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Adolescent development and family functioning in youth with spina bifida

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Abstract. The purpose of this article is to review research concerning adolescent development and family functioning among youth with spina bifida myelomeningocele (SBM). Adolescence is a developmental period characterized by substantial changes in biological, psychological, and social functioning, as well as transformation and reorganization within the family system. A biopsychosocial-contextual model of development was utilized to describe the interface between normative adolescent development and the experience of a chronic health condition among youth with SBM. Major empirical findings relevant to family functioning in adolescents with SBM are presented, including the family environment, parenting behaviors, and marital and parental functioning. There is variability with regards to the influence of SBM on the family system and research identifies both disruption and resilience in families. Current research suggests that families of youth with SBM may have higher levels of family stress, difficulties with family roles, lower levels of cohesion, less adaptive parental control and overprotection, and a greater risk for child and parental psychosocial adjustment difficulties. The review concludes with a discussion of the clinical implications of these findings for the care of youth with SBM and directions for future research.

Keywords: Spina bifida myelomeningocele, adolescence, family functioning

1. Adolescents with chronic health conditions: An introduction

Adolescence is a transitional period characterized by substantial biological, psychological, and social changes in development [17]. Given the magnitude of such changes, it is not surprising that adolescence is a critical period in an individual's development of health-related behaviors [65]. Throughout this developmental period, individuals will establish and consolidate life-long patterns of positive health behaviors (e.g., exercise, diet), health risk behaviors (e.g., substance use, risky sexual behaviors), and health-related self-advocacy. Moreover, the transition from childhood to adulthood is remarkable for considerable continuities and discontinuities in the development of health problems and psychological disorders [55]. Individuals sharing characteristics at a specific starting point are likely to later experience diverse outcomes depending on a variety of individual and environmental outcomes (referred to as multifinality [17]). This is particularly true among individuals with chronic illnesses and/or disabilities where variability is even more pronounced [28].

Adolescents with chronic health conditions, such as spina bifida myelomeningocele (SBM), not only face the developmental changes of normative adolescence, but they also confront unique challenges owing to their health condition [65]. These illness-specific challenges occur within a larger environmental context that also undergoes transformation over time. For example, adolescents with chronic health conditions must learn to successfully negotiate changes within the health-care system (e.g., transition from pediatric-oriented care to adult-oriented care), the family environment (e.g., transition from parent-controlled health care to self-care), the social environment (e.g., participation in romantic

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relationships), and the school environment (e.g. transition into high school). As youth with SBM navigate these developmental milestones, family relationships have been regarded as a particularly important source of support and predict a variety of important psychological and physical outcomes [23,26,66].

SBM is a complex congenital birth defect affecting nearly 18 out of every 100,000 live births [47]. It originates during the early stages of gestation when one or more vertebrae fail to close normally and the surrounding bone and muscle cannot form around the spinal cord. Associated health complications include neurogenic bladder and bowel dysfunction, weakness and paralysis of the lower extremities, hydrocephalus, endocrine dysfunction, neurocognitive challenges, and seizure disorders. The severity of SBM will vary depending on the location and size of the spinal lesion and the presence of neurological complications, such as Chiari II malformation. Despite the complexity of SBM, advances in healthcare and technology have increased the life expectancy of youth with SBM and at least 75% of children are expected to reach adult years [9]. Accordingly, patients with SBM and families are confronted with new health care concerns and must learn to negotiate the developmental tasks of adolescence and adulthood.

A diagnosis of SBM necessitates a significant commitment by families to maintain the neurologic and social functionality of their child, such as assistance with illness-management, activities of daily living (e.g., eating dressing, bathing), and instrumental activities of daily living (i.e., activities required for independent living, such as meal preparation and balancing money). Moreover, families are often confronted with significant financial stress as they manage their child's illness. Ouyang and colleagues [51] estimated an average medical expenditure cost 13 times greater among youth with SBM, as compared to medically healthy youth. These additional stressors (e.g., psychological, financial) place caregivers at risk for individual psychological distress and family maladjustment [29,41,46,61]. Nevertheless, researchers have also noted that many families of children with SBM demonstrate significant resiliency, if not more positive outcomes, as compared to families of youth without SBM [26,44,58]. Therefore, researchers and medical health providers have focused their efforts both on isolating predictors that may increase vulnerability to child and family psychosocial difficulties and on identifying mechanisms that may buffer, exacerbate, or mediate the impact of a chronic pediatric health condition on adjustment outcomes [e.g. [21,44]).

