

Spina Bifida Guideline

Family functioning guidelines for the care of people with spina bifida

Tessa K. Kritikos* and Grayson N. Holmbeck
Loyola University Chicago, Chicago, IL, USA

Abstract. Research supports a resilience-disruption model of family functioning in families with a child with spina bifida. Guidelines are warranted to both minimize disruption to the family system and maximize family resilience and adaptation to multiple spina bifida-related and normative stressors. This article discusses the spina bifida family functioning guidelines from the 2018 Spina Bifida Association's Fourth Edition of the Guidelines for the Care of People with Spina Bifida, and reviews evidence-based directions with the intention of helping individuals with spina bifida achieve optimal mental health throughout their lifespan. Guidelines address clinical questions pertaining to the impact of having a child with spina bifida on family functioning, resilience and vulnerability factors, parenting behaviors that may facilitate adaptive child outcomes, and appropriate interventions or approaches to promote family functioning. Gaps in the research and future directions are discussed.

Keywords: Spina bifida, myelomeningocele, family functioning, neural tube defects

1. Introduction

Research supports a resilience-disruption model of family functioning in families with a child with spina bifida [1,2], wherein families display both disruption in some aspects of family functioning as well as resilience, when compared with families of healthy children. Disruption is considered to be present when there are high levels of behaviors such as family conflict or parental psychological control. Resilience in the family system is represented either by more adaptive behaviors, such as parental acceptance, or by patterns of family functioning that are similar to those of typically developing youth [2]. Overall, there are relatively low rates of family-level dysfunction (10–15%) in families with a child with spina bifida [3,4]. However, family dysfunction has been found to be associated with important variables related to spina bifida care: for example, family conflict is associated with diminished medical adherence [5]. Additionally, certain families appear to be

at greater risk for disruption, such as those with children who have more significant cognitive impairment; such families tend to experience lower family cohesion [6].

Aspects of family functioning that are particularly salient for families of an individual with spina bifida include the parent marital relationship, sibling adjustment, parental stress and competency, and promotion of autonomous development. In terms of marital functioning, families of children with spina bifida show few differences compared to families of typically-developing children [3,4,7]. Interestingly, there is some evidence that having a child with a disability may even strengthen a marriage [8]. The quality of the marital relationship prior to the birth of a child with spina bifida is an important predictor of parental adjustment. It appears that high quality partner relationships may positively affect the extent to which spina bifida affects parents [9].

Regarding sibling adjustment, findings suggest both positive and negative effects of having a sibling with spina bifida [10]. Positive family attitudes toward spina bifida, overall family satisfaction, and the degree of sibling conflicts are important predictors of sibling adjustment [11,12]. In support of the resilience of families of children with spina bifida, some siblings reported that

*Corresponding author: Tessa K. Kritikos, Loyola University Chicago, 1000 W Sheridan Rd, Chicago, IL 60626, USA. Tel.: +1 610 764 0711; E-mail: tessakritikos@gmail.com.

the shared spina bifida experience created family bonds that seemed stronger than those of other families [10].

Overall, parental stress in families who are raising children with spina bifida is higher than in the general population, particularly among mothers, single parents, older parents, and/or economically disadvantaged and culturally-diverse parents. This is an example of cumulative risk, which is an important construct relevant to both family functioning and spina bifida care [6,7]. The complexity of the child's condition and parental personality traits (e.g., extraversion, agreeableness, emotional stability) have also been found to be significant predictors of parental stress [13].

It is common for parents of children with spina bifida to feel less satisfied and competent as parents, to have a lower quality of life, to have smaller social networks, to be less optimistic about the future, and to feel more isolated [4,14]. Spina bifida has a significant impact on parental adjustment and outcomes such as sleep duration, especially for mothers [9,15]. Parents may also experience Post Traumatic Stress Disorder (PTSD) and depression [4,9,16]. The alterations in parental functioning that occur as a result of having a child spina bifida impact the psychosocial adjustment of children. Greater parenting stress, poorer parent psychosocial adjustment, and low marital satisfaction are associated with negative child adjustment outcomes, especially externalizing symptoms [17].

Importantly, the relationship between psychosocial and family functioning may vary according to socio-cultural variables. For example, research has found that Latino youth with spina bifida exhibit less family conflict than non-Latino Caucasian youth with spina bifida. Furthermore, amongst Latino youth, family conflict was associated with psychosocial functioning, whereas family cohesion, conflict, and stress were associated with psychosocial functioning in non-Latino Caucasian youth [18].

Parents of youth with spina bifida exhibit higher levels of parental intrusiveness and overprotectiveness, than the parents of typically developing children without spina bifida. These behaviors are often linked with less functional child outcomes such as lower levels of independent decision-making. However, these effects are often modified by the child's cognitive level (e.g., children with lower IQs have parents who are more controlling) [19].

