

Family Functioning in Children and Adolescents with Spina Bifida: An Evidence-based Review of Research and Interventions

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ABSTRACT. Research on the adjustment of families of children with spina bifida is reviewed, with a focus on delineating the impact of spina bifida on family functioning, the strengths and weaknesses of past research, and the needs for future evidence-based research on family interventions with this population. PsychINFO and MEDLINE literature searches were used to identify studies of family functioning and family-based interventions for children with spina bifida. Identified studies were empirically evaluated for the presence or absence of key methodological or analytic criteria. Thirty-two studies of family functioning were identified from 25 separate research groups; most studies displayed significant methodological limitations. No published studies of interventions to promote adaptive family functioning were identified. Methodologically sound, longitudinal, and theory-driven studies of family functioning are needed, as are randomized family-based intervention trials to promote adaptive functioning and better psychosocial outcomes in families of children with spina bifida. Specific recommendations for future work as well as clinical implications are noted. *J Dev Behav Pediatr* 27:249-277, 2006. Index terms: *spina bifida, family interventions, adaptive family functioning.*

Spina bifida is one of the most common congenital birth defects, affecting roughly 1 of every 1,000 live births.¹ It is caused by a failed closure of 1 or more vertebrae during the early weeks of gestation. Associated health complications include weakened or paralyzed lower extremities, urinary and bowel incontinence, and hydrocephalus. The severity of spina bifida varies in accordance with the spinal lesion level and neurological complications (e.g., the number of shunt replacements and infections).

Such clinical symptoms require intensive medical management and place considerable physical, psychological, and social demands on the individuals and families involved.²⁻⁵ Indeed, parents of children with spina bifida appear to experience more stress than parents of typically developing children.^{6,7} Similarly, children with spina

bifida appear to be at risk for higher rates of adjustment problems, including internalizing and social problems and difficulties with attention and concentration.^{8,10}

It is also the case, however, that there is considerable variability in the degree to which such children, their parents, and their siblings experience stress and adjustment difficulties.^{4,6} Indeed, some research has documented considerable resilience in these families.¹¹⁻¹³ As a result, investigators have attempted to isolate predictors of such variability and/or suggest mechanisms that buffer, exacerbate, or mediate the impact of family stress on child adjustment outcomes (e.g., demographics, coping resources, illness appraisal, social support, parental psychopathology, and quality of parenting).^{14,15}

The current article focuses on the adjustment of families of children and adolescents with spina bifida. Family relationships are particularly salient and influential social relationships for youth with spina bifida, given past research which suggests that children with spina bifida tend to be more socially isolated from their peers than are typically developing children.⁹ Given this focus, one might

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ask why we reviewed the literature on just 1 condition and why we chose spina bifida as the focus. Although a review that cuts across multiple conditions (i.e., a noncategorical approach) would aid in isolating generic illness dimensions that are associated with family adjustment outcomes, illnesses vary considerably across multiple dimensions (e.g., illness visibility and neurological impact). Therefore, the impact of each illness type on family functioning is likely to vary widely, depending on which dimensions are present for a given illness. For this reason, we decided to focus on a single chronic condition.

Why did we choose to focus on spina bifida? As noted above, spina bifida is a complex condition that impacts on most domains of child functioning. All of the following are condition-related stressors that will likely have a cumulative and pervasive impact on family functioning and family relationships: (1) the cognitive and neurological effects of spina bifida (e.g., executive functioning deficits, attention problems, and learning difficulties), (2) the effects of spina bifida on physiological development (e.g., precocious puberty is more common in this population than in the general population), (3) the multiple surgical procedures (e.g., shunt revisions and orthopedic surgeries), (4) difficulties with bowel and bladder control and management as well as the ambulation difficulties, (5) social skills deficits, and (6) difficulties in mastering developmental milestones (e.g., autonomy development). Given the potential multifaceted impact of spina bifida, a study of such a condition makes it more likely that we will detect an effect of condition on family functioning if an effect actually exists (as opposed to a study of a less severe condition). From a developmental psychopathology perspective, studies of atypical populations (such as spina bifida) can also provide information about typically developing youth. In the present instance, a review of the literature on family functioning in children with spina bifida can provide general information about whether and how the quality of a child's functioning impacts on family functioning.

Specifically, we sought answers to the following questions.

(1) How does spina bifida impact on family functioning (parent psychosocial adjustment, sibling adjustment, and family systemic functioning), and what is the evidence in support of a significant "impact"? (2) What are the methodological strengths and limitations of past research on the "impact" of spina bifida? (3) What are the needs for future evidence-based research on family functioning? (4) What evidence is there to support the use of family interventions to improve family functioning? (5) What are the methodological strengths and limitations of past research on family interventions? and (6) What are the needs for future evidence-based research on family interventions? Finally, conclusions based on this evidence-based review are provided. Interestingly, only 2 attempts have been made to review this literature, with one being 20 years old¹⁶ and the other only reviewing a small subset of the available published articles.⁵ Thus, the goal of this review was to provide a thorough evaluation of the literature on families of children with spina bifida since the earlier Spaulding and Morgan¹⁶ review by focusing on the strengths and weaknesses of this literature and discussing fruitful future directions.

THEORETICAL AND CONCEPTUAL BACKGROUND

Although the goal of this discussion is not to present a comprehensive theory of family functioning in children with spina bifida (e.g., see Kazak et al² and Rolland¹⁷), it is important to discuss where family functioning fits into a more general conceptualization of psychosocial functioning in children with spina bifida. Thus, we provide a bioneuro-psychosocial model of psychological adjustment in children with spina bifida (Fig. 1). As illustrated in Figure 1, the adjustment of children with spina bifida is likely determined by the interacting influences of multiple biological, neuropsychological, and social factors. That is, family functioning is but 1 factor that impacts on levels of child psychosocial adjustment. Moreover, all of these influences 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 indicated in the figure evolve with development and over time).

Each construct within Figure 1 can be considered a second-order domain with multiple subdomains. For example, the family domain includes multiple subdomains, such as the following: parental adjustment, parenting behaviors, parenting satisfaction, parenting stress, family system-level constructs (e.g., conflict, affect, and cohesion), family burden, family problem-solving abilities, family coping, family life events, and marital functioning; some of which have proved to be sensitive to the presence of a child with spina bifida (see review below). Not only are there multiple subdomains, but also multiple ways to assess each subdomain as well (e.g., questionnaire vs observational methods and parent vs child report). Moreover, the manner in which spina bifida 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 spina bifida is in grade school, but has difficulty adjusting to new parenting roles when the same child transitions into adolescence. Similarly, the way in which spina bifida impacts upon a family can also vary between family systems. For example, some families may have access to an extensive support network, whereas others may not. Such factors, when present, may contribute to a more resilient or adaptive family system. Teasing apart the illness-related factors that are most likely to have an impact upon the family system is a challenging task. It is our hope that, by pooling the research in this area of the literature, we can begin to understand ways in which spina bifida significantly impacts family functioning.

Family functioning is also viewed as 1 of several social influences, which also include peer-related factors (Fig. 1). Moreover, within this social domain, a child's family relationships and peer relationships may impact each other as each influences child adjustment. At the most complex level, the influence of any one factor in the model may interact with other factors in influencing child adjustment. For example, the impact of parenting and family relationships on child medical adherence may be moderated by the child's level of neuropsychological functioning. That is, some forms of parenting may be more

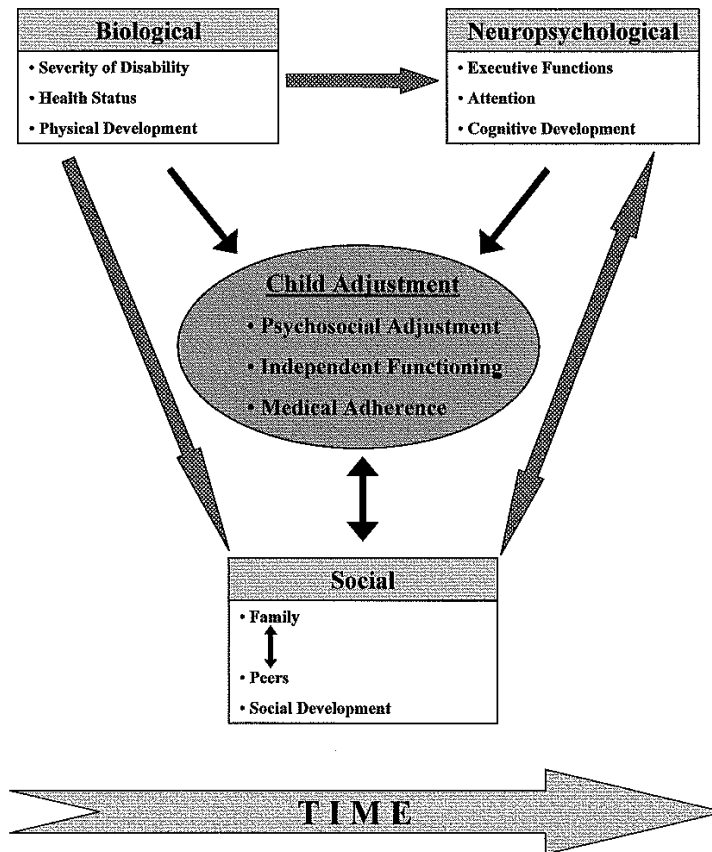


FIGURE 1. A bio-neuropsychosocial model of psychological adjustment in children with spina bifida.

effective in supporting a child’s medical adherence if certain attentional and executive functioning abilities are present in the child (i.e., higher order cognitive abilities such as problem solving, planning ability, organizational skills, and mental flexibility)^{18–20} than if such abilities are not present.

Finally, there is certainly some overlap between constructs that are included in different domains. For example, although medical adherence is viewed by many as a medically related adjustment outcome, it is also intimately intertwined with family process, particularly during the period of development when medical regimen responsibilities are shared between parent and child. In short, although the focus of this evidence-based review is on family functioning, it is critical to note that such functioning must be understood within the broader context of multiple biological, neurological, psychological, social, and contextual factors.

We also maintain that an understanding of family functioning in children with chronic conditions must be firmly grounded within a developmental framework.^{21,22} Space considerations do not permit a complete discussion of the relevant issues (see Holmbeck²¹ for a more complete discussion), but it is clear that the family management of a chronic illness is often at odds with the typical

developmental changes of childhood and, particularly, with the typical developmental changes of adolescence. With respect to adolescence, the cognitive developmental changes of this period make it more likely that youth with chronic medical conditions will increasingly come to view the management of their illness as falling within their own decision-making jurisdiction. In response to such developmental change, and as noted earlier, parents may begin to share more medical responsibilities with their offspring.²³ Thus, the multiple familial processes that govern child outcomes in youth with spina bifida (or any other illness for that matter) are dynamic in nature, and the salience of each family-oriented contributor to child outcome is likely to change over time as well.^{24,25}

HOW DOES SPINA BIFIDA IMPACT ON FAMILY FUNCTIONING AND WHAT IS THE EVIDENCE IN SUPPORT OF A SIGNIFICANT “IMPACT”?

