

Paternal correlates of cognitive and behavioral functioning in children with myelomeningocele

Melissa M Wohlfeiler MA, Department of Psychology, The Citadel, Charleston;

Michelle M Macias* MD, Department of Pediatrics, Medical University of South Carolina, Charleston;

Conway F Saylor PhD ABPP, Department of Psychology, The Citadel, Charleston, South Carolina, USA.

*Correspondence to second author at Developmental Pediatrics, Medical University of South Carolina, 135 Rutledge Avenue, PO Box 250567, Charleston, SC 29425, USA.
E-mail: maciasm@musc.edu

DOI: 10.1111/j.1469-8749.2008.03070.x
Published online August 20 2008

This study examined paternal correlates of the cognitive and behavioral functioning of children with myelomeningocele, when controlling for maternal and biological/child correlates as possible sources of variance. Participants were 48 parent dyads of children with myelomeningocele (21 males, 27 females) between the ages of 4 and 12 years (mean 8y, 2mo, SD 2y 3mo). Lesion levels of participants ranged from the thoracic to sacral (thoracic–L3: $n=15$; L4–L5: $n=15$; sacral or lipomeningocele: $n=18$), of whom 38 had been shunted for hydrocephalus. Half of the participants ($n=24$) were community ambulators. Potential predictors of cognitive and behavioral functioning included paternal and maternal parenting stress, as assessed by the Parenting Stress Index – Short Form paternal, and maternal perceptions of support and resources, as assessed by the Family Resource Scale and the Family Support Scale, and child medical severity. Paternal variables significantly correlated with behavioral functioning but not with cognitive functioning. Regression analyses revealed that paternal personal distress and maternal perceived adequacy of social support accounted for significant variance in overall child behavioral functioning. Only child medical severity and annual household income explained significant variance in overall child cognitive functioning. These findings add to the growing body of theory and research documenting that fathers make unique and significant contributions to child adjustment in children with myelomeningocele. Both fathers and mothers need to be considered in interventions supporting development and adjustment of children with myelomeningocele and their families.

The association between myelomeningocele and risk for medical, social, intellectual, and/or psychosocial adjustment problems is well established.^{1–3} Research has shown relationships between medical and environmental factors and cognitive and behavioral functioning of children with myelomeningocele. Biological/medical factors linked to cognitive functioning include hydrocephalus and associated complications,^{4,5} shunt status,⁶ number of shunt revisions,^{7,8} and level of spinal cord lesion.^{7,8} Socioeconomic status of individuals with myelomeningocele has been associated with verbal cognitive outcome.⁷

The association between biological/medical factors and behavioral and psychosocial outcomes of children with myelomeningocele has been less frequently studied.^{4,6} Medical factors associated with behavioral adjustment of children with myelomeningocele may include hydrocephalus, shunt status, and/or shunt revisions;⁴ however, some studies suggest a less significant relationship between these variables.^{9,10} Environmental factors associated with behavioral adjustment in children with myelomeningocele include parental adjustment,¹¹ parenting behaviors,¹² and maternal social support.¹³ Further research is needed to determine specific factors that predict the outcomes of children with myelomeningocele to enhance interventions for treatment and secondary prevention.

Theoretical and empirical literature highlight the importance of all family members and their support systems in the development and adjustment of children with chronic health concerns such as myelomeningocele. One disability-stress-coping model¹⁴ identifies social-ecological factors of parent adjustment and social support as key resistance factors against stress for children with chronic health conditions. Thompson's stress and coping model of child adjustment describes maternal appraisal of stress and supportive family functioning as key aspects of parental and child adaptation.¹⁵

Research is consistent with theory in suggesting that families of children with myelomeningocele have increased stress related to child development and adjustment.^{16,17} Most studies have examined the possible stress effects of having a child with myelomeningocele on the parents, rather than the possible effects of parental stress on child outcomes. Disease characteristics and child adjustment function are reciprocally interactive with subjective experience of parental stress and its resistance factors. Parental stress and adequacy of family support and resources are also key factors in models of development and adjustment.

