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Psychosocial and Family Functioning in Spina Bifida

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Abstract

A developmentally-oriented bio-neuropsychosocial model is introduced to explain variation in family functioning and psychosocial adjustment in youth and young adults with spina bifida (SB). Research on the family functioning and psychosocial adjustment of individuals with SB is reviewed. The findings of past research on families of youth with SB support a resilience-disruption view of family functioning. That is, the presence of a child with SB disrupts normative family functioning, but many families adapt to such disruption and exhibit considerable resilience in the face of adversity. Parents of youth with SB, and particularly those from lower SES homes, are at-risk for psychosocial difficulties. Individuals with SB are at-risk for developing internalizing symptoms, attention problems, educational difficulties, social maladjustment, and delays in the development of independent functioning. Emerging adults are often delayed in achieving milestones related to this stage of development (e.g., vocational and educational achievements). Methodologically-sound, longitudinal, and theory-driven studies of family and psychosocial functioning are needed, as are randomized family-based intervention trials, to promote adaptive functioning and better psychosocial outcomes in families of individuals with SB.

Keywords

spina bifida; psychosocial adjustment; family; children; adolescents; emerging adults

Psychosocial and Family Functioning in Spina Bifida

Spina bifida (SB) is a relatively common congenital birth defect that has a pervasive multi-systemic impact on the physical, neurocognitive, psychological, and social functioning of affected individuals. Historically, most of the research on SB has focused on the physical and neurocognitive domains, with less attention paid to the psychological and social domains of functioning. However, it is well known that the clinical symptoms of SB place considerable physical, psychological, and social demands on the individuals and families involved [Greenley et al., *2006*; Holmbeck et al., *2003*; Kelly et al., *2008*; Singh *2003*]. Specifically, all of the following SB-related stressors likely have a significant and cumulative impact on individual and family functioning: (1) the cognitive and neurological features of SB, particularly the most common type of spinal lesion, myelomeningocele (e.g., executive functioning deficits, attention problems, learning difficulties; see Dennis and Barnes, in press), (2) the effects of SB on physical development [e.g., precocious puberty, short stature, and obesity are all more common in this population than in the general population; Dosa et al., *2009*], (3) the multiple surgical procedures experienced by most individuals in this population (e.g., shunt revisions, orthopedic surgeries), (4) the difficulties with bowel and bladder management as well as ambulation challenges, (5) the characteristic

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social skills deficits, and (6) individuals' difficulties in mastering developmental milestones (e.g., autonomy development).

Although there is considerable variability in the degree to which children with SB, their parents, and their siblings experience stress and adjustment difficulties, children with SB are at-risk for higher rates of adjustment problems, including internalizing and social problems [Appleton et al., *1997*; Holmbeck et al., *2003*; Lavigne and Faier-Routman *1992*]. Similarly, parents of children with SB appear to experience more stress than parents of typically developing children [Holmbeck et al., *1997*; Wallender et al., *1990*]. However, research has documented considerable resilience in these families as well [e.g., Holmbeck, Coakley et al., *2002*].

The current paper focuses on the family functioning and psychosocial adjustment of individuals with SB. *Family relationships* are particularly salient and influential social relationships for youth with SB, given that children with SB tend to be more socially isolated from their peers than are typically developing children [Holmbeck et al., *2003*]. Further, we are interested in family relationships because SB impacts not only the affected child but the parents and other family members as well. Given the pervasive impact of this condition, we were also interested in the level of *psychosocial adjustment* in such individuals across multiple adjustment domains [e.g., internalizing symptoms, externalizing symptoms, self-concept, and observed behavior; the literature on quality of life in this population is reviewed in a separate paper in this special issue; Sawin et al., press].

First, we introduce a developmentally-oriented bio-neuropsychosocial model to explain variation in family functioning and psychosocial adjustment in youth and young adults with SB (see Figure 1). Next, we provide a review of past research findings related to family functioning and psychosocial adjustment in youth with SB. Finally, we discuss clinical implications and recommendations for future research.