Our review of “impact” articles covers 32 studies published between 1986 and 2002; all of which focus on some aspect(s) of family functioning in children and adolescents with spina bifida (see results of review in Appendix A). Inclusionary criteria for the review were as follows: (1) the article was published in or after 1986; (2)

the article included families of children or adolescents with spina bifida in the sample; (3) the study emerged based on the following search key words in PsychINFO or MEDLINE: spina bifida, myelomeningocele, meningo-myelocele, family, parent, and adjustment; (4) the study focused, at least in part, on the impact of spina bifida on some aspect of family functioning; and (5) the study was published in a peer-review journal. Dissertations and other unpublished work were not reviewed because of our intention to review the literature that is easily accessible to physicians, psychologists, and other health professionals. One implication of Criterion 2 above is that some studies focused exclusively on families of children with spina bifida, whereas others included such families as part of a larger study of families of children with any one of several chronic illnesses. This latter type of study rarely provides findings separately by illness group; thus, findings from these studies are less likely to generalize to families of children with spina bifida than studies that focused only on families of children with spina bifida. Appendix A includes a thorough review of the 32 studies based on 25 independent data sets (these 32 published studies are asterisked in the References section).

With respect to authors, only 5 authors (Ammerman, Barakat, Holmbeck, Kronenberger, and Williams) have published more than 1 article in this area, and only 4 journals (*Journal of Pediatric Psychology*, *Journal of Developmental and Behavioral Pediatrics*, *Children's Health Care*, and *Developmental Medicine and Child Neurology*) include more than 1 article on the topic of functioning in families of children with spina bifida. Thus, this literature tends to be fragmented, with few programmatic lines of research. Interestingly, the articles were published across numerous disciplines: psychology, psychiatry, pediatrics, family studies, disability research, nursing, and rehabilitation medicine. Although a multidisciplinary focus is a strength of this literature, such a broad focus may also contribute to the fragmented nature of this body of research.

Methodologically, sample sizes ranged from 7 to 201 (with the next highest n after 201 being 68). The mean sample size was 44.8 (38.3 without the $n = 201$ study). The total number of participants with spina bifida across studies was $N = 1121$. With respect to comparison samples, nearly 50% of the 25 nonoverlapping data sets included only families of children with spina bifida (i.e., no comparison samples; $n = 12$; 48%), 28% ($n = 7$) included an able-bodied comparison sample, and 24% ($n = 6$) included either a different chronically ill comparison sample (e.g., cerebral palsy and cystic fibrosis) or a mixed chronic illness sample. Only 12% (3/25) of the data sets included observational data, and only 33% included responses from both parents and children. Some studies had very large age ranges (differences between the youngest and oldest child in each sample ranged from 1–30 years), with the average age range covering 12.0 years from youngest to oldest. Only 1 of the 25 data sets was longitudinal.

With respect to measures used, and as noted earlier, multiple constructs have been assessed using multiple methods and with multiple measures within each method.

For example, with respect to “family environment,” the Family Assessment Device (FAD),^{26,27} the Family Environment Scale,^{11,16} and coding of observed family interactions^{11,28} are just some of the measures that have been used to assess multiple family-level systemic constructs (e.g., cohesion and conflict). In the area of parenting behaviors and parenting attitudes, the Child Report of Parent Behavior Inventory,^{29,30} the Hereford Parent Attitude Scale,¹⁶ coding of observed parenting behaviors,^{29,30} and the Parenting Stress Index^{6,31} have all been used. Finally, for parental psychological functioning, investigators have relied on the Brief Symptom Inventory (BSI),³² the Symptom Checklist 90,^{6,33,34} and the Center for Epidemiological Studies—Depression Scale.³¹

Although most studies in this review used previously validated measures of family and parent functioning, most of the measures have been validated only with families of typically developing children. Most authors do not report extensive psychometric information for the measures (see Kronenberger and Thompson³⁵ for an exception); thus, the validity of their use in populations of children with spina bifida, or more generally for children with chronic medical conditions, is not well understood. Finally, the research examined in this review has tended to focus on general family processes, with less attention to family processes that are specific to the management of spina bifida (e.g., conflict around completion of illness-management tasks, degree of parenting stress associated with illness management, etc.).

Between-group comparison studies yield more information about the impact of spina bifida (vs no spina bifida) on family functioning than do within-group studies. Thus, when reviewing the findings of the studies, results from these studies will be emphasized. Comparisons between a spina bifida sample and a comparison sample (a matched comparison sample or a normative sample) were conducted in only 14 of 25 data sets. Significant differences between groups were found in only 6 of these data sets^{11,26,27,33,36,37} (findings based on the Ammerman, Holmbeck, and Kronenberger data sets appear in 2 or more published articles, as noted in Appendix A). Thus, group differences that are reported here should be interpreted against a backdrop of mixed findings across the larger literature.

On the other hand, significant differences were more likely to be found when (1) the study had a larger sample size, (2) the study had a stronger research design, and (3) the comparison was to normative data. Specifically, the average n for the 8 data sets where no group differences were found was 29.00 (range, 10–56). The average n for the 6 data sets where differences were found was 51.83 (range, 30–68). Moreover, there were 4 data sets where the samples being compared differed significantly on at least 1 demographic variable; interestingly, none of these studies yielded differences between spina bifida and able-bodied comparison families (i.e., in 4 of the 8 data sets that showed no differences, the “group” variable was confounded with at least 1 demographic variable, thus complicating interpretation). Given the possibility that the lack of significant differences in these 8 “no differences” studies may have been caused by less than adequate power or design flaws,

interpretations based on these 8 studies will be made cautiously, and findings from the 6 higher quality "group differences" studies will be emphasized in this section. In the next section, we will provide a critique of the full sample of 32 studies (see Appendix A).

Findings from 2 studies revealed that 12% to 13% of families of children with spina bifida exhibited clinical levels of "family dysfunction" (Ammerman et al²⁶ found that 13% of families were in the clinically problematic range, with *T* scores >62, on the General Functioning Scale of the FAD; Wiegner and Donders²⁷ found that 12% were in the clinical range on the same FAD scale). Such rates of family dysfunction are lower than those found in families of children with cerebral palsy (35%; Wiegner and Donders²⁷). A significant number of family members with children who have spina bifida report difficulties in maintaining clear roles and responsibilities in the family system (23% in the clinically problematic range in the study of Ammerman et al²⁶; also see Wiegner and Donders²⁷). On the other hand, many families of children with spina bifida evidence high levels of resilience. In fact, most 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.¹¹

Despite the relatively low levels of family dysfunction at the family systems level, it appears that a sizable minority of parents of children with spina bifida exhibits clinical levels of global psychological distress (e.g., anxiety, depressive symptoms, and somatic complaints).^{6,33} In one study, 41% of parents scored more than a *T* score of 62 on the General Severity Index of the BSI or more than a *T* score of 62 on at least 2 of 9 BSI subscales.²⁷ Although most studies that report on parental functioning have focused on maternal functioning, fathers exhibited higher levels of global distress than comparison families in one study.⁶ Across several studies, parents of children with spina bifida tended to experience more stress in their roles as parents than did comparison parents. 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.^{6,32,38} It appears that parents who are single, socially isolated, older, or from an ethnic minority or low socioeconomic status (SES) background are particularly at risk for such outcomes.^{11,34,39-41}

The following areas of family functioning have also received attention: marital functioning, sibling adjustment, and quality of parenting. With respect to marital functioning, the findings are mixed. Some studies show no differences in marital functioning between families of children with spina bifida and able bodied.^{6,16,31} Interestingly, at least one study found that having a child with a disability can strengthen a marriage.^{12,31} It appears that the quality of the marital relationship before the birth of the affected child is an important predictor of the subsequent adjustment of family members. The few studies of siblings show few differences in the adjustment of siblings in spina bifida versus comparison samples.^{42,43} Parents of children with spina bifida tend to exhibit higher levels of over-

protectiveness, psychological control (i.e., parenting that undermines the autonomy development of their offspring), and authoritarian parenting.^{29,30} On the other hand, group differences on these variables appear to be mediated by child cognitive ability, such that children with spina bifida tend to have lower IQs, and children with lower IQs tend to have parents who are more controlling.²⁹

Severity of illness was related to parental and family adjustment in several studies, although findings are mixed. In 2 studies, illness severity was positively correlated with family dysfunction,^{33,44} but across 3 studies, lesion level was positively related to maternal confidence in the parenting role, marital satisfaction, attachment to child, and negatively related to conflict frequency.^{12,31,45}

In summary, although the findings of past research are mixed with respect to whether spina bifida impacts on the family environment (with some studies showing group differences and others showing no group differences), studies that do show differences tended to have larger *n*'s and had better design features (e.g., comparison samples were well matched on demographic variables). When differences across groups occurred, findings suggest relatively low rates of family-level dysfunction (and high levels of family resilience) but higher rates of parental psychological distress and parenting stress particularly for mothers, single parents, older parents, and parents from low SES and ethnic minority backgrounds. Thus, a family with several of these characteristics would be particularly at risk for parental adjustment difficulties. In general, the findings of past work support a resilience-disruption view of family functioning (see Costigan et al⁴⁶ for a description of this view). That is, spina bifida 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.

WHAT ARE THE METHODOLOGICAL STRENGTHS AND LIMITATIONS OF PAST RESEARCH ON THE "IMPACT" OF SPINA BIFIDA?

Our review of past work on the "impact" of spina bifida revealed both strengths and weaknesses (as detailed in Appendix A). Regarding methodological strengths of past research, many studies have used measures with adequate psychometric characteristics or have attempted to verify illness severity with information from medical chart reviews. In addition, the inclusion of able-bodied or chronically ill comparison samples in many studies is a strength, and the involvement of ethnic minority families in some studies has contributed to the generalizability of findings. Finally, the assessment of both child and parental perspectives, coupled with the collection of multisource multimethod data in some investigations, is a strength that helps to rule out common method variance explanations of findings, as well as ensure that family functioning is evaluated from several perspectives.

Regarding statistical or analytic strengths, some recent studies focused on the clinical significance of findings (e.g., the number of families who score more than certain

T score cutoff levels in relation to normative data.)²⁷ Of course, we cannot determine the number of “families” that meet diagnostic criteria (given that available diagnostic systems focus on individual functioning, we can use family-based measures that have adequate psychometric integrity and normative data to determine the frequency with which families fall in the clinical range). In addition, some recent studies have recruited sample sizes with an adequate *n* to ensure that analyses are not underpowered, thereby increasing the likelihood of detecting a significant effect if one is present. Finally, when large samples are available, the use of sophisticated data analytic techniques, such as structural equation modeling (SEM; i.e., path analytic models that posit multiple causal pathways between variables), has afforded the opportunity to address new research questions that more traditional analytic techniques cannot answer adequately.

Although the above strengths are evident in some studies, the majority of studies reviewed were hampered by significant design flaws and other limitations (see Appendix A; also see Singh⁵ for a recent review of a subset of these studies with similar conclusions). Several methodological weaknesses of past research were evident (*n*'s listed in parentheses refer to the number of studies for which this weakness was evident and are based on the total of 25 nonoverlapping data sets). First, nearly half of the studies sampled only white families (*n* = 11), whereas more than 25% relied solely on middle-class participants (*n* = 7); therefore, the degree to which we can generalize findings to diverse populations is limited. Similarly, the failure to include information on the ethnic background of the sample (*n* = 2) in 2 cases makes generalizations to the larger population of families affected by spina bifida impossible. Generalizability to populations of families from low SES and ethnic minority backgrounds may be particularly important, given evidence to suggest that these groups are more at risk for difficulties with family functioning (see above).