While paternal variables are implicitly included in models alluding to 'parental' influence and stress relating to the adjustment of children with chronic health issues, fathers are under-researched relative to mothers. Child development research suggests that both the quantity and quality of paternal involvement are associated with positive child outcomes¹⁸ and that both mothers and fathers influence their child's development with unique contributions.^{19–21} A study that examined paternal influences on the adjustment of children with myelomeningocele aged 8 to 11 years, found that fathers' stress relating to parenting, predicted externalizing and internalizing symptoms, and that both fathers' and mothers' psychosocial functioning predicted externalizing symptoms.¹¹ Therefore, parents, including fathers, may play an important role in the behavioral functioning of children with myelomeningocele.

See list of abbreviations at end of paper.

Given that few studies have specifically examined paternal contributors to cognitive and behavioral functioning of children with myelomeningocele, the purpose of this study was to examine paternal correlates of cognitive and behavioral functioning of children with myelomeningocele, when controlling for maternal and biological/child factors as possible sources of variance. This study examined some of the same variables as Friedman et al.¹¹ (e.g. parenting stress, and child internalizing and externalizing behaviors), and examined other factors included in theoretical models (e.g. child cognitive functioning, family resources/support, child medical severity). It was hypothesized that at least some paternal variables would account for significant variance in cognitive and behavioral functioning, over and above the contributions of maternal and biological/child factors, and that fathers and mothers would have unique correlates with the cognitive and behavioral functioning of children with myelomeningocele.

Method

PARTICIPANTS

The study population was derived from a broader study of social support in families of children with myelomeningocele, which included a statewide sample of 93 families of children with myelomeningocele aged 4 to 18 years.³³ This study included participants aged ≤12 years with a mother and a father.

Experts from several disciplines were consulted to establish an operational definition for 'fathers.' Father involvement was defined as participation in the child's care at least 20 hours a week. If there was an uninvolved father and a father figure in the home, information from the father figure who lived in the home was used. The final sample included 48 parent dyads of children aged ≤12 years of with myelomeningocele that had both parents involved with the child's care at least 20 hours per week and information on both dependent variables (cognitive and behavioral functioning). Paternal caretakers included 42 biological or adoptive fathers, five stepfathers, and one grandfather raising the child (all referred to as 'fathers'). Maternal caretakers included 45 biological or adoptive mothers, one stepmother, one grandmother assuming the mother role, and one long-term foster mother (all referred to as 'mothers'). Family and child characteristics are summarized in Tables I and II.

MEASURES

Dependent measures.

Cognitive functioning was measured using the Kaufman Brief Intelligence Test (K-BIT),²² administered by a nurse trained in developmental assessment. The K-BIT (suitable for ages 4–9y) consists of Matrices (nonverbal) and Vocabulary (verbal) subtest standard scores resulting in an overall IQ Composite score (mean 100, SD 15). Internal reliability has been reported as 0.94.²² Behavioral functioning was measured by the Child Behavior Checklist (CBCL; *n*=39 for 6–18 year version, *n*=9 for 1.5–5 year version).^{23,24} The CBCL comprises parent-report rating scales that assess child behavior and emotional problems. T-scores (mean 50, SD 10) are generated for syndrome subscale scores and overall Internalizing, Externalizing, and Total behaviors. Test–retest reliability is 0.85 across scales for the 1.5 to 5 year version and 0.88 to 0.90 for the 6 to 18 year version.

Although potential for overlap exists between the CBCL Somatic Complaints syndrome scale and physical symptoms associated with myelomeningocele, this scale was included as all participants had myelomeningocele and no comparison was made with individuals without myelomeningocele.

Child and family measures.