In addition to the health-related stressors of caring for a child with SBM, there are several additional reasons why examining family functioning in youth with SBM is critical. *First*, children with SBM are confronted with social difficulties that may increase isolation from peers and increase reliance on families for social relationships and support. Youth with SBM tend to be more socially immature and are more likely to have difficulties initiating and establishing peer relationships as compared to their medically healthy peers [36]. Moreover, the physical manifestations of SBM (e.g., unusual gait, use of braces or other forms of assistive ambulation devices) frequently increase stigmatization and rejection by peers [54,57]. Adolescents with physical disabilities acknowledge that these social (e.g., child viewed as different by peers) and physical obstacles (e.g., school playgrounds or public places) often disrupt their ability to develop peer relationships and participate in extracurricular activities [57]. As a result, youth with SBM may rely on parents to help them engage in peer group activities. *Second*, youth with SBM, particularly those with hydrocephalus, frequently experience difficulties with functional independence. For example, many youth with SBM and hydrocephalus report significant issues with sphincter control, locomotion, and self care, and they require support with transfers (e.g., for a wheelchair), social cognition, and communication [60]. Functional independence may be further limited by the commonly experienced neurological sequelae of SBM (e.g., difficulties with attention, abstract reasoning, and non-verbal task [11,14,20]). On average, youth with SBM acquire autonomy skills 2 to 5 years after their same-aged peers [19]. Thus, many youth with SBM benefit from increased planning and logistics by family members to promote autonomy and carry out tasks of daily living. *Finally*, youth with SBM are at greater risk for psychosocial adjustment difficulties (e.g., internalizing symptoms), as compared to medically healthy peers [1,3,43]. These adjustment difficulties will likely influence parental and familial stress levels.

As researchers continue to draw attention to family relationships as a particularly influential source of support among youth with SBM [7,66], research studies have been employed to better understand the impact of SBM on family functioning. Therefore, the purpose of this article is to review literature on the adjustment of families of youth with SBM as youth are confronted with the developmental changes of the adolescent years. First, to capture the tremendous variability of changes that occur during adolescence, a

biopsychosocial-contextual model of normative adolescent development will be presented that emphasizes biological, psychological, and social changes of adolescence. Primary developmental changes and milestones of adolescence will be delineated and a discussion concerning the interplay between developmental issues of adolescence and the experience of SBM will be provided. Second, a review of the major empirical findings relevant to family functioning in adolescents with SBM will be presented. Finally, we will discuss the clinical implications of these findings for the care of youth with SBM and directions for future research.

2. Biopsychosocial-contextual model of adolescent development

A contextual framework for understanding adolescent development and adjustment was proposed by Holmbeck and Shapera [34] and emphasizes biological, psychological, and social changes of adolescence (see Fig. 1). The presented model conceptualizes the relationship between primary developmental changes of adolescence (e.g., biological/pubertal, psychological/cognitive, social roles) and developmental outcomes (e.g., achievement, autonomy, and identity) as occurring via the interpersonal contexts in which adolescents develop (e.g., family, peer, and school). In other words, developmental changes impact on relationships and environmental factors which, in turn, influence the individual's ability to master critical milestones of adolescence. For example, associations between primary developmental changes and developmental outcomes may be mediated by the degree of conflict and/or cohesion within the family environment. This contextual model integrates information regarding normative adolescent development. A modified version of this model is easily applied to adolescents with SBM with the primary changes of normative adolescent development also being influenced by features of SBM. Three areas of change will be described below: biological/ pubertal, social roles, and psychological/ cognitive changes.