Second, reliance on single-source (*n* = 12) and/or single-method (*n* = 9) data collection strategies was common, making it difficult to rule out common method variance explanations for significant findings. That is, it may be that a significant association between 2 variables is caused by biases inherent in having the same respondent report on both variables than to an actual overlap between the 2 constructs assessed (e.g., parents who are depressed may be biased in their reports of all family-related constructs; such biases will artificially enhance correlations between parental symptoms and family functioning variables). Many studies are based solely on maternal report, with nearly half of all studies reviewed failing to include data from the perspective of fathers (*n* = 12), despite a growing recognition of the unique contribution of fathers to individual and family functioning. Similarly, more than 60% of studies failed to include both parent- and child-reported data (*n* = 16). Moreover, several studies relied exclusively on telephone or mail-in, data-gathering strategies (*n* = 4), whereas nearly all studies failed to include observational indices of family functioning (*n* = 22). Third, although more than half of all studies used comparison

samples, between-group differences on demographic variables were evident in many cases (*n* = 4 of 14 studies where comparisons between groups were made), thus producing confounded group comparisons in data analyses which threaten the internal validity of findings (see earlier discussion). In several cases, no comparison sample was used at all, and no attempt was made to compare data to a normative sample (*n* = 11), making interpretation of findings difficult.

Regarding analytic or statistical weaknesses, the cross-sectional nature of the data in all but 1 study (*n* = 24) precludes statements about directionality of effects and does not allow for the use of analytic techniques to investigate enduring patterns of family functioning over time. In addition, in a few cases, researchers used inappropriate data analytic strategies (*n* = 3) to address their question of interest. Finally, with respect to theoretical weaknesses, the large age ranges used in many past studies were insensitive to the particular developmental issues that are unique to early childhood, middle childhood, and adolescence. Moreover, the lack of a sound theoretical framework makes the development of appropriate hypotheses and the inclusion of suitable measures challenging and makes it more difficult to understand the meaning and importance of any significant findings.

WHAT ARE THE NEEDS FOR FUTURE EVIDENCE-BASED RESEARCH ON FAMILY FUNCTIONING?

Given these strengths and limitations of past work, 5 recommendations for future work are provided:

Recommendation 1

Most past investigations lack a theoretical framework. It is recommended that future work be theory-driven, where hypotheses, measure selection, and statistical strategies follow directly from a theoretical framework. For example, prospective mediational prediction models where intervening mechanisms are proposed are likely to yield significant and useful information, which will have important implications for interventions. For example, Holmbeck, Johnson et al²⁹ found that associations between intrusive parenting and child adjustment outcomes in families of children with spina bifida 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, when mediational prediction models are applied in studies that examine differences between spina bifida 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. Suppose one finds that spina bifida and able-bodied samples differ on level of parenting stress. If one also finds that this relationship is mediated by parental social support (group→social support→parenting stress), such a model suggests that having a child with spina bifida results in more parenting stress and that 1 mechanism by which this occurs is through decreases in parental social support (see Holmbeck^{47,48} and Rose et al⁴⁹ for a more

complete explanation of mediational models). Such a finding would have implications for interventions (e.g., assisting parents in developing support networks may be an effective way of reducing their levels of stress).

The literature on family functioning in children with spina bifida 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,²⁴ (3) models that examine mediational processes which potentially explain group differences, 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).

Recommendation 2

It is recommended that more work be programmatic and longitudinal, where variables on the predictor side (e.g., family, parent, and peer variables) and variables on the outcome side (e.g., medical adherence and psychosocial adjustment) are all assessed over time, particularly during critical developmental periods or transition points (e.g., early childhood, the transition to school, and the early adolescent transition, the transition to adulthood). It appears that multidisciplinary studies which focus on multiple domains (as illustrated in Fig. 1) will yield the most important information on the functioning of families of children with spina bifida.

Recommendation 3

It is recommended not only that work be conducted on deficits in family functioning, but also that future research attempt to isolate areas of resilience (i.e., adaptive functioning despite exposure to stressors or risk factors) that can be the basis for future interventions.^{2,5} Some past work has found that families of children with spina bifida exhibit resilience across several areas of functioning. In fact, Holmbeck et al¹¹ found support for a resilience-disruption view⁴⁶ of systemic functioning of families of children with spina bifida. Specifically, families in the spina bifida sample exhibited lower levels of cohesion (i.e., disruption) but did not differ from comparison families on measures of conflict or negative life events (i.e., resilience). Given the mixed findings of past work (see review above), it appears that a resilience-disruption perspective should be given serious consideration in future work.

Recommendation 4

Regarding sampling and methods of data collection, it is recommended that future studies include samples 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 spina bifida in this population.⁵⁰ Studies can also be improved by examining the perspectives of multiple family members (mothers, fathers, and children) and using multiple

methods (e.g., as noted above, only 3 of 25 data sets included observational data on families).

Recommendation 5

Several research design issues should be addressed in future work. 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 underpowered 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 data⁵¹ and that methods be put in place where spina bifida and comparison groups are sampled in such a way as to produce matched samples. One strategy is to recruit comparison families from the same schools that include children with spina bifida (see Holmbeck et al²⁹ for an example of this strategy). An alternative strategy would be to select psychometrically sound measures for which there exists normative data that could be used for comparison.

In fact, the choice of a comparison sample is a decision that will impact on the types of conclusions one can draw. If one seeks to determine whether a study's findings apply only to families of children with spina bifida or if they apply to a general population of families with children who have other chronic conditions, a comparison sample that includes children with another type of chronic condition would be most appropriate (see Wade et al⁵² for an example of this strategy). For example, one could compare conditions with and without central nervous system involvement or one could compare a condition which has physical manifestations that are apparent to others versus one where the physical effects are not apparent to others. With respect to small sample sizes, it is recommended that multisite projects be initiated; such projects will generate larger sample sizes and permit more sophisticated data analytic strategies.

WHAT EVIDENCE IS THERE TO SUPPORT THE USE OF FAMILY INTERVENTIONS TO IMPROVE FAMILY FUNCTIONING?

As noted above, our literature review of studies that examined the impact of spina bifida on family functioning yielded 32 studies based on 25 data sets. Contrary to this, our review of studies that examined interventions for families of children with spina bifida yielded no published studies. The literature search was conducted in a manner similar to that used above. PsychINFO and MEDLINE were searched using the following key words: chronic illness, chronic disability, spina bifida, family treatment, family intervention, family therapy, group therapy, empirically validated treatment, empirically supported treatment, pediatric, parent, parent training, efficacy, and adherence.

Thus, the answer to this question is simply: there is no evidence that supports or fails to support the use of family interventions to improve family functioning. Given that

there were no studies to review, we will bypass our fifth question (i.e., what are the methodological strengths and limitations of past research on family interventions?) and move directly to the final question posed above.

WHAT ARE THE NEEDS FOR FUTURE EVIDENCE-BASED RESEARCH ON FAMILY INTERVENTIONS?

Given the lack of published intervention work on families of children with spina bifida, we draw on past intervention work in families of children with other chronic illnesses (e.g., diabetes, end-stage renal failure, cancer, asthma, sickle cell disease, painful medical procedures, cystic fibrosis, neurogenic bowel, and pediatric AIDS) in making recommendations for future intervention work in this literature. We also draw extensively on recommendations in the special series on empirically supported treatments in pediatric psychology published over several issues of the *Journal of Pediatric Psychology* from 1999 to 2001 (also see Rodrigue⁵³). Interestingly, interventions for some aspect of family functioning exist for nearly every pediatric illness, except spina bifida. Thus, our primary recommendation is that some form of intervention work begin immediately with families of children with spina bifida. We also provide recommendations regarding areas that can be addressed with interventions as well as recommendations for how such work might be conducted to yield the most interpretable data.

In the field of psychology, there are clear criteria that enable one to determine whether an intervention is "empirically supported" or "evidence based"⁵⁴; these criteria have become known as the "Chambless criteria," named after the Chair of this task force.⁵⁵ As revealed in the special series of the *Journal of Pediatric Psychology*, few interventions of any kind with pediatric populations (including family-focused interventions) meet the criteria for a "well-established" treatment.^{24,56} Although it is unfortunate that no family interventions have been tested with families of children with spina bifida, the lack of such work is also an opportunity. Given that clear criteria exist for demonstrating evidence-based interventions, we will be able to develop these new interventions by using state-of-the-art strategies for designing effective family interventions. Given that this literature is currently in its infancy, we assume that controlled efficacy studies will be conducted first, followed by "real world" effectiveness efforts.

Recommendation 1

We recommend that the targets (i.e., outcomes) of interventions for families of children with spina bifida be clearly delineated. Family-based interventions could focus on the following: (1) medical adherence (e.g., involving catheterization and bowel programs; see Holmbeck et al⁵⁷ for information on a measure of adherence in children with spina bifida), (2) parent or marital stress⁵⁸ and parenting behaviors, (3) social adaptation of children and adolescents, (4) sibling adjustment, (5) the development of autonomy and independent functioning, and (6) managing

the transition to adulthood. Although different family interventions could be developed to target each of these important outcomes, it is also likely that several of these outcomes could be targeted by a single intervention. For example, medical adherence could be addressed in the context of working on autonomy development and independent functioning as well as focusing on managing the transition to adulthood.

Given that family-based interventions targeting medical adherence are among the most advanced in the field of pediatric psychology,^{59,60} this may be a good place to start. For example, multifamily interventions have been used to improve the adherence of children with Type 1 diabetes.⁶¹ Behavioral family systems therapy has been used to reduce diabetes-related⁶² and cystic fibrosis-related⁶³ family conflicts and increase medical adherence, although this intervention appears to be more effective at reducing conflicts than increasing adherence behaviors. Anderson et al⁶⁴ have proposed a "teamwork" intervention for adherence in adolescents with Type 1 diabetes that emphasizes parents and teens sharing responsibility for diabetes-related tasks. Interventionists could begin by testing modified versions of "well-established" or "probably efficacious" treatments from the aforementioned literatures with families of children with spina bifida.

Recommendation 2

Based on theory, investigators can propose mechanisms that account for the onset and maintenance of the problem outcome (e.g., low adherence, high parenting stress) as well as mediational mechanisms that may account for significant treatment effects.

Recommendation 3

We recommend that interventions for families of children with spina bifida be manualized and that manuals be flexible so that treatments can be matched to each family (perhaps via a therapeutic "toolbox" approach).⁶⁵ This strategy may be particularly important for families of children with spina bifida, given the variability in child cognitive and neuropsychological functioning.

Recommendation 4

It is recommended that interventions be adapted for use with ethnic minority families, particularly Spanish-speaking Hispanic/Latino families. As noted earlier, this population of families of children with spina bifida is underserved, and we have virtually no data on how these families function. Culturally sensitive treatments that address the specific strengths and vulnerabilities of these families are sorely needed.⁵⁶ Reducing barriers to intervention and increasing engagement in interventions once enrolled are significant hurdles that must be confronted.

More generally, interventions should be developed to target at-risk groups. Earlier, we described how single parents, parents from low SES homes, older parents, and parents from minority backgrounds are at risk for experiencing higher levels of parenting stress than are other parents. If treatments

are developed with these groups in mind, they are more likely to be effective for those in greatest need.