Mothers and fathers independently completed three measures reflecting perceived personal adjustment, resources, and social support. The Parenting Stress Index–Short Form (PSI-SF)²⁵ measures parenting stress. It includes 36 items assessed on a 5-point Likert-scale, yielding subscale scores for Parental Distress, Parent–Child Dysfunctional Interaction, Difficult Child, and a Total score. The Parental Distress (PD) subscale was used to assess parental perceptions of stress related to personal adjustment factors, including parenting competence, social support, depression, conflict with the other parent, and stress related to restrictions on other life roles. For the PD subscale, test–retest reliability and internal reliability are 0.85 and 0.87 respectively.²⁵

The Family Resource Scale (FRS)²⁶ and the Family Support Scale (FSS)²⁷ were completed by each parent. The FRS measures parents' perceived resources and includes 30 items on adequacy of household resources (e.g. money, food, time), with higher scores indicating more satisfaction. Internal consistency is 0.92 and test–retest reliability is 0.52.²⁶ The FSS measures parents' perceived social support and includes 18 questions about the helpfulness of various sources of support in raising a child, including relatives, friends, and professional services with higher scores indicating more satisfaction. Internal consistency is 0.77, and test–retest reliability is 0.91.²⁷

A Medical Severity Index (MSI) was calculated by rating the severity of physical and medical variables related to myelomeningocele. Information was obtained by interview and chart review and verified by the principal investigator. Variables included for the analyses in this study were level of lesion, shunt status, and shunt revisions. The MSI is an additive score with each variable rated on a Likert-type scale (e.g. thoracic lesion level=3); higher MSI scores represent greater impairment.

Table I: Family characteristics

Fathers' age	Range 25–77y, Mean 39y 7mo, SD 9y 3mo
Mothers' age	Range 23–76y, Mean 38y 8mo, SD 8y 10mo
Marital status, %	
Married	86
Divorced	8
Separated	4
Single	2
Income ^a , %	
<\$10 000	10
\$10 000 to \$25 000	15
\$25 000 to \$50 000	44
\$50 000 to \$75 000	25
>\$75 000	4
Did not report	2

^aIncome per annum in US dollars.

PROCEDURE

Ethical approval for the study was obtained from the Institutional Review Board (IRB) of the Medical University of South Carolina. Participants were recruited statewide from family

Table II: Child characteristics

Age	<i>Range 4-12y, Mean 8y 2mo, SD 2y 3mo</i>	
Sex, <i>n</i>		
Males		21
Females		27
Ethnic group, %		
Caucasian		71
African-American		27
Native American		2
Level of lesion, %		
Thoracic to lumbar 3		31
Lumbar 4 to lumbar 5		31
Sacral, lipomeningocele, or other		38
Ambulation ^a , %		
Community ambulatory		50
Household ambulator		4
Non-functional ambulator		6
Non-ambulator		40
Bowel control, %		
Continent or too young		31
Infrequent accidents (1-2/month)		27
Occasional accidents (1-2/week)		27
Frequent accidents (1-2/day)		15
Bladder, %		
Continent or too young		21
Clean intermittent catheterization: dry		25
Clean intermittent catheterization: wet		25
Vesicostomy		4
Incontinent		25
Shunt, %		
Yes		79
No		21
Shunt revisions, %		
None		50
One revision		15
Two or more revisions		35

Variable	Range	Mean (SD)
Description of independent and dependent variables scores		
K-BIT Composite	40-124	88.02 (22.04)
K-BIT Matrices	35-141	87.06 (24.32)
K-BIT Vocabulary	40-128	89.48 (22.12)
CBCL Total	25-75	54.27 (10.16)
CBCL Internalizing	33-81	54.02 (9.62)
CBCL Externalizing	33-72	50.98 (10.59)
MSI	1-6	3.58 (1.66)
Paternal PSI-PD	1-96	42.55 (31.53)
Maternal PSI-PD	1-97	45.38 (33.53)
Paternal FRS-Total	33-150	113.92 (23.69)
Maternal FRS-Total	45-145	115.31 (20.44)
Paternal FSS-Total	6-68	28.46 (15.46)
Maternal FSS-Total	6-58	28.10 (13.00)

^aHoffer classification³²

K-BIT, Kaufman Brief Intelligence Test; CBCL, Child Behavior Checklist; MSI, Medical Severity Index; PSI-PD, Parenting Stress Index Short Form - Parental Distress subscale centile; FRS, Family Resource Scale; FSS, Family Support Scale.