2.1. *Biological/pubertal changes*

As mentioned earlier, substantial physical growth and change take place throughout adolescence with considerable variability across individuals [17]. In particular, there is substantial intraindividual variability regarding the time of onset, duration, and termination of the pubertal cycle [10]. Medically-healthy youth that experience an early onset of pubertal development are at increased risk for a variety of adaptational difficulties, such as depression, substance use, early sexual behavior, eating problems and disorders, and family conflicts. Interestingly, youth with SBM are more likely to experience a precocious period, particularly females with hydrocephalus [18,59]. Secondary sex characteristics of youth with SBM may begin to emerge as early as 7 years of age. In addition to the psychosocial risk factors confronted by typically developing adolescents, youth with SBM may also experience a worsening of pre-existing adaptational difficulties (e.g., low self worth, depression). For instance, weight gain associated with puberty may worsen the adolescent's pre-existing body dissatisfaction and insecurities about her physical appearance. In general, disruptions in normal pubertal timing set the stage for potential asynchronies between adolescents' physical and social development. Children with SBM may appear more mature than their medically healthy peers; yet in actuality most of these youth are less socially mature and more dependent on adults [36]. Early physical maturation in children with SBM may also incorrectly imply advanced cognitive sophistication. As a consequence, premature transfer of responsibility for medical self-care tasks from parent to adolescent may take place, thus increasing the likelihood for medical problems and adherence difficulties. In general, an early or precocious pubertal development necessitates earlier discussions between parent and child about associated physical and emotional changes, sexuality, and self-care tasks; however, these discussions must be tailored to the child's cognitive capacities and level of maturity.

2.2. *Changes in social roles*

A variety of changes in social status occurs during the adolescent developmental period. Social redefinition from childhood to adolescence is a universal experience, yet the specific changes that take place are culturally-specific. In Western industrialized societies, changes in social roles tend to occur across four domains: interpersonal relationships (e.g., changes in family status), political (e.g., eligibility to vote), economic (e.g., adolescents begin establishing employment), and legal (e.g., late adolescents can be tried in adult court systems). The experience of SBM may disrupt the nature and timing of such role changes. Holmbeck and colleagues [36] found that youth with SBM tend to be less likely to make independent decisions

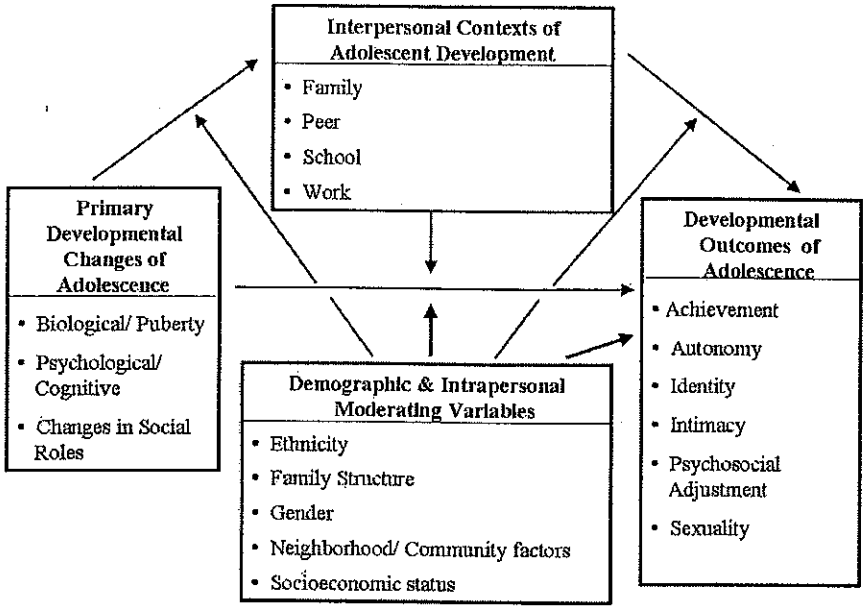

Fig. 1. A biopsychosocial model of adolescent development and adjustment (Reproduced with permission from G.N. Holmbeck, and W. Shapera, Research methods with adolescents, in: *Handbook of Research Methods in Clinical Psychology*, (2nd ed.), P.C. Kendall, J.N. Butler and G.N. Holmbeck, eds, J Wiley & Sons, New York, 1999, pp. 634–661.)

and are more dependent on adults, as compared to their medically healthy peers. Furthermore, physical and cognitive disabilities may limit the adolescent's ability to experience the benefits of these newly gained rights. For example, obtaining a driver's license is a highly desired privilege among adolescents, but may be unattainable for some adolescents with motor and visual disabilities, and consequently, further increase feelings of social isolation and place limits on their independence.