Theoretical Basis for Research on Family Functioning and Psychosocial Adjustment in SB

There are several theories that identify multiple factors and contexts that directly and indirectly influence child development and family functioning in children with chronic health conditions [e.g., social-ecological theory; Kazak et al., *2009*]. However, the goal of this review is to discuss ways in which family functioning fits into a more specific conceptualization of psychosocial functioning in children with SB. Therefore, we provide a bio-neuropsychosocial model of psychological adjustment in children, adolescents, and emerging adults with SB (Figure 1). As illustrated in Figure 1, the adjustment of individuals with SB is likely determined by the interacting influences of multiple biological, neuropsychological, social (including family functioning), and contextual factors. Moreover, all of these factors likely have causal relations with each other, with each evolving and changing over time ("Time" is included in the model to indicate that associations among the processes evolve with development and over time).

Each construct within Figure 1 can be considered a second-order domain with multiple sub-domains. For example, the family domain includes multiple sub-domains, such as the following: parental adjustment, parenting behaviors, parenting satisfaction, parenting stress, family system-level constructs (e.g., conflict, affect, cohesion), family burden, family problem-solving abilities, family coping, family management of the medical condition and adherence, family life events, and marital functioning. These sub-domains may impact each other, in addition to having an impact on the individual's level of adjustment. Additionally, each subdomain can be evaluated in multiple ways (e.g., questionnaire vs. observational methods; parent vs. child report). Moreover, the manner in which SB may impact upon a family system can vary *within* a family system over time. For example, a family may

function adaptively while their child with SB is in grade school, but have difficulty adjusting when the same child transitions into adolescence.

Review of Past Research on Family Functioning in Spina Bifida

Family system and marital functioning—Holmbeck and colleagues [*2006*] published a review of research that examined the impact of SB on family functioning [also see Ammerman et al., *1998*; Greenley et al., *2006*; Kelly et al., *2008*; Singh *2003*]. In general, we found that significant differences between SB and comparison groups are more likely to be found when: (1) the study has a larger sample size, (2) the study has a stronger research design, and (3) the comparison is to normative data (rather than to a matched comparison sample).

Findings from two studies revealed that 12-13% of families of children with SB exhibited clinical levels of "family dysfunction" [Ammerman et al., *1998*; Wiegner and Donders *2000*]. Interestingly, such rates of family dysfunction are lower than those found in families of children with cerebral palsy [35%; Wiegner and Donders, *2000*]. A significant number of family members with children who have SB report difficulties in maintaining clear roles and responsibilities in the family system [23% in the clinically problematic range in the Ammerman et al., *1998* study]. With respect to risk factors, Holmbeck, Coakley et al. [*2002*] found that families of youth with SB who were also from lower SES backgrounds were particularly at-risk for lower levels of family cohesion, supporting a cumulative risk view of such families (i.e., SB and lower SES have additive effects on family functioning).

Despite these difficulties, many families of children with SB also evidence high levels of resilience. In fact, most past studies reveal differences on some family variables but not on others. For example, one study found significant group differences on family cohesion (with comparison families being higher) but no group differences on level of family conflict [Holmbeck, Coakley et al., *2002*]. With respect to conflict, Coakley et al. [*2002*] found that, unlike their typically developing peers, families of youth with SB did not evidence normative increases in family conflict as a function of pubertal development. These investigators speculated that families of youth with SB may be less responsive to developmental change. Indeed, parents of youth with SB are less likely to discuss issues of sexuality with their offspring than are parents of typically developing youth [Blum et al., *1991*]. In support of this "attenuated response to development" hypothesis, Jandasek, Holmbeck and colleagues [*2009*] conducted longitudinal growth analyses over the age period of 9-15 years of age and found that family conflict intensity increased over this early adolescent age range in families of typically developing youth but did not increase with age in families of children with SB.