support groups and/or medical clinics, in accordance with the IRB protocol. The regional study coordinator contacted families for participation in the study. Informed consent and Health Insurance Portability and Accountability Act documents were reviewed and signed with copies provided to the families. Interviews were conducted at home, in the clinic, or in the local health department. Parents completed the family measures (PSI-SF, FSS, and FRS) while the interviewer assessed the child (K-BIT). If both parents were not present, one set of family measures was completed at the interview and the other parent mailed their completed measures in an addressed, stamped envelope. Parents completed one parent report of child behaviors (CBCL) with the interviewer. In 17% of parent dyads both parents completed forms during the interview, in 54% both completed the forms at home and returned by mail, and in 29% the mother completed the forms on site and the father returned completed forms by mail. The *t*-tests comparing families completing forms in the interview versus completing them on their own showed no significant difference in child, parent, or demographic characteristics.

ANALYSIS

Preliminary Pearson's correlation analyses aimed to identify any significant child, maternal, or paternal correlates of both outcomes. Stepwise regression analyses were used to analyze the hypothesis that paternal variables would contribute to the cognitive and behavioral functioning of children with myelomeningocele, even after controlling for the contributions of biological/child and maternal variables. Stepwise regressions were chosen because they allowed the entering of one specific variable ahead of others. Only those variables that were significantly correlated with specific outcomes were included in the regression analyses.

To control for myelomeningocele-related impairments in the regression analyses for behavioral functioning, child MSI and K-BIT Composite scores were entered into the regression before paternal or maternal variables. Four different models were used to analyze the variables. In the first model, contributions of fathers were tested with maternal variables removed, first entering biological/child variables significantly correlated with behavioral functioning and then entering significant paternal variables. In the second model, contributions of mothers were tested with paternal variables removed, again, first entering significant biological/child variables and then entering maternal variables that were significant correlates of behavioral functioning. The third model tested the contributions of fathers over and above mothers on behavioral functioning. All biological/child variables significant in Models 1 or 2 were entered first, followed by maternal variables significant in Model 2, followed by paternal variables significant in Model 1. The final model included all biological/child variables found to be significant in the previous stepwise regressions followed by significant paternal variables and then significant maternal variables.

Results

CORRELATIONAL ANALYSES

Correlations among the parent self-report measures ranged from 0.12 to 0.59. Most measures were significantly correlated with one another, raising concerns of collinearity. However, highly correlated variables (e.g. paternal stress, paternal social support) were included with the rationale that the sig-

nificant correlations only explained a small percentage of the variance, and the regression models selected would further determine the variance contributed by each variable. Measures with overlapping item content (e.g. CBCL Total score, CBCL Externalizing and Internalizing scores) were never used in the same model. Tolerance levels ranged from 0.62 to 1.00 for variables included in the models across regression analyses, indicating that multicollinearity was not a serious problem. Results of the Pearson's correlations are summarized in Table III.

COGNITIVE FUNCTIONING

Stepwise regression analyses revealed that the model accounting for the greatest amount of variance in cognitive functioning as measured by K-BIT Composite scores included both child medical severity (MSI; standardized $\beta=-0.48, p<0.001$) and annual household income ($\beta=0.30, p<0.02$), explaining 32% of the variance. Child MSI score ($\beta=-0.43, p<0.003$) was the only significant correlate of K-BIT Matrices scores and explained 18% of the variance. The model accounting for the most amount of variance in K-BIT Vocabulary scores (19%) included mothers' education ($\beta=0.29, p<0.05$) and annual household income ($\beta=0.42, p<0.02$). No paternal variables correlated with cognitive outcomes.

BEHAVIORAL FUNCTIONING

The unique contributions of maternal and paternal variables were tested using the four stepwise regression models previously described, with CBCL Total scores as the dependent variable. In all models, MSI and K-BIT scores were entered into the model first to account for any variance related to child myelomeningocele status. The four models are summarized in Table IV.