Adolescence is a challenging time as parents and adolescents negotiate the gradual transfer of medical management from parent to child [7]. In many ways, it can seem as though the demands of caring for an

adolescent with a chronic health condition are at odds with the normative adolescent goals of increased autonomy. This leaves parents feeling reluctant to transfer decision-making responsibility to their child, and can frequently lead to parent-child conflict [20]. However, when compared to their typically developing peers, youth with spina bifida do not experience normative increases in family conflict as a function of pubertal development [21,22]. Parents of youth with spina bifida tend to be less responsive to pubertal development than is the case in families of typically-developing children [21].

Family can often be central to and particularly influential in the lives of youth with spina bifida, as they are frequently more socially isolated from their peers than their typically developing counterparts [23]. Indeed, positive family relationships have the capacity to buffer youth with spina bifida from some of the negative psychosocial outcomes that they may otherwise be at risk of developing. For example, research has found that satisfaction with family functioning may protect youth with spina bifida from depressive symptoms [24]. Furthermore, qualitative research on quality of life amongst adolescents and young adults with spina bifida found that youth most frequently endorsed family as contributing to their quality of life [25].

Given the centrality and importance of the family system in caring for an individual with spina bifida, adequate attention and ample support should be given to the family members of people with spina bifida. Few family intervention studies specific to spina bifida have been conducted to better understand this important topic. It is recommended that family-based interventions target "modifiable" aspects of family functioning, including problem-solving, facilitation of shared responsibility for medical care, parenting stress, intrusive parenting, and coping [26,27].

2. Guidelines goals and outcomes

The goals of the family functioning guidelines were both practical and aspirational. Below are the primary, secondary, and tertiary outcomes for the family functioning guidelines.

Primary

1. Maximize family resilience and adaptation to multiple spina bifida-related and normative stressors as appropriate for developmental level.

Secondary

1. Maximize parental adaptation, expectations, and responsiveness to the changing developmental

Table 1
Clinical questions that informed the family functioning guidelines

Age group	Clinical questions
0 months – 18+ years	<ol style="list-style-type: none"> 1. What is the impact of having a child with spina bifida on family functioning (including parental adjustment, marital functioning, effect on the siblings and extended family, and familial participation in social activities) and how does this impact change as children move through various stages of lifespan development? 2. What resilience and vulnerability factors are predictive of familial adaptation at each level of child development? 3. What parenting behaviors facilitate adaptive child outcomes (including independence-related outcomes such as self-management and the transfer of health care responsibilities from parent to child) and how do these adaptive parenting behaviors vary developmentally? 4. What interventions or approaches can promote family functioning?

level of the child by identifying and reinforcing effective parenting techniques.

2. Maximize independence of the child within the family context, given developmental level and condition-related constraints.

Tertiary

1. Minimize parental and marital stress and maladaptation when raising a child with a serious chronic health condition.
2. Maximize family engagement in social activities, including parental self-care activities.
3. Maximize parental knowledge of spina bifida and advocacy.

3. Methods

As part of an initiative within the Spina Bifida Collaborative Care Network, literature review and consensus-building methods were combined to develop and expand the fourth edition of the Guidelines for the Care of People with Spina Bifida (“guidelines”). Details regarding the full scope and methodology of the development of all of the Guidelines for the Care of People with Spina Bifida can be found in the methodology paper published by Dicianno and colleagues [28]. These guidelines were published via the Spina Bifida Association website [29].

A family functioning working group was formed consisting of a team of clinical and research experts. This group devised a list of “clinical questions,” the answers to which provide guidance on how best to care for people with spina bifida and achieve the primary, secondary, and tertiary outcomes agreed upon and defined above.

Evidence based-research and consensus methodologies were used to develop these guidelines. Specifically, a systematic review of multiple databases was conducted and the consensus building methodology, One-Text Procedure, was followed to draft and review docu-

ments. This working group presented their guidelines at a face-to-face meeting using the Nominal Group Technique (NGT). The goal of these guidelines was to not only guide health care providers but also patients and families, so that people with spina bifida can enhance their quality of life throughout their lifespan.

4. Clinical questions framing the guidelines

Table 1 presents the clinical questions that informed the family functioning guidelines.

5. Results

Table 2 presents the family functioning guidelines developed by the family functioning working group. These guidelines are organized by age group. For each age group, the guidelines aim to address pertinent clinical questions relating to the impact of having a child with spina bifida on family functioning and how this impact changes across development, resilience and vulnerability factors, parenting behaviors that may facilitate adaptive child outcomes, and appropriate interventions or approaches to promote family functioning.

6. Discussion

A resilience-disruption model of family functioning recognizes the ways in which having a child with spina bifida can both cause disruption in the family system but also fortify relationships and promote adaptive, resilient behaviors. Risk and protective factors differentially contribute to relative levels of disruption and resilience. By bolstering protective factors and mitigating risk factors at each developmental level, it is possible to maximize family adaptation to spina bifida-related and normative stressors.