2.3. *Psychological/cognitive changes*

Adolescence has long been described as a critical period of cognitive development, with particular growth of the capacity for complex and abstract reasoning, and increases in processing capacity, knowledge base, cognitive self-regulation, and socially-relevant cognitions (e.g., empathy [24]). Similar to the other arenas of change, advances in cognitive and psychological development may also be affected by the presence of SBM. Cognitive limitations frequently associated with SBM may limit adolescents' growth towards autonomy in medical self-care and normative activities of living. For instance, youth with SBM are at risk for executive functioning and memory impairments (i.e., prospective, immediate and delayed episodic memory [20]), attentional difficulties [14,36], and below average non-verbal skills [11]. Several factors place youth with SBM at particular risk for cognitive impairment, including the presence of Chiari II malformation [63], multiple shunt revisions [12,20,37], and a history of seizures [12]. Furthermore, cognitive ability (e.g., memory) in youth with SBM is associated with functional independence [20]; thus significantly impaired youth often require greater assistance from family members to achieve autonomy and perform activities of daily living. Diminished cognitive ability may also disrupt the adolescents' ability to form positive relationships with same-aged peers, thereby increasing social isolation and psychological maladjustment. Internalizing problems (e.g., depression, anxiety) are particularly common concerns among youth with SBM [1,3].

3. Overview of literature on family functioning in youth with spina bifida

Throughout adolescence, changes in family relationships take place as the child-rearing practices of parents are altered in response to a child's developmental level [52]. Ideally, parenting behaviors are gradually altered to promote increased autonomy in youth [33]. Parents of adolescents are confronted with the important task of facilitating responsibility and autonomy in their adolescent, as they also maintain a high level of cohesiveness within the family structure. Par-

enting an adolescent with a chronic health condition or physical disability may be a particularly complex task because the demands of caring for their child are often at odds with the normative adolescent goals of increased responsibilities and autonomy. Consequently, parents may be hesitant to relinquish appropriate decision-making responsibilities to their child with a disability, particularly in regards to medical issues [2].

Although some researchers argue that peers have a greater influence on social development than parents, the family context among most adolescents with SBM is particularly influential due to decreased peer interactions [36,37,57]. As previously discussed, the number of obstacles these adolescents face make social interactions with peers complicated and simultaneously increases the importance of family social relationships. Researchers have drawn attention to several areas of family functioning in youth with SBM, including the family environment, parenting behaviors, and marital and parental functioning [23].

3.1. Family environment

Research findings regarding the impact of caring for a child with a chronic health condition on family stress are mixed [23,61]. Some researchers suggest that families of youth with SBM experience considerable disruption within the family system [1,64]. For example, Ammerman and colleagues [1] found that 13% of families of youth with SBM fell within the clinically problematic range of family functioning (i.e., problem solving, communication, affective involvement, affective responsiveness, roles, behavioral control), as indicated by parental reports, and 23% of families reported difficulties establishing and maintaining family roles and responsibilities. Similarly, another study found that 12% of parents of youth with SBM report significant family dysfunction (e.g., difficulty maintaining family roles [64]); however this percentage was lower than rates in families of youth with other congenital disabilities (i.e., 35% for the cerebral palsy group and 15% for the limb deficiency group). On the other hand, several researchers have found no significant differences between families of youth with SBM and medically healthy youth in reports of stressful life events [26] or disruptions in family functioning [58].