When only biological/child variables (MSI, K-BIT Composite) and paternal variables (paternal PSI-PD, FSS, FRS) were included in the regression analysis, paternal parental distress (PSI-PD) alone explained significant variance (16%) in total behavior problems. When only biological/child variables and maternal variables (maternal PSI-PD, FSS, FRS) were entered

into the stepwise regression, maternal adequacy of family support (FSS) alone accounted for significant variance (14%) in total behavior problems.

When the significant predictors from Models 1 and 2 were entered with maternal variables entered into the model before paternal variables (Model 3), only maternal perceived family support accounted for significant variance (14%) in CBCL Total scores. However, when the significant paternal variable (from Model 1) was entered before the significant maternal variable (from Model 2), only paternal parental distress explained significant variance (16%) in CBCL Total scores.

To determine if different patterns emerged with internalizing and externalizing behaviors as the dependent variables, stepwise regression analyses were completed using CBCL Internalizing and Externalizing scores. As with CBCL Total scores, when only paternal variables were entered (Model 1) using internalizing behaviors as the dependent variable, paternal parental distress was the only significant variable, accounting for 11% of the variance in CBCL Internalizing scores ($\beta=0.34, p<0.02$). In Model 2 the only maternal variable that explained significant variance (11%) in CBCL Internalizing scores was maternal perceived parental distress ($\beta=0.33, p<0.02$). When the two significant variables in Models 1 and 2 were entered with maternal variables first, only maternal parental distress accounted for significant variance (11%) in child internalizing behaviors. When paternal parental distress was entered before maternal parental distress, only paternal parental distress explained significant variance (11%) in CBCL Internalizing scores.

When CBCL Externalizing scores were used as the dependent variable a different pattern emerged. The only significant correlates in the preliminary Pearson's correlations were paternal variables; therefore, only paternal variables were entered into the stepwise regression (see Table III). After controlling for biological/child variables, the model that accounted for the most variance (23%) in externalizing behavior included fathers' age ($\beta=-0.27, p<0.05$) and paternal parental distress ($\beta=0.38, p<0.005$).

Table III: Pearson correlations to examine potential correlates of cognitive and behavioral functioning^a

Potential predictors	K-BIT (n=48)			CBCL (n=48)		
	Composite	Matrices	Vocabulary	Total	Internalizing	Externalizing
MSI	-0.48 ^f	-0.4 ^f	-0.24	0.13	0.24	-0.09
Children's sex ^b	0.22	0.24	-0.03	-0.07	-0.05	-0.02
Children's age	0.10	0.10	-0.03	0.16	0.15	0.01
Annual income	0.29 ^d	0.23	0.44 ^f	0.06	0.17	-0.04
Fathers' age	0.12	0.04	-0.03	-0.15	-0.18	-0.29 ^d
Mothers' age ^c	0.17	0.13	-0.01	-0.09	-0.13	-0.20
Fathers' education	0.05	0.08	0.15	0.03	0.03	-0.04
Mothers' education	0.12	-0.03	0.29 ^d	-0.17	0.02	-0.19
Paternal PSI-PD	-0.08	-0.09	-0.11	0.40 ^c	0.34 ^c	0.39 ^c
Maternal PSI-PD	0.03	0.02	0.03	0.25 ^d	0.33 ^d	0.07
Paternal FRS Total score	0.17	0.16	0.10	-0.25 ^d	-0.29 ^d	-0.15
Maternal FRS Total score	-0.00	0.15	-0.05	-0.30 ^d	-0.32 ^d	-0.15
Paternal FSS Total score	0.14	0.04	0.06	-0.36 ^c	-0.31 ^d	-0.28 ^d
Maternal FSS Total score	-0.01	0.01	-0.01	-0.37 ^c	-0.29 ^d	-0.23

^aOne-tailed test of significance was used for all correlations. ^bHigher values equal female and lower equal male. ^cInformation on age was missing for one mother so variables n=47 for this variable on all outcome measures. ^d $p\leq 0.05$. ^e $p\leq 0.01$. ^f $p\leq 0.001$. K-BIT, Kaufman Brief Intelligence Test; CBCL, Child Behavior Checklist; MSI, Medical Severity Index; PSI-PD, Parenting Stress Index Short Form - Parental Distress subscale centile; FRS, Family Resource Scale; FSS, Family Support Scale.