The Family Functioning Guidelines for the Care of People with Spina Bifida strive to provide evidence-based directives with the following desired goals for in-

Table 2
Family functioning guidelines

Age group	Guidelines	Evidence
0–11 months	<ol style="list-style-type: none"> 1. Refer families who have received a prenatal diagnosis of spina bifida for prenatal counseling and consultation with members of a spina bifida multidisciplinary clinical team. Assess family dynamics and adjustment in response to diagnosis. 2. Assess for postpartum depression. Provide information about spina bifida, parenting, treatments, support groups, and the Spina Bifida Association. 3. Coordinate services during the transition from the hospital stay to subsequent clinic follow-up, stressing the need for ongoing multi-specialty care. 4. Teach necessary home care procedures such as post-surgical care, skin care, and clean intermittent catheterization, as needed. 5. Assess family dynamics and adjustment (e.g., post-traumatic stress disorder in parents) during infancy. 6. Refer the parents or caregivers to infant intervention and appropriate state programs (e.g., Supplemental Security Income) and financial resources as needed. Provide financial counseling if necessary. 7. Provide support and ongoing counseling as needed to parents, the child, and siblings. 8. Provide anticipatory guidance for parents regarding strengths and possible cognitive and behavioral challenges in children with spina bifida and their siblings. 9. Teach parents to advocate for themselves and their child when working with medical, educational, and agency staff. 10. Be aware that although interventions should target all families, some families are particularly at-risk for adjustment and adherence difficulties (e.g., those from lower socioeconomic backgrounds and single-parent families). 11. Assess the family's ability to carry out medical regimens, and identify possible barriers to adherence, such as need for caregiver support and parental beliefs regarding alternative therapies. 	See mental health guidelines Clinical consensus as well as [4,5,9,11,12,16,26]
1–2 years, 11 months	<ol style="list-style-type: none"> 1. Provide support and ongoing counseling as needed to parents, the child, and siblings. 2. Be aware that although interventions should target all families, some families are particularly at-risk for adjustment and adherence difficulties (e.g., those from lower socioeconomic backgrounds and single-parent families). 3. Promote effective parenting techniques or provide referral for such services. 4. Provide anticipatory guidance for parents regarding possible behavioral challenges and autonomy needs in children with spina bifida and their siblings. 5. Assess family's need for additional counseling, financial resources, or other support services. 6. Inform families of advocacy resources and encourage them to contact the appropriate governmental and non-governmental authorities to obtain additional information, referrals, and support. 7. Encourage the parents or other primary caregivers to teach other family members or close friends how to provide for the child's specialized care needs and how to access other needed services. Alternatively, families can arrange for child care by trained professionals. 8. Educate parents about the importance of engaging in personal activities that promote parental well-being. 9. Refer the parents to early intervention services, if these are not already in place. 10. Assess the family's ability to carry out medical regimens, and identify possible barriers to adherence, such as need for caregiver support and parental beliefs regarding alternative therapies 	See mental health and neuropsychology guidelines Clinical consensus as well as [4,5,12,16,19,26]
3–5 years, 11 months	<ol style="list-style-type: none"> 1. Provide support and ongoing counseling as needed to parents, the child, and siblings. 2. Provide anticipatory guidance for parents regarding possible behavioral challenges and autonomy needs in children with spina bifida and their siblings. 3. Teach parents to advocate for themselves and their child when working with medical, educational, and agency staff. 4. Be aware that although interventions should target all families, some families are particularly at-risk for adjustment and adherence difficulties (e.g., those from lower socioeconomic backgrounds and single-parent families). 5. Re-assess parenting skills such as discipline, behavior management, and sibling relationships. 6. In the context of family functioning, address self-care abilities and refer to therapies (OT, PT). 7. Discuss issues that affect children with spina bifida when they transition to school. 8. Advise parents of their child's right to free and appropriate education in the least restrictive environment through the public schools (i.e., explain services available under the Individuals with Disabilities Education Act). 9. Assess the family context for helping the child to develop self-management skills and to carry out medical regimens and identify possible barriers to adherence. 	See mental health, neuropsychology guidelines, self-management and independence guidelines, and transition guidelines Clinical consensus, as well as [5,12,16,19,26]