McCormick and colleagues [48] identified several variables that are likely to influence the amount of stress experienced within families of youth with SBM: the child's health status (e.g., limitations to daily functioning), resources for dealing with a child with a chronic health condition (e.g., maternal education, medical insurance, family income), number of doctors' visits, and employment status of the father. Severity of the child's health problems has also been associated with increased family difficulties in daily living [25], such as bladder problems [13] and a higher lesion level [8]. The presence of hydrocephalus is another risk factor due to impaired cognitive abilities [6] and functional independence [60]. Moreover, lower SES, ethnic minority status, and single-parent status have also emerged as salient predictors of higher levels of parental stress within families of children with a chronic health condition [16]. SBM has been identified across socioeconomic status (SES) levels, yet there are moderately higher prevalence rates among families of lower SES [39,50]. Thus, in addition to tackling the stressors of caring for a child with a chronic health condition, some families are confronted with the additional stressors that accompany lower SES (e.g., substantial medical costs). In brief, the presence of SBM alone does not cause family dysfunction; rather the degree of family support (e.g., family resources, social support) and nature of the condition (e.g., severity of health problems) influences the risk for disruption within the family system.

The degree of conflict and cohesion within the parent-child dyad has been an area of interest for developmental researchers [23,61]. Serious parent-child conflict is not common [33], but there is typically increased conflictive engagement and emotional withdraw during adolescence, most notably at the peak of pubertal development [31]. Thus, a moderate level of conflict is normative within healthy families and, in fact, some researchers have suggested that this conflict may facilitate positive familial adaptation to developmental changes [31]. Among families of preadolescents with SBM, Holmbeck and colleagues [26] found less family cohesion, as compared to families of medically healthy children. However, the child's cognitive functioning (i.e., verbal intelligence) mediated this relationship. In other words, youth with SBM tend to have lower levels of cognitive functioning, which, in turn, is associated with lower family cohesion. These researchers also failed to find significant group differences regarding the degree of family conflict. In sum, the systematic functioning among families of youth with SBM is characterized by some disruption (e.g., low levels of cohesion), as well as resiliency (e.g., no group differences for family conflict).

A follow up study conducted by Coakley and colleagues [18] suggests that the experience of SBM may

alter qualitatively the familial response to the transitional changes of puberty. Among medically healthy youth, there is an association between early pubertal maturation and higher levels of conflict and lower levels of cohesion. On the other hand, families of youth with SBM fail to display the pubertal effects evident among medically healthy youth. Rather, families of youth with SBM tend to display either no response to pubertal timing or increased levels of cohesion. Analyses also indicate that levels of cohesion increase over time in families of youth with SBM, yet cohesion levels tend to decrease among medically healthy youth [18]. Moreover, Holmbeck and Faier-Routman [27] suggest that the family environment is further qualified by the severity of the youth's illness. These researchers reported that mothers of children with higher lesion levels (e.g., thoracic), as compared to lower lesion levels, demonstrated less family conflict, more maternal attachment to their child, and greater willingness to grant their child behavioral autonomy. In conclusion, familial responses to developmental changes vary in interesting ways, depending on the context of development (spina bifida, medically healthy) and the severity of the child's illness.

3.2. *Parenting behaviors*

Parenting behaviors have been identified as another crucial area of research in families of youth with chronic health conditions, particularly in the areas of psychological control, behavioral control, acceptance, and parental overprotectiveness [23]. A cross-sectional study conducted by Holmbeck and associates [32] examined the association between parenting behaviors, namely observed and perceived parental overprotection, and psychosocial adjustment in preadolescents with SBM. Parental overprotection refers to an excessive amount of parental protection that surpasses the degree of protection necessary given a child's developmental level. In general, mothers and fathers of children with SBM demonstrate significantly higher levels of overprotection than parents of medically healthy children, as measured by questionnaires and observational assessments. However, this relationship was partially mediated by the child's cognitive abilities, such that children with SBM tend to have lower cognitive abilities that, in turn, were associated with greater levels of overprotection. In general, a higher level of parental overprotection was associated with lower levels of decision making autonomy and parental reluctance to grant autonomy in the future. Parental overprotection (measured by questionnaire only) was also associated with preadolescent externalizing problems, yet this relationship was mediated by the degree of behavioral autonomy. In other words, parental overprotection was associated with less behavioral autonomy, which, in turn, was associated with externalizing problems.

