

Maternal mental health in families of children with spina bifida

LC Ong, NAR Norshireen, V Chandran

Kuala Lumpur, Malaysia

Background: This study aimed to compare mental health of mothers of children with spina bifida with mothers of able-bodied controls.

Methods: Eighty-one mothers of children with spina bifida aged 1-18 years completed the General Health Questionnaire-12 (GHQ-12) and Parenting Stress Index Short Form (PSI/SF). The controls were 69 mothers of children with acute, non-disabling illnesses. Each child's adaptive skills were assessed using the Vineland Adaptive Behaviour Scales (VABS). Logistic regression analysis was used to determine factors related to a high GHQ score (≥ 3) in all patients.

Results: Compared to the controls, mothers of children with spina bifida had lower educational levels and were more likely to be the main caregivers and not working. Nineteen (23.5%) of them had a high GHQ score compared to 5 (7.2%) of the controls. They also had significantly higher scores for total PSI/SF and the parent domain, difficult child (DC) and parent-child dysfunctional interaction subscales. Children with spina bifida had lower scores for the composite VABS and communication, socialization, daily living skills and motor sub-domain than the controls. Spina bifida (odds ratio [OR] 4.3, 95% confidence interval [CI] 1.30-14.23), higher DC scores (OR 1.1, 95% CI 1.00-1.16), and higher life stress scores (OR 1.1, 95% CI 1.01-1.71) were associated with a high GHQ score.

Conclusion: Spina bifida, recent stressful life change events and maternal perception of a child as 'difficult' are associated with poor maternal psychological health.

World J Pediatr 2011;7(1):54-59

Author Affiliations: Department of Pediatrics, Universiti Kebangsaan Malaysia Medical Centre, Jalan Yaacob Latif, 56000 Kuala Lumpur, Malaysia (Ong LC, Chandran V); Department of Pediatrics, Institute of Pediatrics, Jalan Raja Muda, 50300 Kuala Lumpur, Malaysia (Norshireen NAR)

Corresponding Author: Ong Lai Choo, MRCP, Department of Pediatrics, Universiti Kebangsaan Malaysia Medical Centre, Jalan Yaacob Latif, Bandar Tun Razak, 56000 Kuala Lumpur, Malaysia (Tel: 603 91455383; Fax: 603 91456637; Email: onglc@ppukm.ukm.my)

doi:10.1007/s12519-011-0246-z

©Children's Hospital, Zhejiang University School of Medicine, China and Springer-Verlag Berlin Heidelberg 2011. All rights reserved.

Key words: life change events; mental health; parent-child relations; spina bifida

Introduction

Spina bifida (SB) is a common congenital birth defect worldwide, and long-term problems related to the disorder include motor weakness and ambulatory difficulties, bowel and urinary incontinence, hydrocephalus and shunt malfunction, skin breakdown and school difficulties.^[1] The variability of morbidity in each individual with SB highlights the importance of not only providing multidisciplinary care, but also "individualising" the care to cater to the child's needs that change over time. This invariably places demands on both the community and family. Families of children with SB have to deal with large amounts of medical information, make major adjustments to their daily routines, and make major decisions on medical, educational and social care at critical stages of the children's life.

A meta-analysis showed that SB has a negative effect on parental psychological adjustment, especially the parent-child relationship.^[2] However, this effect is more heterogenous for mothers than for fathers, and identification of possible reasons for this effect has yielded mixed findings. An evidence-based review of family functioning in children with SB highlighted the limitations in generalizability of the studies due to sampling bias, as most of the studies involved the Western Caucasian middle income population.^[3] In a multicultural society such as Malaysia, with a different social, educational and medical services model, it is even more difficult to extrapolate results from the available literature. A previous local study revealed a high incidence of co-morbidities in SB children but did not explore the psychological impact of chronic disability on the caregiver.^[1] Hence, this study was undertaken to determine whether mothers of SB children had poorer mental health than those of healthy able-bodied children, and to explore other factors related to poor maternal mental health.

Methods

Patients and controls

This prospective cross-sectional hospital-based study was conducted on mothers of children aged 1-18 years attending the SB Clinics of the Institute of Pediatrics Kuala Lumpur and Universiti Kebangsaan Malaysia Medical Center. Both clinics catered for the surrounding urban population but also served as tertiary centers for patients from adjacent rural areas. Controls were mothers of children with acute, non-disabling conditions (febrile fits, viral fevers, acute respiratory tract infections and acute gastroenteritis) attending the general pediatric clinics or admitted into the pediatric wards of both hospitals. Exclusion criteria included non-citizens (who did not have access to government funded healthcare), those who had incomplete multidisciplinary assessments, and those who were unable to read and complete the questionnaire or declined to participate in.