Findings are mixed with respect to marital functioning. Some studies show no differences in marital functioning between families of children with SB and able-bodied families [Cappelli et al., *1994*; Holmbeck et al., *1997*; Spaulding and Morgan *1986*]. Interestingly, some studies have found that having a child with a disability can strengthen a marriage [Cappelli et al., *1994*; Kazak and Clark *1986*]. It appears that the quality of the marital relationship *prior to* the birth of the affected child is an important predictor of subsequent family adjustment.

In general, the findings of past work support a "resilience-disruption" view of family functioning [Costigan et al., *1997*]. That is, SB appears to disrupt some aspects of family and parent functioning for many families, but such families also tend to demonstrate considerable resilience across other adjustment domains. Moreover, parents of youth with SB tend to be relatively less responsive to maturational change in their offspring compared to parents with typically developing children.

Adjustment of parents and parenting behaviors—Despite the relatively low levels of family dysfunction at the family systems level, it appears that a sizable minority of parents of children with SB exhibit clinical levels of global psychological distress [e.g., anxiety, depressive symptoms, somatic complaints; Holmbeck et al., *1997*; Kronenberger and Thompson *1992*a]. Although most studies that report on parental functioning have focused on maternal functioning, fathers of children with SB exhibited higher levels of global distress than fathers from comparison families in one study [Holmbeck et al., *1997*]. In a recent meta-analysis of 15 studies, Vermaes, Janssens, Bosman, and Gerris [*2005*] found medium to large effect sizes for the impact of SB on mother and father psychological adjustment, with somewhat larger effects sizes for mothers (d=.73) than for fathers (d=.54).

Across several studies, parents of children with SB tend to experience more stress in their roles as parents than do comparison parents [Holmbeck et al., *1997*; Macias et al., *2007*; Vermaes, Gerris et al., *2007*]. Typically, such parents feel less satisfied and competent as parents, feel more isolated, are less adaptable to change, and hold less optimistic views about the future than comparison parents [Barakat and Linney *1995*; Grosse et al., *2009*; Holmbeck et al. *1997*; Sawin et al., *2003*]. Parents who are single, socially isolated, older, or from an ethnic minority or a low SES background are particularly at-risk for such outcomes [Barakat and Linney *1992*; Fagan and Schor *1993*; Holmbeck, Coakley et al., *2002*; Kronenberger and Thompson *1992*b; Macias et al. *2001*].

With respect to parenting behaviors, it has been found that increases in parental responsiveness are associated concurrently with increases in adaptive coping strategies in youth with SB [e.g., problem-focused coping; McKernon et al., *2001*[. However, parents of children with SB tend to exhibit higher levels of intrusiveness, psychological control, and authoritarian parenting [i.e., parenting that undermines the autonomy development of their offspring; Holmbeck, Johnson et al., *2002*; Holmbeck, Shapera et al., *2002*; Sawin et al., *2003*; Seefeldt et al., *1997*; Vermaes et al., *2007*] and these behaviors tend to be linked with less desirable child outcomes. Specifically, higher levels of intrusiveness (sometimes referred to as overprotectiveness) tend to be associated with lower levels of decision-making autonomy which are, in turn, related to higher levels of externalizing symptoms [Holmbeck, Johnson et al., *2002*; Sawin et al., *2003*]. Moreover, group differences on these types of parenting variables appear to be mediated by child cognitive ability, such that children with SB tend to have lower verbal IQs and children with lower verbal IQs tend to have parents who are more controlling [Holmbeck, Johnson et al., *2002*]. In general, high levels of intrusive parental control tend to be related to adjustment difficulties in any population; thus, the fact that there are higher levels of these parenting behaviors in families of youth with SB may be one factor in explaining why these children are at-risk for adjustment difficulties.