Anderson and Coyne [2] proposed the theory of *Miscarried Helping* to describe a process by which a child's chronic health condition may influence parenting behaviors. Frequently children with chronic health conditions, such as SBM, require additional assistance from caregivers to perform daily self-care and illness-related tasks. Although parents' helpfulness initially serves a practical function, over time their assistance may spill over into domains that the youth could feasibly manage on his/her own. Parents may then begin to exercise a large amount of psychological control, which impacts negatively on the child's level of independent functioning in these domains.

Holmbeck and colleagues [35] provided partial support for the theory of *Miscarried Helping* in their study that investigated the influence of observed and perceived parenting behaviors (i.e., psychological control, behavioral control, and acceptance) on adjustment outcomes among preadolescents. In general, mothers of children with SBM tend to be more psychologically controlling as compared to mothers of medically healthy youth. Notably, parental psychological control was associated with maladjustment for both groups, thus placing youth with SBM at risk for psychosocial difficulties. This study also found parental acceptance to be associated with positive adjustment outcomes, particularly among youth with SBM. No significant findings emerged regarding the influence of behavioral control on psychosocial adjustment. A longitudinal study following the same group of participants investigated the influence of parenting behaviors (responsiveness, demandingness) and the family environment (cohesiveness, conflict) on the development of coping behaviors among preadolescents with SBM [49]. Maternal and paternal responsiveness and family cohesiveness were significant predictors of preadolescent problem-focused coping strategies for both groups. Further analyses indicated that changes in parenting behaviors were concurrently associated with changes in coping strategies. The findings of these studies suggest that certain parenting behaviors are critical for the positive adjustment of youth with SBM and the development of positive coping strategies.

Greenley et al. [22] also found that the degree of family conflict and parenting stress were significant

predictors of parenting behaviors. In general, higher levels of familial conflict were significantly associated with less adaptive parenting behavior. This study failed to find support for the influence of marital functioning and parental psychological functioning on parenting behaviors. An interesting finding emerged in regards to familial conflict and parenting behaviors across time, such that higher levels of conflict during early childhood were associated with adaptive parenting changes during the adolescent developmental period. Thus, the degree of conflict within the family may facilitate the development of adaptive parental behaviors during adolescence. This finding supports developmental researchers' theory that a moderate level of conflict is normative and facilitates family adaptation to developmental changes [31].

3.3. Marital and parental functioning

Only a handful of studies have investigated the influence of raising a child with SBM on marital functioning [23]. Such studies found no differences between parents of SBM youth and medically healthy youth in marital satisfaction or marital quality [15,29,58] or partner support [53]. Vermaes and colleagues [61] conducted a review of literature investigating families of youth with SBM and did not find a significant negative impact of SBM on marital happiness, marital communication, or marital stability. Surprisingly, Kazak and Clark [39] found that severity of impairment among youth with SBM was positively associated with marital satisfaction, in that parents of the most severely impaired youth had the highest levels of marital satisfaction. Cappelli and associates [15] also found a positive association between severity of disability and marital functioning. These researchers found that parents with reported lower marital satisfaction had children with fewer ambulation problems. Collectively, these data suggest that caring for a child with a disability in certain contexts may strengthen the marital relationship.

Contradictory results have emerged regarding the psychosocial adjustment among parents of youth with SBM [61]. Several researchers suggest that parents of youth with SBM are more likely to experience psychological difficulties. A meta analysis conducted by Vermeas and colleagues [62] found that SBM in families predicted higher levels of psychological strain in parents. Nonetheless, their analyses revealed great variability in regards to the levels of psychopathology experienced, particularly among maternal caregivers, such that many parents do not experience psychopathology. Moreover, Kronenberger and Thompson [41] found that mothers of SBM youth were at increased risk for adjustment problems, including elevated levels of depression, anxiety, and global psychological distress. Although mothers frequently endorsed medical illness related factors as the greatest source of stress, there were no significant associations between medical indices (e.g., lesion level, ambulation) and maternal psychological symptoms. Holmbeck and colleagues [29] found that, compared to parents of medically healthy children, mothers of youth with SBM were at risk for lower levels of parenting satisfaction, lower levels of perceived parenting competence, and higher social isolation, and fathers were at risk for psychosocial difficulties (i.e., higher levels of psychological symptoms, lower parenting satisfaction). Nonetheless, it is noteworthy that within this sample 75% of parents of youth with SBM did not exhibit dysfunctional or symptomatic levels of psychosocial functioning. In other words, most parents are resilient and positively adjust to the increased parental demands of caring for a child with SBM. In fact, several studies have found no group differences (SBM versus medically healthy) in regards to parental adjustment, including maternal psychological adjustment [4] or psychological distress and parenting satisfaction [44].