Recommendation 5

It is recommended that “modifiable” spina bifida–relevant variables be selected as targets for family-based interventions and as outcomes. For example, family-oriented interventions might focus on problem-solving strategies of families of children with spina bifida (including conflict management), facilitation of shared responsibility for the management of and adherence to one’s medical regimen, parenting stress, parenting behaviors (including intrusive parenting), and family coping strategies.²⁴

CONCLUSIONS AND CLINICAL IMPLICATIONS

The purpose of this article was to review current knowledge regarding the impact of spina bifida on family functioning and the effectiveness of existing family interventions. We concluded that the “impact” findings were mixed but tended to support a resilience-disruption perspective in families of children with spina bifida. Such families appear to be at risk for high levels of parenting stress, although certain demographic groups appear to be most at risk. Numerous weaknesses of past work were identified, and several recommendations for future work were discussed. Surprisingly, no studies of family inter-

ventions were found in the existing literature. More generally, it is our hope that this review will stimulate more evidence-based research on families of children and adolescents with spina bifida.

The findings of this review have several clinical implications. For example, interventions should target the most at-risk families (i.e., single parent, those of low SES, and ethnic minority families) and should focus on parenting stress. In addition, basic research and intervention research should focus on similar variables so that knowledge resulting from the former can inform designs for the latter. Unfortunately, we still have only a modest amount of data on many of the constructs which are relevant to families of children with spina bifida (e.g., how these families facilitate shared responsibility for illness management and family coping strategies). Moreover, few spina bifida clinics have incorporated interventions focused on psychosocial or family issues. It is likely that considerable benefit would be derived from integrating such strategies into comprehensive care programs.

Acknowledgments. This manuscript was supported in part by research grants from the National Institute of Child Health and Human Development (NICHD; R01 HD048629) and the March of Dimes Birth Defects Foundation (12-FY01-0098).

Based on presentation at the conference “Evidence-Based Practice in Spina Bifida: Developing a Research Agenda,” May 2003, Washington, DC.

REFERENCES

- McLone DG, Ito J. *An Introduction to Spina Bifida*. Chicago, IL: Children’s Memorial Spina Bifida Team; 1998.
- Kazak AE, Rourke MT, Crump TA. Families and other systems in pediatric psychology. In: Roberts M, ed. *Handbook of Pediatric Psychology*, 3rd ed. New York: Guilford; 2003:159–175. chapter 10.
- Quittner AL, DiGirolamo AM. Family adaptation to childhood disability and illness. Boston: Allyn & Bacon; 70–102. chapter 4.
- Shepard MP, Mahon MM. Family considerations. In: Hayman LL, Mahon MM, Turner JR, eds. *Chronic Illness in Children: An Evidence-based Approach*. New York: Springer; 2002:143–170. chapter 6.
- Singh DK. Families of children with spina bifida: a review. *J Dev Phys Disabil*. 2003;15:37–55.
- Holmbeck GN, Gorey-Ferguson L, Hudson T, et al. Maternal, paternal, and marital functioning in families of preadolescents with spina bifida. *J Pediatr Psychol*. 1997;22:167–181.
- Wallander JL, Pitt LC, Mellins CA. Child functional independence and maternal psychosocial stress as risk factors threatening adaptation in mothers of physically or sensorially handicapped children. *J Consult Clin Psychol*. 1990;58:818–824.
- Appleton PL, Ellis NC, Minchom PE, et al. Depressive symptoms and self-concept in young people with spina bifida. *J Pediatr Psychol*. 1997;22:707–722.
- Holmbeck GN, Westhoven VC, Phillips WS, et al. A multi-method, multi-informant, and multi-dimensional perspective of psychosocial adjustment in pre-adolescents with spina bifida. *J Consult Clin Psychol*. 2003;71:782–796.
- Lavigne JV, Faier-Routman J. Psychological adjustment to pediatric physical disorders: a meta-analytic review. *J Pediatr Psychol*. 1992;17:133–157.
- Holmbeck GN, Coakley RM, Hommeyer JS, et al. Observed and perceived dyadic and systemic functioning in families of preadolescents with spina bifida. *J Pediatr Psychol*. 2002;27:177–189.
- Kazak AE, Clarke MW. Stress in families of children with myelomeningocele. *Dev Med Child Neurol*. 1986;28:220–228.
- Masten AS. Ordinary magic: resilience processes in development. *Am Psychol*. 2001;56:227–238.
- Lavigne JV, Faier-Routman J. Correlates of psychological adjustment to pediatric physical disorders: a meta-analytic review and comparison with existing models. *Dev Behav Pediatr*. 1993;14:117–123.
- Murch RL, Cohen LH. Relationships among life stress, perceived family environment, and the psychological distress of spina bifida adolescents. *J Pediatr Psychol*. 1989;14:193–214.
- Spaulding BR, Morgan SB. Spina bifida children and their parents: a population prone to family dysfunction? *J Pediatr Psychol*. 1986;11:359–374.
- Rolland JS. Chronic illness and the life cycle: a conceptual framework. *Fam Process*. 1987;26:203–221.
- Dise JE, Lohr ME. Examination of deficits in conceptual reasoning abilities associated with spina bifida. *Am J Phys Med Rehab*. 1998;77:247–251.
- Fletcher JM, Dennis M, Northrup H. Hydrocephalus. In: Yeates KO, Ris MD, Taylor HG, eds. *Pediatric Neuropsychology: Research, Theory, and Practice*. New York: Guilford; 2000:25–46. chapter 2.
- Wills KE. Neuropsychological functioning in children with spina bifida and/or hydrocephalus. *J Clin Child Psychol*. 1993;22:247–265.
- Holmbeck GN. A developmental perspective on adolescent health and illness: an introduction to the special issues. *J Pediatr Psychol*. 2002;27:409–416.

22. Williams PG, Holmbeck GN, Greenley RN. Adolescent health psychology. *J Consult Clin Psychol.* 2002;70:828–842.
23. Anderson BJ, Ho J, Brackett J, et al. Parental involvement in diabetes management tasks: relationships to blood glucose monitoring adherence and metabolic control in young adolescents with insulin-dependent diabetes. *J Pediatr.* 1997;130:257–265.
24. Drotar D. Relating parent and family functioning to the psychological adjustment of children with chronic health conditions: what have we learned? What do we need to know? *J Pediatr Psychol.* 1997;22:149–165.
25. Murphy MA. The family with a handicapped child: a review of the literature. *J Dev Behav Pediatr.* 1982;3:73–82.
26. Ammerman RT, Kane VR, Slomka GT, et al. Psychiatric symptomatology and family functioning in children and adolescents with spina bifida. *J Clin Psychol Med Settings.* 1998;5:449–465.
27. Wiegner S, Donders J. Predictors of parental distress after congenital disabilities. *J Dev Behav Pediatr.* 2000;21:271–277.
28. Coakley RM, Holmbeck GN, Friedman D, et al. A longitudinal study of pubertal timing, parent-child conflict, and cohesion in families of young children with spina bifida. *J Pediatr Psychol.* 2002;27:461–473.
29. Holmbeck GN, Johnson SZ, Wills KE, et al. Observed and perceived parental overprotection in relation to psychosocial adjustment in preadolescents with a physical disability: the mediational role of behavioral autonomy. *J Consult Clin Psychol.* 2002;70:96–110.
30. Holmbeck GN, Shapera WE, Hommeyer JS. Observed and perceived parenting behaviors and psychosocial adjustment in preadolescents with Spina Bifida. In: Barber BK, ed. *Intrusive Parenting: How Psychological Control Affects Children and Adolescents.* Washington, DC: American Psychological Association; 2002:191–234. chapter 7.
31. Cappelli M, McGrath PJ, Daniels T, et al. Marital quality of parents of children with spina bifida: a case-comparison study. *Dev Behav Pediatr.* 1994;15:320–326.
32. Barakat LP, Linney JA. Optimism, appraisals, and coping in the adjustment of mothers and their children with spina bifida. *J Child Fam Stud.* 1995;4:303–320.
33. Kronenberger WG, Thompson RJ. Medical stress, appraised stress, and the psychological adjustment of mothers of children with myelomeningocele. *Dev Behav Pediatr.* 1992;13:405–411.
34. Kronenberger WG, Thompson RJ. Psychological adaptation of mothers of children with spina bifida: association with dimensions of social relationships. *J Pediatr Psychol.* 1992;17:1–14.
35. Kronenberger WG, Thompson RJ. Dimensions of family functioning in families with chronically ill children: a higher order factor analysis of the Family Environment Scale. *J Clin Child Psychol.* 1990;19:380–388.
36. Edwards-Beckett J. Parental expectations and child's self concept in spina bifida. *Child Health Care.* 1995;24:257–267.
37. Lemanek KL, Jones ML, Lieberman B. Mothers of children with spina bifida: adaptational and stress processing. *Child Health Care.* 2000;29:19–35.
38. Wallander JL, Varni JW, Babani L, et al. The social environment and the adaptation of mothers of physically handicapped children. *J Pediatr Psychol.* 1989;14:371–387.
39. Barakat LP, Linney JA. Children with physical handicaps and their mothers: the interrelation of social support, maternal adjustment, and child adjustment. *J Pediatr Psychol.* 1992;17:725–739.
40. Fagan J, Schor D. Mothers of children with spina bifida: factors related to maternal psychosocial functioning. *Am J Orthopsychiatry.* 1993;63:146–152.
41. Macias MM, Clifford SC, Saylor CG, et al. Predictors of parenting stress in families of children with spina bifida. *Child Health Care.* 2001;30:57–65.
42. Williams PD, Williams AR, Graff JC, et al. Interrelationships among variables affecting well siblings and mothers in families of children with a chronic illness or disability. *J Behav Med.* 2002;25:411–424.
43. Williams PD, Williams AR, Hanson S, et al. Maternal mood, family functioning, and perceptions of social support, self-esteem, and mood among siblings of chronically ill children. *Child Health Care.* 1999;28:297–310.
44. Bier JB, Liebling JA. Parents' and pediatricians' views of individuals with meningomyelocele. *Clin Pediatr.* 1996;35:113–118.
45. Holmbeck GN, Faier-Routman J. Spinal lesion level, shunt status, family relationships, and psychosocial adjustment in children and adolescents with spina bifida myelomeningocele. *J Pediatr Psychol.* 1995;20:817–832.
46. Costigan CL, Floyd FJ, Harter KSM, et al. Family process and adaptation to children with mental retardation: disruption and resilience in family problem-solving interactions. *J Fam Psychol.* 1997;11:515–529.
47. Holmbeck GN. Toward terminological, conceptual, and statistical clarity in the study of mediators and moderators: examples from the child-clinical and pediatric psychology literatures. *J Consult Clin Psychol.* 1997;65:599–610.
48. Holmbeck GN. Post-hoc probing of significant moderational and mediational effects in studies of pediatric populations. *J Pediatr Psychol.* 2002;27:87–96.
49. Rose BM, Holmbeck GN, Coakley RM, et al. Mediator and moderator effects in developmental and behavioral pediatric research. *Dev Behav Pediatr.* 2004;25:1–10.
50. Lary JM, Edmonds LD. Prevalence of spina bifida at birth—United States, 1983–1990: a comparison of two surveillance systems. *Morb Mortal Wkly Rep.* 1996;45:15–26.
51. Cohen J. A power primer. *Psychol Bull.* 1992;112:155–159.
52. Wade SL, Taylor HG, Drotar D, et al. Interpersonal stressors and resources as predictors of parental adaptation following pediatric traumatic injury. *J Consult Clin Psychol.* 2004;72:776–784.
53. Rodrigue JR. Beyond the individual child: innovative systems approaches to service delivery in pediatric psychology. *J Clin Child Psychol.* 1994;23:32–39.
54. Task Force on Promotion and Dissemination of Psychological Procedures. Training in and dissemination of empirically-validated psychological treatments: report and recommendations. *Clin Psychol.* 1995;48:3–23.
55. Lemanek KL, Kamps J, Chung NB. Empirically supported treatments in pediatric psychology: regimen adherence. *J Pediatr Psychol.* 2001;26:253–275.
56. Kaslow NJ, Collins MH, Loundy MR, et al. Empirically validated family interventions for pediatric psychology: sickle cell disease as an exemplar. *J Pediatr Psychol.* 1997;22:213–227.
57. Holmbeck GN, Belvedere MC, Christiansen M, et al. Assessment of adherence with multiple informants in pre-adolescents with spina bifida: initial development of a multidimensional, multitask parent-report questionnaire. *J Pers Assess.* 1998;70:427–440.
58. Walker JG, Johnson S, Manion I, et al. Emotionally focused marital intervention for couples with chronically ill children. *J Consult Clin Psychol.* 1996;64:1029–1036.
59. La Greca AM, Bearman KJ. Commentary: if "an apple a day keeps the doctor away," why is adherence so darn hard? *J Pediatr Psychol.* 2001;26:279–282.
60. La Greca AM, Bearman KJ. Adherence to pediatric treatment regimes. In: Roberts M, ed. *Handbook of Pediatric Psychology*, 3rd ed. New York: Guilford; 2003:119–140. chapter 8.
61. Satin W, La Greca AM, Zigo M, et al. Diabetes in adolescents: effects of multifamily group intervention and parenting simulation of diabetes. *J Pediatr Psychol.* 1989;14:259–275.
62. Wysocki T, Greco P, Harris MA, et al. Behavioral family systems therapy for adolescents with diabetes. In: Drotar D, ed. *Promoting Adherence to Medical Treatment in Chronic Illness: Concepts,*