Adjustment of siblings—There are few studies that examine the functioning of siblings of children with SB. Findings have been contradictory, as an early study using teacher report found a four times greater likelihood of adjustment problems for siblings as compared to a comparison sample [Tew and Laurence *1973*], while a more recent study of siblings of youth with SB reported no differences in self-concept compared to siblings of typically developing youth [Kazak and Clark *1986*]. Qualitative research has identified both positive and negative outcomes related to having a sibling with SB. For example, siblings report significant levels of concern for the health of their sibling with SB, emotional upset in relation to their siblings' experience with discrimination, teasing, and bullying, and sadness related to the lack of opportunities to engage in physical activities with their siblings with SB [Bellin et al., *2008*; Kiburz *1994*]. Siblings have also identified some positive effects, such as increased empathy for their sibling and a greater appreciation for their own physical abilities [Bellin et al., *2008*; Kiburz *1994*].

Although there are only a few studies that examine siblings of children with SB, there are several studies that combine siblings of different illness groups and examine the functioning of siblings of children with chronic conditions (rather than focusing on only one condition). A recent meta-analysis combining the results from over 50 studies found that psychological functioning, peer activities, and cognitive development scores were lower for siblings of children with chronic conditions compared to comparison samples [Sharpe and Rossiter *2002*]. Larger effect sizes were found for internalizing symptoms (e.g., depression, anxiety) as compared with externalizing symptoms (e.g., aggression), and for studies that employed comparisons to normative data versus comparison samples. While there may be an increased risk for negative psychological effects for siblings of children with chronic illnesses, siblings are not uniformly at-risk and there are many factors that influence psychological outcomes for siblings.

The behavioral and psychological functioning of siblings are significantly associated with socioeconomic status, family cohesion, perceptions of social support, and their knowledge of and attitudes towards the illness [Taylor et al., *2001*; Williams et al., *1999*; Williams et al. *2002*]. Gender and birth order of the sibling have been examined in several studies, but no consistent findings have emerged [Sharpe and Rossiter *2002*; Tew and Laurence *1973*; Williams *1997*]. For siblings of children with SB, more positive attitudes toward SB, greater family satisfaction, lower levels of sibling conflict, and increased social support from classmates significantly predicted higher levels of self-concept and prosocial behavior, and lower rates of behavior problems [Bellin et al., *2009*]. In this study by Bellin and colleagues [*2009*], family satisfaction was the only significant predictor across all three sibling adjustment outcome measures, suggesting that family variables may be particularly salient for sibling adjustment.

Review of Past Research on Psychosocial Functioning in Spina Bifida

Research on children and adolescents—Youth with SB are at-risk for exhibiting higher levels of internalizing symptoms (e.g., depression, anxiety) and lower levels of self-concept than comparison children [Ammerman et al., *1998*; Appleton et al., *1997*; Holmbeck et al., *2003*; Shields et al., *2008*]. Those with hydrocephalus often exhibit difficulties in certain areas of cognitive functioning and school performance [e.g., arithmetic, nonverbal cognitive skills; Fletcher and Dennis *2010*]. Such children are also more likely to exhibit attention and concentration difficulties in school settings and tend to score at the low end of the average range of intelligence [Fletcher and Dennis *2010*; Holmbeck DeLucia et al., *2009*; Hommeyer et al., *1999*].

To date, more work has been done in evaluating children with SB in the areas just noted than has been conducted in the area of social adjustment. This is surprising given that this area of psychosocial functioning is problematic for most children with SB [Blum et al., *1991*; Holmbeck et al., *2003*]. Based on our own analyses and the work of others, youth with SB, as compared to typically developing youth and those with other chronic conditions, tend to be socially immature and passive, have fewer friends, be less likely to have social contacts outside of school, and date less during adolescence [e.g., Blum et al., *1991*; Ellerton et al., *1996*; Holmbeck et al., *2003*] and these difficulties appear to be maintained over time [Holmbeck DeLucia et al., *2009*].