A longitudinal study conducted by Friedman and colleagues [21] found that maternal and paternal psychosocial adjustment prospectively and concurrently predicted child adjustment outcomes, particularly in regards to child externalizing symptoms. Lower marital satisfaction, greater paternal stress in the parenting role, and greater maternal and paternal symptomatology (e.g., global psychosocial functioning) were prospectively associated with higher levels of child externalizing behaviors. Furthermore, greater maternal and paternal symptomatology were concurrently associated with higher levels of child internalizing behaviors. In general, associations between parent and child adjustment tended to be in the direction of parent to child. These data suggest that interventions aimed at helping parents successfully adjust to the increased demands of caring for a child with SBM may have significant implications for adolescent adjustment.

Several factors have been identified that increase the likelihood of positive psychosocial adjustment among parents of youth with SBM. These factors include increased social support [25,38,46], younger maternal age [45], and a positive marital relationship [42]. Other features of family life that may help protect parents from psychosocial maladjustment include greater lev-

els of parent-centered caregiving [40], lower levels of conflict within the family [42], lower levels of child behavior problems [12,46], and adaptive forms of coping [5,40,42]. Additional research has shown that increased levels of hope in parents, as well as the utilization of social support, are related to a higher quality of life for parents of adolescents with SBM [38,56].

4. Summary and clinical implications

Current research suggests that families are both disrupted by and resilient to the stress associated with raising a child with SBM [23,61]. Studies have identified a variety of family functioning domains impacted by the presence of SBM. Specifically, researchers have documented that families of children and adolescents with SBM may have higher levels of family stress, more difficulties with communication and family roles, lower levels of cohesion, more maladaptive forms of parental control and overprotection, and a greater risk for child and parental psychological maladjustment.

Although the presence of SBM represents a risk factor for psychosocial difficulties, considerable variability exists within children and adolescents and their families, such that many families will not experience negative outcomes. In fact, a variety of factors may be less negatively influenced or even enhanced as a consequence of the impact of SBM. Some families of adolescents with SBM may demonstrate more cohesion over time as compared to their medically-healthy peers. Furthermore, a healthy marital relationship prior to the birth of the child with SBM, effective child rearing techniques by parents, increased social support, higher socioeconomic status, and a higher verbal intelligence in the child may decrease the likelihood of negative outcomes.

Together, these data suggest several risk and protective factors that could be a focus of monitoring and intervention efforts in individuals with SBM and their families. To our knowledge, however, there are no published data that examine the effectiveness of interventions for families of children with SBM. Intervention efforts should target families that are most at risk for dysfunction and adjustment difficulties (e.g., single parent, low SES, ethnic minority families). Consideration of the context of development (e.g., family resources, social support) and nature of the child's condition (e.g., severity of health problems) can also help medical and mental health providers identify families that may benefit from additional support. Given the multitude of changes that individuals with SBM undergo during adolescence, it is critical that such programs consider the developmental needs of the adolescent. Interventions, therefore, should be flexible so as to account for the immense variability between families of youth with SBM.