- Methods, and Interventions*. Mahwah, NJ: Erlbaum; 2000:367–381. chapter 16.
63. Quittner AL, Drotar D, Ievers-Landis C, et al. Adherence to medical treatments in adolescents with cystic fibrosis: the development and evaluation of family-based interventions. In: Drotar D, ed. *Promoting Adherence to Medical Treatment in Chronic Illness: Concepts, Methods, and Interventions*. Mahwah, NJ: Erlbaum; 2000:383–407. chapter 17.
 64. Anderson BJ, Brackett J, Ho J, et al. An intervention to promote family teamwork in diabetes management tasks: relationships among parental involvement, adherence to blood glucose monitoring, and glycemic control in young adolescents with type 1 diabetes. In: Drotar D, ed. *Promoting Adherence to Medical Treatment in Chronic Illness: Concepts, Methods, and Interventions*. Mahwah, NJ: Erlbaum; 2000:347–365. chapter 15.
 65. Weisz JR, Hawley KM. Developmental factors in the treatment of adolescents. *J Consult Clin Psychol*. 2002;70:21–43.
 66. Ammerman RT, Van Hasselt VB, Hersen M. Parent-child problem-solving interactions in families of visually impaired youth. *J Pediatr Psychol*. 1991;16:87–101.
 67. Bower AM, Hayes A. Mothering in families with and without a child with disability. *Int J Disabil Dev Educ*. 1998;45:313–322.
 68. Havermans T, Eiser C. Mothers' perceptions of parenting a child with spina bifida. *Child Care Health Dev*. 1991;17:259–273.
 69. Horton TV, Wallander JL. Hope and social support as resilience factors against psychological distress of mothers who care for children with chronic physical conditions. *Rehab Psychol*. 2001;46:382–399.
 70. King G, King S, Rosenbaum P, et al. Family centered caregiving and well-being of parents of children with disabilities: linking process with outcome. *J Pediatr Psychol*. 1999;24:41–53.
 71. Loebig M. Mothers' assessments of the impact of children with spina bifida on the family. *MCN Am J Matern Child Nurs*. 1990;19:251–265.
 72. Loomis JW, Javornisky JG, Monahan JJ, et al. Relations between family environment and adjustment outcomes in young adults with spina bifida. *Dev Med Child Neurol* 1997;39:149–165.
 73. McCormick MC, Charney EB, Stemmler MM. Assessing the impact of a child with spina bifida on the family. *Dev Med Child Neurol* 1986;28:53–61.
 74. Samuelson JJ, Foltz J, Foxali MJ. Stress and coping in families of children with myelomeningocele. *Arch Psychiatr Nurs* 1992;6:287–295.
 75. Seefeldt T, Holmbeck GN, Belvedere M, et al. Socioeconomic status and democratic parenting in families of preadolescents with spina bifida. *Psi Chi J Undergrad Res* 1997;2:5–12.

Appendix A: Summary of Studies Assessing the Impact of Spina Bifida on Family Functioning

Authors	Title	Year	Publication Source	Method	Results	Comments
Ammerman et al ²⁶	Psychiatric Symptomatology and Family Functioning in Children and Adolescents with Spina Bifida	1998	<i>Journal of Clinical Psychology in Medical Settings</i>	Sample consisted of 54 children and adolescents with SB (ages 6-18 yr; mean age, 12.94 yr) Measures included parent report of child psychiatric symptoms and family functioning Data were collected via mail	43% of the sample met criteria for one psychiatric diagnosis, whereas 13% met criteria for 2 or more psychiatric diagnoses Most prevalent diagnoses among youth with SD included ADHD (33%) and oppositional defiant disorder (13%) Overall, internalizing symptoms were more prominent than externalizing symptoms No differences in psychological symptoms emerged as a function of age, sex, ambulation status, or lesion level 13% of families fell in the clinically problematic range of family functioning, and 23% of families reported difficulties in the establishment and maintenance of family roles and responsibilities Child psychological functioning was significantly and positively related to problematic family functioning; however, child age, sex, ambulation status, and level of lesion were not	Strengths include use of measures with demonstrated reliability and attention to clinical significance of psychiatric symptoms (i.e., whether child met criteria for diagnosis) Limitations include reliance on a primarily white sample; questionnaires completed via mail; reliance on a single-method (questionnaire), single-reporter (mother) data collection strategy; exclusion of fathers; and use of a cross-sectional research design
Ammerman et al ²⁶	Parent-Child Problem Solving Interactions in Families of Visually Impaired Youth	1991	<i>Journal of Pediatric Psychology</i>	Sample consisted of 24 visually impaired (VI) adolescents, 25 adolescents with SB, and 25 AB adolescents, and their parents Measures included observational assessment of parent-adolescent dyadic problem solving	No differences as a function of group status were documented in patterns of problem solving in either mother-adolescent or father-adolescent dyads	Strengths include use of a comparison sample of AB adolescents, as well as a comparison sample of adolescents with another disability; inclusion of mothers and fathers; and utilization of observational assessments of family interaction Limitations include cross-sectional research design and reliance on single-method (observational) data collection strategy

Barakat and Linney ³⁹	Children with Physical Handicaps and their Mothers: The Interrelation of Social Support, Maternal Adjustment, and Child Adjustment	1992	<i>Journal of Pediatric Psychology</i>	<p>Sample consisted of 29 families of children with SB (between the ages 6 and 11 yr) and a comparison sample of 28 families of AB children</p> <p>Measures included the PPVT-R as an index of receptive vocabulary; parent-report measures of child adaptive behavior, child behavioral functioning, parental adjustment, social support, demographic information, and child illness severity; child self-report of self-assessment of mother-child and father-child play</p> <p>Data were collected through interviews with parents and children during a hospital appointment</p>	<p>Children in the SB group evidenced significantly lower self-concept scores and lower adaptive behavior ratings than did children in the AB group</p> <p>No significant differences in child internalizing or externalizing symptoms were observed between the SB and AB groups</p> <p>No significant differences in maternal social support and maternal psychological adjustment were observed between groups</p> <p>Social support accounted for significant variance in the prediction of maternal adjustment in the SB sample, with increasing levels of social support indicative of better adjustment</p> <p>Maternal social support significantly and negatively predicted child externalizing problems within the SB sample</p> <p>Maternal adjustment did not significantly predict child adjustment within the SB sample</p> <p>No significant differences in the nature or strength of the relationship between (1) social support and maternal adjustment, (2) social support and child adjustment, or (3) maternal adjustment and child adjustment were documented as a function of group status (SB vs AB), suggesting that the process of maternal stress adaptation, and the relationship between maternal adjustment and child outcomes is similar across groups</p>	<p>Strengths include use of a multivariate, ecological prediction model and inclusion of a comparison sample</p> <p>Limitations include exclusion of fathers; significant differences between the SB and comparison sample on a number of demographic factors (e.g., SES, and parent levels of education, ethnicity); and cross-sectional research design</p>
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Appendix A (continued)

Authors	Title	Year	Publication	Method	Results	Comments
Barakat and Linney ³²	Optimism, Appraisals, and Coping in the Adjustment of Mothers and Their Children with Spina Bifida	1995	<i>Journal of Child and Family Studies</i>	Sample consisted of 29 families of a child with SB and a demographically matched comparison sample of 28 families with an AB child; all children ranged in age from 6-11 yr Measures included PPVT-R as an index of receptive vocabulary; parent report of coping, child adaptive behavior, demographic information, child illness severity, and child behavior problems; child report of perceived self-competence; and observational indices of parent-child play Data were collected during a visit to a children's hospital or a university clinic	Mothers of children with SB appraised stressful child situations as having less of an impact on child self-esteem than did mothers of AB children Mothers of children with SB evidenced lower levels of optimism in comparison to mothers of AB children Maternal avoidant coping was positively associated with threats to self-esteem and negatively associated with (1) threats to a loved one and (2) maternal optimism Maternal coping strategies, particularly avoidant coping, were significantly predictive of maternal adjustment, such that adaptive forms of coping were positively associated with adjustment, whereas maladaptive forms of coping were negatively associated with adjustment Maternal problem-focused coping was positively associated with child internalizing symptoms	Strengths include use of a comparison sample of AB children Limitations include exclusion of fathers, unsuccessful matching of samples on SES and race, and use of a primarily white sample
Bier and Liebling ⁴⁴	Parents' and Pediatricians Views of Individuals with Meningomyelocele	1996	<i>Clinical Pediatrics</i>	Sample consisted of 63 parents (48 mothers and 15 fathers) of individuals with SB (age range, 0.5-27 yr; mean age, 9 yr) and 66 pediatricians of individuals with SB Measures included parent report of the impact of SB on family functioning, physician perceptions of the impact of SB on family functioning, and medical chart review for medical status and psychosocial functioning variables	Higher lesion level was associated with parental report of a more negative impact of SB on family functioning and social functioning No differences in maternal and paternal perceptions were documented Pediatricians predicted a more negative impact of illness on family functioning than was perceived by parents	Strengths include inclusion of mothers and fathers and use of a multisource data collection strategy (mothers, fathers, and pediatricians) Limitations include lack of comparison sample, cross-sectional research design, wide age range of children used, use of a primarily middle-class sample, failure to report information on family ethnicity