The degree to which an adolescent exhibits decision-making autonomy in both medical and non-medical areas is another highly salient developmentally-oriented variable for these youth and their families [Anderson and Coyne *1993*; Friedman et al., *2009*]. Typically-developing adolescents view more issues as falling within their own decision-making jurisdiction than they did during childhood and they are also increasingly likely to question the legitimacy of parental authority [Darling et al., *2008*; Smetana *1988*]. Interestingly, our

own results on youth with SB run contrary to this typical developmental trend. Specifically, findings revealed that children and adolescents with SB (and especially boys and those with lower levels of intelligence) tend to be more dependent on adults for guidance, less likely to exhibit behavioral autonomy at home, less likely to exhibit intrinsic motivation at school, and less likely to express their own viewpoints during observed family interactions [Davis et al., *2006*; Friedman et al., *2009*; Holmbeck, Johnson et al., *2002*; Holmbeck et al., *2003*]. Variation in intrinsic motivation (i.e., interest in learning and mastery, curiosity, preference for challenge) proved to be the most robust predictor of psychosocial adaptation (i.e., scholastic success, social acceptance, and positive self-worth) in one of our studies [Coakley et al., *2006*].

Research on emerging adults—Emerging adulthood is a critical period in the life of older adolescents with SB [Holmbeck et al., in press]. Indeed, the transition to adulthood and "community participation" in youth with chronic physical conditions have received considerable attention recently [Blum et al., *2002*], with large portions of national conferences being devoted to these topics [e.g., First World Congress on Spina Bifida Research and Care, March, *2009*, Orlando, FL] and entire volumes being published on transition issues [e.g., Lollar in press]. In general, many young adults with SB are capable of high levels of independent functioning across multiple domains but most have not been successful in fully engaging in the larger community of typically developing emerging adults [Buran et al., *2004*]. In this section, we review findings related to many of the major milestones of emerging adulthood (i.e., psychosocial adjustment, educational achievement, and employment and vocational outcomes). We refer the reader to other articles in this special issue for discussions of romantic relationships and sexuality, independent living, and the transition to adult medical care (see Webb, in press; Sawyer, in press).

Regarding *psychosocial adjustment*, emerging adults with SB, like their younger counterparts, are at-risk for depressive symptoms and anxiety [Bellin et al., in press; Dicianno et al., *2009*]. Regarding *educational outcomes*, emerging adults with SB are less likely to go to college [41-49% of individuals with SB go to college vs. 66% of typically developing youth; Bowman et al., *2000*; Cohen et al., *2003*; Zukerman *2008*]. With respect to *vocational outcomes*, recent studies report rates of full- or part-time employment ranging from 36-41% [Liptak et al., *2009*; McDonnell and McCann *2000*; Zabel and Bellin *2009*; Zukerman *2008*], which are significantly lower than those found in typically developing youth [e.g., roughly 75%; Cohen et al., *2003*; Hamilton and Hamilton *2006*; Zukerman *2008*] and in those with other chronic conditions [e.g., asthma, cancer; 68%-78%; Gerhardt, Dixon et al., *2007*; Liptak et al. *2009*].

Unfortunately, we know almost nothing about factors that predict whether or not an emerging adult with SB is able to go to college and become employed. Studies that have been conducted on individuals with SB have tended to focus only on demographic or medical severity predictors. For example, Liptak et al. [*2009*] found that communication problems, difficulties with managing responsibilities, lower levels of parental education, and higher rates of parental unemployment were associated with poorer social, vocational, and educational transitions. Zabel and Bellin [*2009*] found that young adult males with SB were more likely to work than females, but that females were more likely to live independently. With respect to medical severity, Hetherington, Dennis, Barnes, Drake, and Gentili [*2006*] found that spinal lesion level and number of shunt revisions were related to employment outcome [with higher lesion levels and more shunt revisions being related to worse occupational outcome; Barf et al., *2009*, found similar results in the Netherlands, as did Zabel and Bellin, *2009*, in the US].