Discussion

This study examined the relative contributions of paternal distress, social support, and resources, to cognitive and behavioral functioning of children with myelomeningocele in the context of maternal and biological/child variables. Paternal variables were hypothesized to account for significant variance in cognitive and behavioral functioning, even when controlling for maternal and biological/child factors. Results showed that paternal personal distress was the biggest contributor to behavioral functioning in all regression analyses, except for internalizing behaviors where maternal and paternal parental distress contributed equally. The only socio-demographic variables related to child cognitive functioning were maternal education and household income.

Different relationships were detected between maternal and paternal variables and specific areas of child functioning. Unique maternal and paternal correlates imply distinct parental contributions to the development and adjustment of children with myelomeningocele and distinct relationships for both maternal and paternal stress and resources. Correlations between child adjustment and paternal variables of personal distress, social support, and resources are consistent with theoretical models linking the adjustment of children with special health-care needs to parental stress and resources.^{14,15,28,29} As theoretical models of development and adjustment of children with special health-care needs are elaborated and tested, paternal roles should be further examined and explicitly included.

Correlational analyses cannot provide information about the direction of causality. The relationship between paternal support or distress and child behavior problems is likely to be bi-directional, and should be addressed from either side (i.e. additional support for fathers or behavioral interventions for the child with myelomeningocele) or ideally from both sides.

These findings are consistent with research across multiple groups of children with disabilities suggesting that child behavior is the single biggest predictor of parenting stress, even more than child physical impairment, diagnosis, sex, or demographic factors.³⁰ Child behavior can be a causal contributor to parenting stress in children with myelomeningocele.¹¹ Regardless of which came first, families of children with myelomeningocele and parents under stress and/or children with behavior problems need additional support

beyond that typically offered in follow-up of children with myelomeningocele.

Most studies examining stress in families of children with myelomeningocele investigated the effects of the child on the parents rather than the possible effects of parental stress on child outcomes.^{16,17} This study found that paternal distress was the most important paternal variable and was comparable to maternal variables in explaining the variance in behavioral functioning. Clearly, fathers co-parenting children with myelomeningocele should be personally involved and supported for the well-being of the fathers and for the enhanced behavioral/emotional adjustment of the child.

Interpretation of our findings requires additional methodological considerations. Parents collaborated to provide unified measures of child behavior; however, all of the potential maternal and paternal predictors were assessed with self-reports, raising the possibility of multicollinearity. Although multicollinearity statistics did not indicate significant problems, it is preferable to have objective data, minimal correlation between variables, and collaborating data from more than one source, especially as behavior may vary across settings. Future studies should use an independent respondent for dependent measures (e.g. teachers), and employ a prospective longitudinal design.

A limitation of this study was the inability to compare between participants with and without an involved father due to the limited number of children without an involved father. Also, this study only examined the potential influence of fathers on children with myelomeningocele without making comparisons with other populations (e.g. other disability groups or children without disabilities). Future research should compare children with myelomeningocele with and without fathers, and with children without disabilities.

Conclusion

Despite some limitations, this study contributes to the sparse research available on fathers and children with disabilities, especially myelomeningocele. The findings further support the importance of fathers and mothers in child behavioral adjustment. Seagull³¹ identified the inclusion of fathers as a critical issue in pediatric populations and suggested that clinicians are 'setting themselves up to fail' if they rely exclusively on maternal report without encouraging father's to

Table IV: Child CBCL Total scores predicted by significant biological/child, maternal, and paternal characteristics (n=48)^a

Variable	β Model 1 (Child, paternal)	β Model 2 (Child, maternal)	β Model 3 (Child, maternal, then paternal)	β Model 4 (Child, paternal, then maternal)
MSI	0.09	0.08	0.08	0.09
K-BIT Composite	-0.10	-0.14	-0.14	-0.10
Paternal PSI-PD	0.40 ^b	-	0.29	0.40 ^b
Maternal PSI-PD	-	0.12	-	-
Paternal FRS Total score	-0.02	-	-	-
Maternal FRS Total score	-	-0.15	-	-
Paternal FSS Total score	-0.17	-	-	-
Maternal FSS Total score	-	-0.37 ^b	-0.37 ^b	-0.24
R ²	0.16 ^b	0.14 ^b	0.14 ^b	0.16 ^b

^aSummary of standard coefficients (standardized betas) and R² (variance accounted for by each model). ^bp<0.01.