Despite the lack of intervention studies targeting families of youth with SBM, several suggestions for intervention and prevention can be made based on the presented data. First, the individual needs of the child should be assessed. Given the body of research that suggests youth with SBM are at risk for internalizing problems (e.g., depression, anxiety), children and adolescents should be routinely screened for these difficulties. Youth with SBM that have cognitive and neuropsychological deficits (e.g., impaired memory, executive functioning, attention, and social cognition) may benefit from neuropsychological screening and school-based interventions (e.g., special education, tutors). Additionally, social difficulties are also found in youth with SBM. Greater emphasis needs to be directed towards increasing youth participation in extracurricular activities and contact with peers. Parents and health-care providers can maximize these children's opportunities for social involvement by helping them navigate the social and physical barriers of their environment (e.g., transportation, identifying accessible environments). Educating the child with SBM about their condition and providing tools to discuss their condition with peers may also reduce peer stigmatization. Furthermore, these youth will likely benefit from interventions that target not only medically-oriented difficulties (e.g., adherence), but also normative concerns of adolescent development (e.g., sexual and reproductive concerns, identity formation). Autonomy has emerged as a particularly salient concern among youth with SBM, as such families are at risk for increased parental overprotectiveness and parental control. These youth will likely benefit from interventions that focus on increasing autonomy within several domains, including emotional (i.e., interventions that decrease childhood dependency on adults), behavioral (i.e., those that increase independent functioning and self reliance), and cognitive autonomy (i.e., those that increase self-confidence in decision making [17]). Health-care providers may need to educate parents on granting developmentally appropriate autonomy.

Second, the individual needs of the parent should be assessed. Parents of youth with SBM are at risk for internalizing problems (e.g., depression and anxiety) and parental stress. These adjustment outcomes will

likely influence parenting behaviors and child adjustment. Thus, routinely screening for these difficulties is recommended. Increasing parental social support (e.g., parent support groups, spousal support) may serve as a protective factor against such adjustment difficulties. Third, there are several areas for intervening at the family-level. Families of youth with SBM are at risk for decreased family cohesion and poor adaptive functioning. Professionals may reduce the risk for these family difficulties by helping families develop problem solving abilities, redefine family roles and responsibilities, learn positive communication styles, and utilize adaptive coping mechanisms (e.g., problem-focused coping, improved sense of hope).

Lastly, the availability of continuous and seamless health services is crucial in supporting youth and their families during important developmental periods, such as adolescence. For example, the transition from pediatric to adult care should be carefully planned and special support should be provided so that not only are the individual's medical needs attended to, but also that the emotional, developmental, and social needs of the young adult are taken into account. Health care providers are encouraged to help families and individuals with SBM understand normative developmental processes and how they may be affected by SBM as well as provide education about difficulties that individuals with SBM often face in family, social, and academic contexts.

5. Future research directions

Research regarding the impact of SBM on adolescent development and family functioning is still in its early stages. The paucity of research on family functioning within the past 5 to 10 years is particularly notable within the current review. Moreover, advances in technology and medical care have increased the life expectancy of youth with SBM and most of these youth are now entering into the adult years. Unfortunately, little is known about the important transition from childhood to adulthood among youth with SBM, including the increased move toward autonomy (emotional, behavioral), vocational training, and participation in romantic relationships. Such transitions are embedded in the context of the family; therefore studies would benefit from investigating family functioning throughout this emerging adulthood stage. Other areas of exploration in need of further research include the complex and often difficult nature of the transition from pediatric-oriented care to adult care for the adolescent and the family. Finally, much research has been focused on the risks and deficits, rather than strengths and resiliency, among families and youth with SBM.

A number of methodological limitations exist in previous work. To date, the majority of studies have utilized single-method, single-source designs, thus making it difficult to rule out common-method variance interpretations of the findings. Additionally, most studies have been cross-sectional, a design feature which creates difficulties in making conclusions about the directional nature of variables. Future research would benefit from the utilization of multi-method (e.g., observational, self-report), multi-source (e.g., parent, child, teacher, nurse) designs that follow families across time. For a more complete discussion of methodological considerations, refer to the review by Holmbeck and colleagues [30].

6. Conclusion

In this article we have provided an overview of empirical findings related to family functioning in youth with SBM in the context of a biopsychosocial model of adolescent development. Using this framework may help improve our understanding of how primary and secondary features of SBM influence processes of normative development. As noted above, there is considerable variability across individuals and families regarding how they manage the demands of living with SBM. Depending on a variety of individual and environmental conditions, individuals with SBM may have very different outcomes [30]. Our challenge is to better understand the diverse individual and environmental factors that contribute to positive outcomes in youth with SBM and their families, with the ultimate goal of leading them towards a positive trajectory into adulthood.

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