In the absence of actual data, others have speculated about why young adults with SB are less likely to successfully negotiate the milestones of emerging adulthood. For example, some have described the complexities in managing "real world" responsibilities with a chronic physical condition, including transportation difficulties and issues related to accessibility [Barf et al., *2009*; Dicianno et al., *2009*; Zabel and Bellin *2009*]. Other explanations for these developmental delays focus on financial concerns [including lack of health insurance; Park et al., *2006*], lack of job training and vocational rehabilitation services, employment discrimination, stigmas related to physical appearance, and a lack of autonomy-related socialization in early childhood [Dicianno et al., *2008*, *2009*; Schriner et al., *1993*].

Clinical Implications of Research on Family and Psychosocial Functioning

The current literature suggests that, although children with SB and their families are at-risk for psychosocial difficulties, many demonstrate significant resilience. These results have clinical implications for providers working with families of children, teens, and young adults with SB [Greenley et al., *2006*]. First, we needed to use the research literature to inform interventions for individuals and families at risk. Basic research and intervention research should focus on similar variables so that results of the former can inform the goals of the latter [Holmbeck et al., *2006*]. Further, adoption of similar theoretical frameworks in both basic and intervention studies will permit development of theory-driven and evidence-based intervention programs. Interventions should target families with the greatest number of risk factors for psychosocial difficulties, such as low SES, single parent, and ethnic minority families.

Clinics could enhance comprehensive care by adopting a model for identifying families in need of treatment for psychosocial difficulties. Kazak [*2006*] presented a Pediatric Psychosocial Preventative Health Model (PPPHM), based on a social-ecological framework. According to this model, all families receive a brief assessment to determine what level of services they need – universal, targeted, or clinical/treatment groups. The largest group of families would fall within the "universal" category, suggesting that they are coping well with the challenges of having a family member with SB. General support and provision of resources could be used to foster the strengths of these families. Families within the "targeted" range demonstrate moderate levels of acute distress or risk factors. Interventions specific to the distress or that reduce risk factors would be appropriate. Finally, the smallest group would be those with high distress or a greater number of risk factors. This group would need the highest level of clinical attention, as they would be at-risk for the most negative outcomes.

Additionally, risk factors can change and families can move between such groups at any time. There may be particular developmental periods that add to risk factors for certain families. For example, the transition to adolescence may be a challenging time, as parents and teens negotiate the gradual transfer of medical management from parent to child. The adolescent's desire for autonomy and independence may conflict with the need for strict adherence to a complex medical regimen. Therefore, assessment must be on-going and relevant to the developmental period of the individual with SB.

Given that children with SB are at-risk for delays in the development of autonomy [Davis et al., *2006*; Friedman et al., *2009*], it would be important to educate parents regarding autonomy and how to help foster independence and adherence. Discussions need to begin during childhood and should focus, if possible, on helping the child work toward the mastery of important developmental milestones, such as educational and vocational achievement, living independently, and community participation.

In sum, theoretical perspectives, such as the bio-neuropsychosocial model discussed in this chapter, will help medical and other providers attend to the broader impact of SB on the psychological and social functioning of all family members. Identifying families at-risk for poor psychosocial outcomes, intervening in programmatic ways, and evaluating such intervention strategies will move the field toward a better understanding of how to promote healthy functioning in individuals with SB.