Bower and Hayes ⁶⁷	Mothering in Families With and Without a Child With Disability	1998	<i>International Journal of Disability, Development, and Education</i>	<p>Sample consisted of 17 mothers with a child with Down syndrome, 14 mothers of a child with SB, and 38 mothers of an AB child</p> <p>Measures included maternal report of coping and family functioning</p>	<p>No significant differences in maternal coping were documented between mothers of children with SB and mothers of children with Down syndrome or between mothers of children with SB and mothers of AB children</p> <p>No significant differences in maternal perceptions about family hardship were documented between the 3 groups</p>	<p>Strengths include use of a comparison sample of AB children, as well as a comparison sample of children with cognitive disabilities</p> <p>Limitations include exclusion of fathers, failure to match samples on potentially relevant demographic characteristics, reliance exclusively on maternal report of individual and family functioning, use of an unsophisticated data analytic strategy, and cross-sectional research design</p>
Cappelli et al ³¹	Marital Quality of Parents of Children with Spina Bifida: A Case-Comparison Study	1994	<i>Developmental and Behavioral Pediatrics</i>	<p>Sample consisted of 46 families of a child with SB and a matched comparison sample of 46 families of an AB child; children ranged in age from 1-15 yr</p> <p>Measures included parent report of marital satisfaction, parenting stress, role strain, attitudes toward traditional marital roles, depressive symptoms, and observational assessment of couples' communication styles</p> <p>Data were collected via a visit to families' homes</p>	<p>No differences in marital quality were documented between groups, even after controlling for differences in SES levels; moreover, within each dyad, mothers and fathers did not significantly differ in their perceptions of marital satisfaction</p> <p>Across groups, increasing parenting stress was associated with decreased marital satisfaction; marital quality, in turn, was negatively associated with depression and role strain</p> <p>No differences in parental psychological functioning were documented between groups; moreover, with each dyad, mothers and fathers did not significantly differ in their psychosocial functioning</p> <p>Within the SB group, mothers of (1) female and (2) children reported higher marital quality than mothers of (1) male and (2) older (i.e., >5 yr) children did</p> <p>Within the SB group, ambulation status was significantly associated with maternal</p>	<p>Strengths include inclusion of both mothers and fathers; use of multimethod data collection strategy (questionnaires and behavioral observation)</p> <p>Limitations include unsuccessful matching on several demographic characteristics (SES and parent education), cross-sectional research design, and inclusion of only intact families</p>

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Authors	Title	Year	Publication	Method	Results	Comments
Coakley et al. ²⁶	A Longitudinal Study of Pubertal Timing, Parent-Child Conflict, and Cohesion, in Families of Young Adolescents with Spina Bifida	2002	<i>Journal of Pediatric Psychology</i>	Sample consisted of a demographically matched group of 68 families with children with SB and 68 families with AB children. Children were aged 8-9 yr at Time 1 and 10-11 yr at Time 2. Measures included maternal report of pubertal timing; and parental, child, and observer report of family conflict and cohesion.	Prospective longitudinal analyses documented an association between early pubertal maturation and (1) higher levels of conflict and (2) lower levels of cohesion within the AB group. Families of children with SB tended to display either no response to pubertal timing or increases in positive functioning (e.g., cohesiveness) over time regardless of pubertal timing. Early maturing males evidenced greater increases in conflict than males maturing on time.	Strengths include the use of a prospective longitudinal design, use of a multisource and multimethod data collection strategy, and inclusion of fathers. Limitations include a focus on early stages of pubertal development, fewer measures of cohesion relative to conflict, and use of a predominantly white sample.
Edwards-Beckett ²⁶	Parental Expectations and Child's Self-concept in Spina Bifida	1995	<i>Children's Health Care</i>	Sample consisted of 30 families with a child with SB and a demographically matched sample of 30 families with an AB child; all children were between the ages 6 and 12 yr (mean age, 9.5 yr). Measures included parent report of child future development and child self-report of self-concept.	Parents of children with SB differed significantly in their future expectations of the adaptive abilities of their children in comparison to parents of AB children; specifically, parents of children with SB expected their children to learn more adaptive skills within the next 5 yr, thus displaying "catch up" growth to their AB counterparts. No significant differences were documented with respect to the self-concept of children with SB compared with the AB children. No significant relationship between parental expectations and child self-concept emerged in the present investigation for either the SB or AB group.	Strengths include use of demographically matched comparison sample and use of standardized measures with adequate reliability. Limitations include reliance primarily on mother report (although 4 of 60 participating parents were fathers), use of a predominantly middle-class white sample, exclusion of children with educational delays, and use of a cross-sectional research design.

Fagan and Schor ⁴⁰	Mothers of Children with Spina Bifida: Factors Related to Maternal Psychosocial Functioning	1993 <i>American Journal of Orthopsychiatry</i>	Sample consisted of 50 mothers of children with SB (mean age, 8.1 yr) Measures included maternal report of demographic information, social support, general family functioning, and maternal psychosocial functioning Data were collected during a visit to the SB clinic of a large urban hospital	Adult companionship and social support significantly predicted maternal satisfaction in the parenting role Neither adult companionship nor marital status predicted maternal well-being or maternal competence African-American and Latino mothers evidenced lower levels of maternal satisfaction compared with white mothers Family functioning significantly predicted maternal perceived parenting competence and maternal well-being	Strengths include use of an ethnically diverse sample (i.e., 62% white, 20% Latino, and 18% African American) Limitations include reliance on single-reporter (mother), single-method (questionnaire) data collection strategy
Havermans and Eisler ⁶⁸	Mothers' Perceptions of Parenting a Child with Spina Bifida	1991 <i>Child: Care, Health, and Development</i>	Sample consisted of mothers of 19 children with SB (mean age, 8.8 yr) Measures included the use of a semistructured interview to obtain information about child's condition, family use of medical and psychological services, school accommodations, and type of school setting; questionnaire measures were used to assess maternal self-confidence in illness management, illness-related child role restrictions, maternal social support, and family difficulties in daily living Data were collected via maternal interview and maternal completion of questionnaires	Mothers of children with high severity ratings reported higher rates of family difficulties in daily living and greater perceived restrictions on child behavior relative to mothers of children with low severity ratings Mothers of children with high severity ratings reported greater confidence in managing their child's illness relative to mothers of children with low severity ratings A positive association was documented between maternal confidence and level of social support	Strengths include a comprehensive assessment of disease severity based on several factors including type of SD, shunt status, status of physical problems associated with SD (e.g., bowel or bladder incontinence), self-help skills, and ambulation status Limitations include use of single reporter (mother) on variables of interest, use of small sample (n = 19), use of unstandardized measures created for the study, no information reported about reliability of measures, no comparison group used, and unsophisticated statistical analyses
Holmbeck et al ¹¹	Observed and Perceived Dyadic and Systemic Functioning in Families of Preadolescents with Spina Bifida	2002 <i>Journal of Pediatric Psychology</i>	Sample consisted of a demographically matched group of 68 families with 8-9 yr olds with SB and 68 families with 8- to 9-yr-old AB children Measures included parent and	Relative to families of AB children, families of children with SB were less cohesive Children in the SB group were more passive during family interactions, relative to their AB counterparts	Strengths include multi-informant, multimethod data collection strategy, use of matched comparison sample, inclusion of mothers and fathers, and assessment of dyadic and systemic functioning

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Authors	Title	Year	Publication	Method	Results	Comments
Holmbeck and Faler-Routman ⁴⁵	Spinal Lesion Level, Shunt Status, Family Relationships, and Psychosocial Adjustment in Children and Adolescents with Spina Bifida Myelomeningocele	1995	<i>Journal of Pediatric Psychology</i>	child report of parent-child conflict and decision making; parent report of family conflict, cohesion, and family life events; and observational ratings of family interaction	Verbal IQ mediated the relationship between group status and family functioning, such that children with SB displayed lower IQs, and lower IQ was associated with lower family cohesion Lower SES status was associated with increased mother-child conflict, less family cohesion, and more stressful life events	Limitations include cross sectional research design, reduced generalizability of findings due to narrow age range used, and exclusion of non-English-speaking families
Holmbeck et al ⁶	Maternal, Paternal, and Marital Functioning in Families of Preadolescents with Spina Bifida	1997	<i>Journal of Pediatric Psychology</i>	Sample consisted of 65 children and adolescents with SB myelomeningocele (age range, 8-16 yr) Measures included maternal report of demographic factors, child behavioral autonomy, family conflict, parent-child attachment, and child grades, as well as child and mother reports of child behavioral functioning and self-competence A medical chart review was conducted to obtain information about IQ and lesion level Data were collected during a scheduled appointment at the SD clinic within an urban children's hospital	Mothers of children with higher lesion levels (e.g., thoracic) reported higher levels of attachment to their children relative to mothers of children with lower lesion levels Mothers of children with higher lesion levels reported less family conflict relative to mothers of children with lower lesion levels Mothers of children with higher lesion levels reported a greater willingness to grant the child behavioral autonomy relative to mothers of children with lower lesion levels Relative to mothers of AB children, mothers in the SB group reported lower levels of parenting satisfaction, lower levels of perceived parental competence, higher levels of social isolation, and less adaptability to change and used less adaptive coping	Strengths include the use of multiple reporters (child, mother) of outcome of lesion interest, verification of lesion level using medical chart, and use of standardized questionnaires with demonstrated reliability Limitations include the fact that significant findings emerged only from maternal report data and not from child report, and parents and children were allowed to complete questionnaires at home and return them via mail, creating the potential for poor child comprehension of questionnaires
Holmbeck et al ⁶	Maternal, Paternal, and Marital Functioning in Families of Preadolescents with Spina Bifida	1997	<i>Journal of Pediatric Psychology</i>	Sample consisted of a demographically matched group of 55 families with an 8- to 9-yr-old children with SB and 55 families with 8- to 9-yr-old AB children	Relative to mothers of AB children, mothers in the SB group reported lower levels of parenting satisfaction, lower levels of perceived parental competence, higher levels of social isolation, and less adaptability to change and used less adaptive coping	Strengths include a focus on both maternal and paternal functioning and the use of a matched comparison sample Limitations include the use of cross-sectional data and the reliance on self-report data

<p>Measures included self-report questionnaires assessing parental coping, adaptability to change, psychological symptoms, parenting satisfaction, perceived parenting competence, and marital satisfaction Data were collected during a 3-hr visit to the families' homes</p>	<p>strategies (e.g., more denial and less active coping and planning) Relative to fathers of AB children, fathers in the SB group reported higher levels of psychological symptoms, lower levels of parenting satisfaction, and lower levels of role restriction and reported the use coping through venting emotions more frequently Despite presence of group differences, 75% of parents were not functioning in the dysfunctional or symptomatic range No differences between the SB and AB groups were documented in levels of marital satisfaction Coping predictors of adjustment varied as a function of parent gender rather than illness status</p>	<p>Strengths include multi-informant, multimethod data collection strategy; use of matched comparison sample; and inclusion of mothers and fathers Limitations include use of cross-sectional research design, reduced generalizability of findings due to narrow age range used and exclusion of non-English-speaking families, and significance of findings may be partially explained by common method variance</p>
<p>Sample consisted of a demographically matched group of 68 families with 8-9 yr olds with SB and 68 families with 8- to 9-yr-old AB children Measures included child, parent, and observational reports of parental overprotectiveness; parent and child reports of behavioral autonomy; and reports of child internalizing and externalizing symptoms and perceived self-competence Data were collected during a 3-hr visit to the families' homes</p>	<p>Mothers and fathers in the SB group evidenced higher levels of overprotection relative to parents in the AB groups on both questionnaire and observational indices; however, this relationship was partially mediated by child cognitive ability Across both the SB and AB samples, mothers displayed higher levels of overprotection than fathers did, with higher levels of overprotection associated with lower levels of behavioral autonomy Within the SB sample, support was found for the mediational role of behavioral autonomy in explaining the relationship between questionnaire reports of overprotection and child externalizing problems</p>	<p>Observed and Perceived Parental Overprotection in Relation to Psychosocial Adjustment in Preadolescents with a Physical Disability: The Mediational Role of Behavioral Autonomy</p>

