as more reliable predictors of psychosocial outcomes than SB severity for both the affected child (Sawin, Buran, et al., 2003) and the parent (Kronenberger & Thompson, 1992). Given the key role the family environment appears to play, the development and testing of family interventions designed to support family resilience seems paramount. However, a recent review of the family literature found no family based interventions in SB (Holmbeck et al., 2006).

Methodological Considerations and Future Directions

It is possible that recruitment via the SBA may have caused a selection-bias effect, and consequently, the findings reported here may not be representative of the larger population. Further, a comparison group comprised of siblings of typically developing youths would help to disentangle risk and protective factors that are truly grounded in the experience of having a brother or sister with SB versus those that are a function of normative developmental processes. Another limitation relates to the cross-sectional nature of the data which prohibits definitive conclusions about the directions of the observed relationships (Holmbeck et al., 2006). Future investigations would benefit from a longitudinal design, because this would enable the identification of risk and protective factors that are influential across developmental periods and those that are associated with a specific phase of development. Lastly, inclusion of perspectives from all members of the family, including the child with SB and parents, would be helpful to provide a more holistic appreciation of fac-

tors influencing sibling adjustment and expand this study's initial contribution to the field of family science.

CONCLUSION

By definition, the web of services in a family-centered care model extends beyond the child with the chronic health condition. This research utilized a social-ecological framework to enrich understanding of key areas to assess and support for siblings of youths with SB. These data identified attitude toward SB, satisfaction with family functioning, qualities of sibling interactions, and experiences of peer support as particularly salient dimensions of sibling life to address in family-centered assessment and intervention processes. Minimizing threats and bolstering protective influences on sibling adjustment are essential ingredients to advance a family-centered perspective in SB.

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