CBCL, Child Behavior Checklist; MSI, Medical Severity Index; K-BIT, Kaufman Brief Intelligence Test; PSI-PD, Parenting Stress Index Short Form – Parental Distress subscale centile; FRS, Family Resource Scale; FSS, Family Support Scale; R², R squared for regression model, which is also percent of variance accounted for by the model.

share their perspective on clinical intervention issues. Fathers should receive as much support, education, and attention as mothers. This may be particularly true when children with myelomeningocele are displaying externalizing behavioral problems, which appear particularly distressing to fathers. Optimal outcomes for children with myelomeningocele depend on both parents having good support, stress management, and parental education.

Accepted for publication 3rd January 2008.

Acknowledgements

This research was supported by a grant from the American Association of Medical Colleges and The Centers for Disease Control. This study was presented in part at the 58th Annual Meeting of the American Academy of Cerebral Palsy and Developmental Medicine in September of 2004.

References

1. Dennis M, Landry SH, Barnes M, Fletcher JM. A model of neurocognitive function in spina bifida over the life span. *J Int Neuropsychol Soc* 2006; **12**: 285–296.
2. Holmbeck GN, Westhoven VC, Phillips WS, et al. A multimethod, multi-informant, and multidimensional perspective on psychosocial adjustment in preadolescents with spina bifida. *J Consult Clin Psychol* 2003; **71**: 782–96.
3. Blum RW, Pfaffinger K. Myelodysplasia in childhood and adolescence. *Pediatr Rev* 1994; **15**: 480–84.
4. Fletcher JM, Brookshire BL, Landry SH, et al. Behavioral adjustment of children with hydrocephalus: relationships with etiology, neurological, and family status. *J Pediatr Psychol* 1995; **20**: 109–25.
5. Wills KE. Neuropsychological functioning in children with spina bifida and/or hydrocephalus. *J Clin Child Psychol* 1993; **22**: 247–65.
6. 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**: 817–32.
7. Bier JB, Morales Y, Liebling J, Geddes L, Kim E. Medical and social factors associated with cognitive outcome in individuals with myelomeningocele. *Dev Med Child Neurol* 1997; **39**: 263–66.
8. Holler KA, Fennell EB, Crosson B, Boggs SR, Mickle JP. Neuropsychological and adaptive functioning in younger versus older children shunted for early hydrocephalus. *Child Neuropsychol* 1995; **1**: 63–73.
9. Wallander JL, Feldman WS, Varni JW. Physical status and psychosocial adjustment in children with spina bifida. *J Pediatr Psychol* 1989; **14**: 89–102.
10. Wallander JL, Varni JW, Babani L, Banis HT, DeHaan CB, Wilcox KT. Disability parameters, chronic strain, and adaptation of physically handicapped children and their mothers. *J Pediatr Psychol* 1989; **14**: 23–42.
11. Friedman D, Holmbeck GN, Jandasek B, Zukerman J, Abad M. Parent functioning in families of preadolescents with spina bifida: longitudinal implications for child adjustment. *J Fam Psychol* 2004; **18**: 609–19.
12. Holmbeck GN, Shapera WE, Hommeyer JS. Observed and perceived parenting behaviors and psychosocial adjustment in preadolescents with spina bifida. In: Barber BK, editor. *Intrusive parenting: how psychological control affects children and adolescents*. Washington, DC: American Psychological Association. 2002: 191–234.
13. Barakat LP, Linney JA. Children with physical handicaps and their mothers: The interrelation of social support, maternal adjustment, and child adjustment. *J Pediatr* 1992; **17**: 725–39.
14. Wallander JL, Varni JW. Adjustment in children with chronic physical disorders: programmatic research on a disability-stress-coping model. In: La Greca AM, Siegel L, Wallander JL, Walker CE, editors. *Stress and coping in child health*. New York: Guilford Press. 1992: 279–98.