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Authors	Title	Year	Publication	Method	Results	Comments
Holmbeck et al. ²⁰	Observed and Perceived Parenting Behaviors and Preadolescents with Spina Bifida	2002	B.K. Barber (ed), <i>Intrusive Parenting: How Psychological Control Affects Children and Adolescents</i>	Sample consisted of a demographically matched group of 68 families with 8-9 yr olds with SB and 68 families with 8- to 9-yr-old AB children Measures included PPVT-R as an index of receptive language; parent, child, and observational reports of parenting behaviors; parent and teacher report of child behavior problems; teacher report of academic performance; and observational report of child adaptive behavior Data were collected during a 3-hr visit to the families' homes	Mothers of children with SB exhibited higher levels of psychological control relative to mothers of AB children (across both questionnaire and observational reports) Parental psychological control was associated with a variety of negative psychosocial outcomes across both the SB and AB groups Parental acceptance was associated with positive psychosocial outcomes across the SB and AB groups; however, acceptance was less strongly related to outcomes in comparison to psychological control Parental behavioral control was rarely related to psychosocial adjustment among children in either the SB or AB group	Strengths include multi-informant, multimethod data collection strategy, use of matched comparison sample, and inclusion of mothers and fathers Limitations include cross-sectional research design, reduced generalizability of findings due to narrow age range used, and exclusion of non-English-speaking families
Horton and Wallander ²⁸	Hope and Social Support as Resilience Factors Against Psychological Distress of Mothers Who Care for Children with Chronic Physical Conditions	2001	<i>Rehabilitation Psychology</i>	Sample consisted of 111 mothers of 5- to 18-yr-old children with cerebral palsy, SD (n = 33), or diabetes Measures included parent-reported feelings of hope, social support, psychological functioning, and disability-related stress	No differences in maternal distress were found as a function of illness type Hope and social support were negatively associated with maternal distress The relationship between disability-related stress and maternal maladjustment was moderated by maternal perceptions of hope, such that, with high levels of stress, hope served a buffering effect	Strengths include use of a large sample Limitations include exclusion of fathers; use of a cross-sectional research design; and reliance on single-reporter (mother), single method (questionnaire) data collection strategy
Kazak and Clark ¹²	Stress in Families of Children with Myelomeningocele	1986	<i>Developmental Medicine and Child Neurology</i>	Sample consisted of 56 families with a child with SB and an age-matched comparison sample of 53 families with an AB child	Children with SB evidenced significantly lower self-concepts than AB children No significant differences in sibling self-concept were	Strengths include use of a multimethod, multi-informant data collection strategy and use of a comparison sample Limitations include group

differences in parent age, level of education, and annual family income	documented between the groups	Measures included an interview to collect relevant demographic data; a medical chart review to collect data on illness severity; parent-report of social network, child independence of bowel and bladder functioning, marital satisfaction, parenting stress, and family functioning; and child report of perceived self-concept	Maternal and paternal perceived marital satisfaction was positively associated with child illness severity	There was a trend for child illness severity to be positively associated with maternal parenting stress	Mothers and fathers of more severely impaired children reported significantly more stress in the areas of child distractibility and activity compared with parents of less severely impaired children. Regardless of severity level, maternal and paternal ratings of stress evidenced clinical elevations
Strengths include use of a sophisticated data analytic strategy; inclusion of a large sample, and inclusion of mothers and fathers	Greater levels of family-centered caregiving was predictive of parent well-being	Sample consisted of 164 parents of children (age range, 3-6 yr) with pediatric chronic illnesses/physical disabilities including cerebral palsy, SD (n = 18), and hydrocephalus	Salient predictors of child well-being included child behavior problems and protective factors within the social environment	Reports of demographic factors, disability parameters, caregiving process, family functioning, satisfaction with social support, psychosocial stressors, coping, satisfaction with care, and emotional functioning	Limitations include use of a heterogeneous sample with respect to illness type (i.e., cerebral palsy, SD, and hydrocephalus); use of a primarily white, urban, middle-class sample; cross-sectional research design; and reliance on single-method (questionnaire) data collection strategy
Parents completed questionnaires via mail	Parents completed questionnaires via mail	Parents completed questionnaires via mail	Parents completed questionnaires via mail	Parents completed questionnaires via mail	Parents completed questionnaires via mail
Strengths include attention to impact of cognitive appraisals as influencing adjustment,	Mothers of children with SB endorsed factors related to their child's medical illness as	Sample consisted of 66 mothers of a child with SB (age range, 2 mo to 18 yr)	Mothers of children with SB endorsed factors related to their child's medical illness as	Mothers of children with SB endorsed factors related to their child's medical illness as	Mothers of children with SB endorsed factors related to their child's medical illness as

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Authors	Title	Year	Publication	Method	Results	Comments
	Children with Myelomeningocele			Measures included questionnaires assessing maternal stress and psychological symptoms, and medical chart data on SB severity	<p>being sources of the greatest stress, with social and other stresses rated as less stressful</p> <p>Mothers evidenced elevated symptoms of depression, anxiety, and global psychological distress, relative to normative comparisons</p> <p>No significant correlations were documented between medical indexes (e.g., lesion level, presence of hydrocephalus, shunt status, CNS infections, etc.) and maternal psychological symptoms</p> <p>Significant positive associations were documented between levels of appraised stress and maternal symptoms of depression, anxiety, and global distress</p> <p>In regression analyses, demographic factors (particularly race) and appraised medical stress accounted for nearly one third of the variance in maternal global distress</p>	<p>testing of a multivariate statistical model, and inclusion of African-American and Indian mothers</p> <p>Limitations include exclusion of fathers, use of cross-sectional research design, reliance on self-report data, and use of single-method (questionnaire) data collection strategy</p>
Kronenberger and Thompson ³⁴	Psychological Adaptation of Mothers of Children with Spina Bifida: Association with Dimensions of Social Relationships	1992	<i>Journal of Pediatric Psychology</i>	Sample consisted of 66 mothers of children with SB (age range, 2 mo to 18 yr) Measures included maternal report of demographic information, family environment, marital adjustment, social support, social coping, and psychological symptoms, as well as SB severity information obtained from medical chart review	<p>Significant positive associations were documented between marital coping and measures of social coping and marital support</p> <p>Increasing levels of family support and decreasing levels of control and conflict were associated with adaptive maternal psychological functioning (lower levels of somatization, depression, anxiety, and global distress)</p>	<p>Strengths include the use of standardized measures, inclusion of African-American and Indian mothers, and evaluation of a multivariate prediction model</p> <p>Limitations include exclusion of fathers, use of cross-sectional research design, reliance on self-report data, and use of single-method (questionnaire) indices of family, marital, and individual psychosocial functioning</p>

Lemanek et al. ⁸⁷	Mothers of Children with Spina Bifida: Adaptational and Stress Processing	2000	<i>Children's Health Care</i>	<p>Data were collected during a routine SB clinic appointment</p> <p>Marital functioning was moderately related to maternal symptoms of anxiety, depression, and global distress</p> <p>Higher levels of emotional regulation coping using friends was associated with greater psychological symptoms</p> <p>In multivariate models, demographic variables (i.e., race and child gender) predicted global distress ratings, as did marital functioning and levels of family control; in contrast, medical indices of severity were unrelated to maternal adjustment, as were social coping variables</p>	<p>Strengths include attention to the differential impact of SB on maternal and paternal functioning and comparison of data with normative values</p> <p>Limitations include reliance primarily on single-reporter (maternal report), single-method (questionnaire) data collection strategy; use of a predominantly middle-class, white sample; and use of a cross-sectional research design</p>
				<p>Maternal perceived efficacy ratings and parenting competence ratings were significantly higher than those for the normative group</p> <p>No differences in maternal ratings of psychological distress or parenting satisfaction were found in comparison to normative ratings</p> <p>Maternal ratings of psychological distress were significantly lower than paternal ratings of psychological distress; however, both maternal and paternal ratings were within the reference range of functioning when compared with normative values; no significant differences were found between maternal and paternal ratings of parenting competence, efficacy, or satisfaction</p>	
				<p>Sample consisted of 59 mothers of children with SB and 19 fathers of children with SB; children ranged in age from 3-16 yr</p> <p>Measures included parent-reported demographic information, parental psychological distress, parenting satisfaction, and child social skills; disease severity information was obtained through medical chart review</p> <p>Parents completed questionnaires either during a regularly scheduled child clinic appointment or at home following the appointment</p>	

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Appendix A (continued) Authors	Title	Year	Publication	Method	Results	Comments
Loebig ⁷¹	Mothers' Assessments of the Impact of Children with Spina Bifida on the Family	1990	<i>Maternal-Child Nursing Journal</i>	Sample consisted of 10 mothers of children with SB, ranging in age between 5 and 11 yrs of age Measures included maternal report of child health history, demographic information, and impact of SB on family Data were collected through audiotaped interviews	Maternal ratings of child social skills were significantly lower than the mean social skill levels of the normative sample, but still within the average range of functioning No significant differences were documented in child behavior problems when comparing children with SB to the normative sample Maternal reports of child social skills were negatively associated with child problem behavior and positively associated with parental satisfaction and medical severity Child behavior problems were negatively associated with parenting satisfaction and positively associated with parental distress and child age; moreover, more problem behaviors were reported among boys than girls	Strengths include comparison of functioning of mothers of children with SB to reference group of mothers of children with other chronic conditions Limitations include use of middle-class white sample and small sample size (n = 10), reliance on maternal report only, and use of an exploratory, descriptive, cross-sectional approach
					No significant differences in the impact of SB on family functioning when comparing families of children with SB to a reference group of families of children with other chronic conditions Positive associations were documented between (1) financial impact of illness and no. of children in family, (2) work impact of illness and no. of children in family, (3) social impact of illness and child special education status, (4) social impact of SD and single parent status, (5) coping and family income, and (6) work impact of illness and family income	