Table 2, continued

Age group	Guidelines	Evidence
6–12 years, 11 months	<ol style="list-style-type: none"> 1. Provide support and ongoing counseling for parents, the child, and siblings, as needed. 2. Provide anticipatory guidance for parents regarding possible behavioral challenges and autonomy needs in children with spina bifida and their siblings. 3. Be aware that although interventions should target all families, some families are particularly at-risk for adjustment and adherence difficulties (e.g., those from lower socioeconomic backgrounds and single-parent families). 4. Assess family dynamics and relationships with school staff. 5. Have detailed discussions about appropriate interventions to address academic and social difficulties. Provide parents with current and accurate information about various school settings. For each type of setting, identify potential gaps and determine the impact that such a setting has on family members and the family system. 6. Encourage advocacy activities and resources and motivate parents to advocate for themselves and their children with medical, educational, and agency staff. 7. Advise parents of their child's right to free and appropriate education in the least restrictive environment through the public schools (i.e., explain services available under the Individuals with Disabilities Education Act and Section 504 of Vocational Rehabilitation Act of 1973). 8. Serve as a resource to school systems regarding health issues, individualized educational planning, and socialization. 9. Reinforce appropriate family leisure activities. 10. Reinforce effective parental discipline, behavioral management, and expectations. 11. Encourage the family to facilitate medical self-management in their children with spina bifida, as developmentally appropriate. 12. Facilitate parents' understanding of the importance of fostering their child's independence and participating in chores and other activities of daily living. 13. Encourage social activities such as sleepovers, camp overnights, dating, and social and recreational activities outside the home. Encourage development and maintenance of friendships. 14. Emphasize positive attitudes, assertiveness, and self-empowerment of family members. 15. Encourage the family to develop strategies that gradually empower their children toward independence such as decision-making and problem-solving. 16. Assess the family context for helping the child to develop self-management skills and to carry out medical regimens and identify possible barriers to adherence. 	See mental health, neuropsychology, self-management and independence, and transition guidelines Clinical consensus, as well as [5,12,16,19,26,30–32]
13–17 years, 11 months	<ol style="list-style-type: none"> 1. Provide support and ongoing counseling for parents, child, and siblings, as needed. 2. Be aware that although interventions should target all families, some families are particularly at-risk for adjustment and adherence difficulties (e.g., those from lower socioeconomic backgrounds and single-parent families). 3. Assess parent-child communication and their relationship. Aid parents to encourage the development of autonomy in their child with spina bifida. 4. Encourage the family to begin planning for their child's transition to adult health care. 5. Begin discussions of other important developmental milestones, including educational and vocational achievement, living independently, and community participation. 6. Give advice to the child and family about the right to free and appropriate education in the least restrictive environment through the public schools (i.e., explain services available under the Individuals with Disabilities Education Act and Section 504 of Vocational Rehabilitation Act of 1973). 7. Assist with normative sexual education, as well as specific issues relevant to the teen's condition. Work with the teen to navigate sexual expression in a safe and mature fashion. 8. Continue to encourage the family to facilitate medical self-management in their child with spina bifida. 9. Assess the family context for helping the child to develop self-management skills and to carry out medical regimens and identify possible barriers to adherence. 	See self-management and independence, transition, sexual health and education guidelines Clinical consensus, as well as [5,12,16,19,26,30,31]
18+ years	<ol style="list-style-type: none"> 1. Provide support and ongoing counseling for parents, young adults, and siblings, as well as older adults with spina bifida, as needed. 2. Work with families to support the development of maximal vocational and social independence. 3. Continue to work with the family to support medical self-management in their young adult. 4. Continue working with the family to ensure a successful transition to adult health care. 5. Work with the young and older adults to navigate sexual expression in a safe and mature fashion. 6. Assess the family context for helping the young adult to develop self-management skills and to carry out medical regimens and identify possible barriers to adherence. 	See transition, sexual health and education, self-management and independence guidelines Clinical consensus, as well as [5,12,16,30,31]

dividuals with spina bifida. Primarily, these guidelines aspire to maximize family resilience and adaptation to multiple spina bifida-related and normative stressors as appropriate for developmental level. Secondly, these guidelines strive to maximize parental adaptation, expectations, and responsiveness to the changing developmental level of the child by identifying and reinforcing effective parenting techniques, as well as maximizing independence of the child within the family context, given developmental level and condition-related constraints. Finally, these guidelines had three intended tertiary outcomes: to minimize parental and marital stress and maladaptation raising a child with spina bifida, to maximize family engagement in social activities, including parental self-care activities, and to maximize parental knowledge of spina bifida and advocacy.

Limitations in the research underscore important areas for continued investigation and development of resources. Future intervention research should develop and empirically evaluate interventions that a) maximize familial resilience and adaptation at each level of the child's development, b) facilitate adaptive parenting behaviors, c) enhance familial, marital, and parental adjustment outcomes, and d) support families as they transfer medical management from parent to child and transition from pediatric to adult healthcare. Additional research is also needed to clarify how the cognitive profile of children and young adults with spina bifida may complicate the unfolding of self-management within the family context.

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The Spina Bifida Association has already embarked on a systematic process for reviewing and updating the guidelines. Future guidelines updates will be made available as they are completed.

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Conflict of interest

The authors have no conflict of interest to report.

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