15. Thompson RJ, Jr, Gustafson KE, Hamlett KW, Spock A. Stress, coping, and family functioning in the psychological adjustment of mothers of children with cystic fibrosis. *J Pediatr Psychol* 1992; **17**: 573–85.
16. 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**: 167–81.
17. Vermaes IPR, Janssens JMAM, Bosman AMT, Gerris JRM. Parents' psychological adjustment in families of children with spina bifida: a meta-analysis. *BMC Pediatr* 2005; **5**: 32.
18. Cabrera NJ, Tamis-LeMonda CS, Bradely RH, Hofferth S, Lamb ME. Fatherhood in the twenty-first century. *Child Dev* 2000; **71**: 127–36.
19. Lewis C, Lamb ME. Fathers' influences on children's development: the evidence from two-parent families. *Eur J Psychol Educ* 2003; **18**: 211–28.
20. Lamb ME, Lewis C. The development and significance of father-child relationships in two-parent families. In: Lamb ME, editor. *The role of the father in child development*. 4th edn. Hoboken, NJ: John Wiley & Sons, Inc. 2004; 272–306.
21. Rohner RP, Veneziano RA. The importance of father love: history and contemporary evidence. *Rev Gen Psychol* 2001; **5**: 382–405.
22. Kaufman AS, Kaufman NL. Kaufman Brief Intelligence Test. Circle Pines, MN: American Guidance Service, 1990.
23. Achenbach TM, Rescorla LA. Manual for the ASEBA Preschool Forms & Profiles. Burlington, VT: University of Vermont, Research Center for Children, Youth, & Families, 2000.
24. Achenbach TM, Rescorla LA. Manual for the ASEBA School-Age Forms & Profiles. Burlington, VT: University of Vermont, Research Center for Children, Youth, and Families, 2001.
25. Abidin RR. Parenting Stress Index Professional Manual. 3rd edn. Odessa, FL: Psychological Assessment Resources, 1995.
26. Dunst CJ, Leet HF. Measuring the adequacy of resources in households with young children. *Child Care Health Dev* 1987; **13**: 111–25.
27. Dunst CJ, Jenkins V, Trivette CM. Family Support Scale: reliability and validity. *J Indiv Fam Community Wellness* 1984; **1**: 45–52.
28. Chaney JM, Mullins LL, Frank RG, et al. Transactional patterns of child, mother, and father adjustment in insulin-dependent diabetes mellitus: a prospective study. *J Pediatr Psychol* 1997; **22**: 229–44.
29. Kazak AE, Rourke MT, Crump TA. Families and other systems in pediatric psychology. In: Roberts M, editor. *Handbook of pediatric psychology*. 3rd edn. New York: Guilford Press. 2003: 159–75.
30. Saylor CF, Macias MM, Spratt EG, Gonzalez A, Wohlfeiler M. Predictors of parenting stress in youth with special health care needs. http://www.pasmeeting.org/2009Baltimore/abstract_archives.asp
31. Seagull EA. Beyond mothers and children: finding the family in pediatric psychology. *J Pediatr Psychol* 2000; **25**(3): 161–70.
32. Hoffer MM, Feiwell E, Perry R, Perry J, Bonnet C. Functional ambulation in patients with myelomeningocele. *J Bone Joint Surg Am* 1973; **55**: 137–48.
33. Macias MM. Social Support in Children and Adolescents with Neural Tube Defects and their Families. Final Grant Report, _MM-0140-02/02. Centers for Disease Control and Prevention, 1973.

List of abbreviations

CBCL	Child Behavior Checklist
FRS	The Family Resource Scale
FSS	Family Support Scale
K-BIT	Kaufman Brief Intelligence Test
MSI	Medical Severity Index
PD	Parental Distress subscale of PSI-SF
PSI-SF	Parenting Stress Index-Short Form