<p>Negative associations were documented between (1) work impact of SD and child special education status and (2) work impact of SD and single-parent status</p>	<p>Perceived family encouragement of achievement was positively associated with employment status, community mobility, and social activity</p> <p>Perceived family encouragement of independence was positively associated with community mobility</p> <p>Perceived family organization was positively associated with social activity</p> <p>Perceived moral/religious emphasis of family was negatively associated with employment</p> <p>Perceived family involvement with intellectual and cultural activities was negatively associated with mobility and social activity</p>	<p>Sample consisted of 32 adults (18-48 yr old) with SB</p> <p>Measures included medical chart report of medical status, assessment of intellectual functioning using WAIS-R, self-report of family environment, and a structured-interview to assess current life situation (e.g., educational history, vocational history, living arrangements, etc.).</p>	<p><i>Developmental Medicine and Child Neurology</i></p>	<p>1997</p>	<p>Relations between Family Environment and Adjustment Outcomes in Young Adults with Spina Bifida</p>	<p>Loomis et al.⁷²</p>
<p>Strengths include a focus on an adult sample, use of questionnaire and interview methods, and inclusion of covariates in multiple regression analyses</p> <p>Limitations include use of retrospective reports of family functioning, wide age range used in the study (18-48 yr), failure to report information on ethnic background of participants, small sample size (n = 32), cross-sectional design, and no comparison sample</p>	<p>Maternal age was significantly and positively associated with perceived parenting stress</p> <p>Maternal education, SES, child medical severity, and family social support were not significantly associated with maternal perceived parenting stress</p> <p>Caregivers with the highest levels of dysfunctional parent-child interactions were less likely to use available support services relative to caregivers with lower levels of parent-child dysfunction</p>	<p>Sample consisted of 56 caretakers of a child with SB (54 mothers, 1 father, and 1 grandparent); children ranged from 1-17 yr old (mean age, 6.27 yr)</p> <p>Measures included caregiver report of parenting stress, perceived family support, and demographic information</p> <p>Data were collected during a routine SB clinic appointment</p>	<p><i>Children's Health Care</i></p>	<p>2001</p>	<p>Predictors of Parenting Stress in Families of Children with Spina Bifida</p>	<p>Macias et al.⁴¹</p>

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Authors	Title	Year	Publication	Method	Results	Comments
McCormick et al ⁷³	Assessing the Impact of Spina Bifida on the Family	1986	<i>Developmental Medicine and Child Neurology</i>	Sample consisted of 201 families with a child with SB (age range, infancy to 18 yr) Measures included a telephone survey administered to parents assessing demographic characteristics of the family, acute and chronic health problems of the child, health care use for the affected child, limitations in child daily living and self-care activities, parental perceptions of child's health, health care-related expenses and services for the child, and maternal perception of the impact on the family of child health problems Data were collected via a telephone survey of families involved in the SB clinic of a major children's hospital	Significant predictors of family stress in rearing a child with SB included (1) indicators of child health status (no. of daily activities limited by child's health and parental perceptions of child health), (2) resources required to deal with child's health including maternal educational attainment, no. of adults in the home, insurance status, and family income, (3) frequency of doctor visits, and (4) employment status of father Parental perceptions of limitations in the child's daily living and self-care activities were the strongest predictor of the impact of SB-associated problems on family functioning, followed by parental perceptions of the child's health	Strengths include use of a large sample size Limitations include reliance on a predominantly white sample, reliance primarily on maternal report of child and family functioning, reliance on a telephone survey data collection strategy, and use of a cross-sectional research design
Samuelson et al ⁷⁴	Stress and Coping in Families of Children with Myelomeningocele	1993	<i>Archives of Psychiatric Nursing</i>	Sample consisted of 17 mothers and 17 fathers of children with SB (age range, preschool to 8th grade) Measures included parent report of family health-related stressors, family hassles, and family coping Data were collected via visits to the families' homes	Significant differences emerged regarding areas in which mothers and fathers desired assistance; help related to play was the most common need reported by mothers, whereas help with school activities was the most common need reported by fathers Overall, mothers evidenced more concerns than fathers; however, the child's future and sufficient income were salient concerns shared by both parents Perceptions of too many things to do were a hassle frequently mentioned by both parents	Strengths include use of a sample of mothers and fathers and comparison of differences in functioning between mothers and fathers Limitations include small sample size, unsophisticated data analytic strategy, use of a primarily middle-class and all-white sample of families, and use of a cross-sectional research design

Family Functioning in Children with Spina Bifida

<p>Seefeldt et al.⁷⁵</p>	<p>Socioeconomic Status and Democratic Parenting in Families of Preadolescents with Spina Bifida</p>	<p>1997</p>	<p><i>Psi Chi Journal of Under-graduate Research</i></p>	<p>Sample consisted of 55 families with a child with SB and a demographically matched sample of 55 families with an AB child; all children were between ages 8 and 9 yr. Measures included observational indices of parenting behaviors. Data were collected during a 3-hr home visit.</p>	<p>Relative to fathers, mothers evidenced higher mean scores on measures of coping. Religious coping through faith in God was the coping method most commonly used by mothers and fathers.</p>	<p>Parents in the high SES groups engaged in more democratic forms of parenting relative to parents in the low SES groups. Parents of children with SB scored higher on indices of authoritarian parental control, relative to parents of AB children. Significant social class × group interactions were documented, such that parents in the SB group with low SES evidenced the lowest levels of democratic parenting relative to (1) parents in the SB group with high SES, (2) parents in the AB group with low SES, and (3) parents in the AB group with high SES.</p>	<p>Strengths include use of a demographically matched comparison sample, use of observational ratings of parenting behaviors with documented moderate to high levels of inter-rater reliability, inclusion of mothers and fathers, and investigation of moderational models. Limitations include cross-sectional research design, use of predominantly white sample, and reliance solely on observed rather than perceived parenting behaviors.</p>
<p>Spaulding and Morgan¹⁶</p>	<p>Spina Bifida Children and Their Parents: A Population Prone to Family Dysfunction?</p>	<p>1986</p>	<p><i>Journal of Pediatric Psychology</i></p>	<p>Sample consisted of 19 families of a child with SB (age range, 5–15 yr) and a matched sample of 19 families of AB children. Measures included parent report of demographic information, parenting attitudes, marital satisfaction, perceptions of child behavior, perceived family functioning, perceived stress, and impact of SB on family functioning; and child report of behavior and self-concept. Data were collected during a visit to each family's home.</p>	<p>No differences in family functioning as a function of illness status were reported. No differences in child adjustment or self-concept as a function of illness status were reported. Neither fathers nor mothers reported that the presence of a child with SB significantly disrupted family functioning. No differences emerged with respect to maternal or paternal perceptions of (1) the impact of the child with SB on the family, (2) parenting attitudes, (3) marital satisfaction, (4) perceived stress, or (5) perceived family functioning both within the SB sample and in comparison to parents in the AB sample.</p>	<p>Strengths include use of a matched comparison sample of AB children, use of a multirespondent (parent, child) data collection strategy, and inclusion of mothers and fathers. Limitations include cross-sectional research design, small sample size, reliance on single method (questionnaire) data collection strategy, exclusion of SB children with cognitive impairments, and use of a primarily white sample of families.</p>	

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Authors	Title	Year	Publication	Method	Results	Comments
Wiegner and Donders ²⁷	Predictors of Parental Distress After Congenital Disabilities	2000	<i>Journal of Developmental and Behavioral Pediatrics</i>	Sample consisted of 60 parents of children with cerebral palsy, 34 parents of children with SB, and 27 parents of children with limb deficiencies. (age range, 3–12 yr); illness groups did not differ on demographic variables of interest Measures included parent-report of demographic variables, family stress, child adaptive behavior, family functioning, and parent psychological functioning Data were primarily collected via phone interview with caregivers (primarily mothers)	On average, children in the SB group showed fewer impairments in adaptive functioning than children with cerebral palsy did, but more impairments in adaptive functioning than children with limb deficiencies did On a measure of parent psychological functioning, 41% of parents in the SB group evidenced clinically significant levels of distress, which was significantly lower than parents of children with cerebral palsy (48%) and not significantly different from parents of children with limb deficiencies (37%) Role restrictions in family functioning and the presence of a significant psychosocial stressor within the past 6 mo were significantly associated with parental distress 12% of parents in the SB group reported significant family dysfunction (compared with 35% of parents in the cerebral palsy group and 15% of parents in the limb deficiencies group), with the degree to which families are able to assign and manage family roles/tasks being the most problematic area of family functioning across all groups	Strengths include use of standardized assessment instruments, comparison of parent and family functioning among different illness groups Limitations include reliance on single-method (questionnaire), single-source (parent) data collection strategy; collection of data via telephone interview; and reliance primarily on maternal report (90% of respondents were mothers)
Williams et al ⁴³	Maternal Mood, Family Functioning, and Perceptions of Social Support, Self-esteem, and Mood Among Siblings of Chronically Ill Children	1999	<i>Children's Health Care</i>	Sample consisted of 22 healthy siblings (age range, 8–15 yr) and parents of chronically ill children with cystic fibrosis, diabetes, or SB Measures include assessments of maternal mood, family	Maternal mood was positively associated with sibling perceptions of higher social support, which in turn was related to higher self-esteem and improved mood Maternal mood was positively	Strengths include attention to the impact of chronic illness on adjustment of siblings Limitations include use of a heterogeneous sample with respect to type of pediatric condition (i.e., cystic fibrosis,

<p>Williams et al.²</p>	<p>Interrelationships Among Variables Affecting Well Siblings and Mothers in Families of Children with a Chronic Illness or Disability</p>	<p>2002</p>	<p><i>Journal of Behavioral Medicine</i></p>	<p>cohesion, sibling perceptions of social support, self-esteem, and mood</p>	<p>associated with family functioning, which in turn was positively associated with sibling mood</p>	<p>diabetes, and SB) and use of a cross-sectional research design</p>
<p>Strengths include attention to the impact of chronic illness on adjustment of siblings, use of sophisticated data analytic technique (structural equation modeling), and use of a large sample of families</p>	<p>SES predicted maternal mood, which in turn influenced family cohesion</p>	<p>Sample consisted of 252 dyads of healthy siblings (mean age, 11 yr) and parents of a chronically ill or disabled child, some of whom had SB (others had cystic fibrosis, cancer, diabetes, or developmental disability)</p>	<p>SES had the strongest impact on sibling behavior, followed by family cohesion and self-concept; other significant predictors of sibling behavior included child age, perceived sibling support, and knowledge of illness</p>	<p>Siblings with cancer, cystic fibrosis, diabetes, SB, autism, seizure disorder, or cerebral palsy), reliance on a primarily middle-class, white sample; and use of cross-sectional research design</p>	<p>Limitations include use of a heterogeneous sample of families affected by pediatric chronic illness or physical disability (i.e., families of children with cancer, cystic fibrosis, diabetes, SB, autism, seizure disorder, or cerebral palsy), reliance on a primarily middle-class, white sample; and use of cross-sectional research design</p>	<p>SES predicted maternal mood, which in turn influenced family cohesion</p>
<p>SES had the strongest impact on sibling behavior, followed by family cohesion and self-concept; other significant predictors of sibling behavior included child age, perceived sibling support, and knowledge of illness</p>	<p>Sibling mood was most strongly affected by one's attitude toward the illness</p>	<p>Measures included parent-report of demographic variables, family cohesion, parent mood, healthy sibling behavior problems, and child-report of self-concept, social support, knowledge of illness, and attitude toward illness</p>	<p>Sibling self-esteem was most strongly predicted by sibling mood and perceived social support; other significant predictors of self-esteem included family cohesion</p>	<p>Sibling-perceived social support was significantly predicted by sibling mood, family cohesion, and age</p>	<p>Sibling attitude about the illness was significantly predicted by family cohesion, self-esteem, and illness-related knowledge</p>	<p>Siblings with cancer, cystic fibrosis, diabetes, SB, autism, seizure disorder, or cerebral palsy), reliance on a primarily middle-class, white sample; and use of cross-sectional research design</p>

SB indicates spina bifida; ADHD, attention-deficit/hyperactivity disorder; AB, able-bodied; CNS, central nervous system; PPVT-R, Peabody Picture Vocabulary Test-Revised; WAIS-R, Wechsler Adult Intelligence Scale-Revised.