Recommendations for Future Research on Family and Psychosocial Functioning

First, it is recommended that future work be theory-driven, where hypotheses, measure selection, and statistical strategies follow directly from a theoretical framework. For example, longitudinal, mediational prediction models where intervening mechanisms are proposed are likely to yield significant and useful information, as well as important implications for interventions. When mediational prediction models are applied in studies that examine differences between SB and comparison samples, we are able to go beyond asking whether there are differences between groups and move toward asking *why* these group differences exist [Holmbeck, Zebracki et al., *2009*]. For example, Holmbeck, Johnson, and colleagues [*2002*] found that associations between intrusive parenting and child adjustment outcomes in families of children with SB were mediated by level of child behavioral autonomy, such that intrusive parenting was associated with lower levels of behavioral autonomy, which were, in turn, associated with higher levels of externalizing symptoms. More generally, the literature on family and psychosocial functioning in individuals with SB will benefit from theoretical advances that include the following features: (1) a developmental emphasis, (2) a focus on both illness-specific and general family processes, (3) models that examine mediational processes, and (4) models that take into account family-related variables (e.g., autonomy-promoting parenting) that serve as buffers for associations between risk factors (e.g., neurological status) and negative outcomes (e.g., academic failure).

Second, it is recommended that research be programmatic and longitudinal, where variables on the predictor side (e.g., family, parent, peer variables) and variables on the outcome side (e.g., medical adherence, psychosocial adjustment) are all assessed over time, particularly during critical developmental periods or transition points (e.g., early childhood, the transition to elementary school, the early adolescent transition, the transition to early adulthood). More specifically, the quality of research studies will improve if they are longitudinal *and* if indices of developmental level and variables that are developmentally-relevant are included (e.g., pubertal status, changes in cognitive developmental level, changes in level of peer intimacy, autonomy development, changes in parenting behaviors). It is not enough to simply document whether a certain outcome increases or decreases over time. Instead, it is of interest to track important outcomes over time (e.g., adherence) as a function of changes in important developmental processes. Simply put, a study of children, adolescents, or emerging adults becomes developmentally-oriented when the researcher includes measures that tap constructs such as those noted in Figure 1 and when development *and* outcome are both tracked longitudinally.

Third, it is recommended that work not only be conducted on deficits in family functioning, but also on areas of resilience (i.e., adaptive functioning despite exposure to stressors or risk factors) that can be the basis for future interventions [Kazak et al., *2009*; Singh, *2003*[. Given the mixed findings of past work, it appears that a resilience-disruption perspective should be given serious consideration in future work.

Fourth, regarding sampling and methods of data collection, it is recommended that future studies include families with more ethnic and SES diversity. Most importantly, Hispanic/Latino families are understudied in this literature. This is surprising given the high

prevalence rates of SB in this population [Lary and Edmonds *1996*]. Studies can also be improved by examining the perspectives of multiple family members (mothers, fathers, *and* children) and employing multiple methods.

Fifth, several research design issues should be addressed in future work [Holmbeck Zebracki et al., *2009*]. Small sample sizes with wide age ranges make it nearly impossible to have adequate representation of the population under investigation and also produce samples that are under-powered for data analyses. When these limitations are combined with group matching problems (where the samples to be compared differ significantly on multiple demographic variables), such a study will yield few interpretable findings. It is recommended that investigators conduct power analyses before collecting data [Cohen *1992*] and that methods be used to produce matched samples of SB and comparison groups. One strategy is to recruit comparison families from the same schools that include children with SB [see Holmbeck Johnson et al., *2002*, for an example of this strategy]. An alternative strategy would be to select psychometrically-sound measures with normative data that could be used for comparison.

In conclusion, using theoretical models such as the bio-neuropsychosocial model to inform our research studies will help move the field towards a better understanding of the various factors that influence child psychosocial adjustment and family functioning in youth with SB. Although we have made progress in these areas, current areas of high priority for research include: (1) evaluating longitudinal models of psychosocial outcomes and examining mediational processes within these models; (2) specifying individual, family, and parenting variables that predict successful achievement of adolescent and emerging adulthood milestones; and (3) identifying areas of resilience and factors associated with resilience in youth with SB and their families.

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Figure 1.
A Bio-Neuropsychosocial Model of Psychosocial Adjustment in Children, Adolescents, and Emerging Adults with Spina Bifida