Measures

Mothers of SB and control groups were given information regarding the purpose and method of the study. Those who consented to participate in the study were required to complete the self-administered General Health Questionnaire-12 (GHQ-12)^[4] and the Parenting Stress Index Short Form (PSI/SF) questionnaire.^[5] Demographic data and medical information were verified by a combination of direct interviews, physical examinations and case notes' review.

The GHQ-12 was originally designed as a screening test to detect minor (non-psychotic) psychiatric disorders in the community setting. With a high degree of validity, it is widely used internationally and has been applicable in both community and clinical settings. Both the original English and translated Bahasa Malaysia versions have been validated in the Malaysian population, and a cut off score between 2 and 3 was reported to have a sensitivity of 86% and a specificity of 85%.^[6] In this study a GHQ score of 3 or more was suggestive of poor mental health.

The PSI/SF has 36 items that make up three subscales: parental distress (PD), parent-child dysfunctional interaction (P-CDI), and difficult child (DC). The PD subscale assesses perceived stress to personal adjustment factors in her role as a parent. The P-CDI subscale focuses on the parent's perception that the child does not fulfil her expectations and their interactions are not reinforcing her as a parent. The DC subscale focuses on some of the basic behavioural characteristics of the children that make them difficult to manage. The total PSI/SF score, obtained by summing the scores of the 3 subscales, indicates the overall level

of stress a parent is experiencing, with higher scores indicating more stress. Mothers who obtain a score of ≥ 90 th percentile in the total PSI/SF or subscales are said to be experiencing clinically significant levels of stress.^[5]

The PSI also yielded a life stress score, which is a cumulative score of life change events that occurred in the past year of the respondent's life that might contribute to stress, e.g., changes in work, financial or marital status, recent birth or death in the family, new home or school environment or problems with the law.

The child's functional skills were measured using the Vineland Adaptive Behavior Scales (VABS), interview edition.^[7] The VABS is a structured interview and was carried out by two of the researchers (VC and NNAR). This yielded four domains of function: communication, daily living skills (DLS), socialization, and motor skills. A composite adaptive score, obtained by averaging the communication, DLS and socialization scores, represents the overall level of adaptive skills of the child. Results are reported in standard scores (normative mean 100 S.D.15) with lower scores indicating more dysfunction. A child is considered to have inadequate adaptive skills if the standard score is less than 85.

Statistical analysis

Based on an alpha of 0.05 and a power of 0.8, a sample size of 74 in each group was required to demonstrate a four-fold risk of obtaining a high GHQ (>3) score among mothers of children with SB, assuming that 5% of the controls had high scores. Univariate analysis was performed using student's *t* test (the Mann-Whitney *U* test for nonparametric data) for continuous variables and Chi-square analysis (Fisher's exact test for cell values <5) for categorical variables. Logistic regression analysis (SPSS for Windows Version 15.0, 2006, SPSS Inc., Chicago, USA) was performed to determine factors associated with a high GHQ score in all mothers. To reduce the number of variables that could be put into the regression equation, only factors that achieved statistical significance on univariate analysis were subjected to binary logistic regression analysis. A *P* value less than 0.05 was considered statistically significant. For the individual dimensions of PSI and VABS, Bonferroni correction was used to control type 1 errors, and between group differences were considered statistically significant only if the *P* value was less than 0.013.

Results

Altogether 81 children with SB and 69 controls participated in the study. The socio-demographic differences between the SB and control groups are shown in Table 1. Compared to the controls, more

mothers of SB children were not working and were the main caregivers. There were a lower maternal educational level, lower family income and fewer siblings in the SB group than the control group. There were no differences between the two groups in terms of gender, ethnicity, paternal educational level and occupation, single parent status or maternal age.

Nine (11.1%) of the SB children had a lesion involving the thoracic region, 6 (7.4%) the upper lumbar region, 39 (48.1%) the lumbosacral region, and 27 (33.4%) the sacral region. Nine (11.1%) of the SB children had a closed myelomeningocele, 49 (60.5%) an open lesion which had been repaired in early life, 19 (23.5%) a lipomyelomeningocele, and 4 (4.9%) other lesions. Thirty-six (44.4%) children had hydrocephalus. Forty-four (54.3%) children were on a clean intermittent urinary catheterization regimen. Forty-two (51.9%) walked without aids, 15 (18.5%) ambulated upright with assistant devices, 19 (23.5%) used wheelchairs, and 5 (6.1%) were totally dependent on their caregivers for mobilization.

There were differences in maternal GHQ-12, PSI/

SF and child VABS scores between the SB and control groups (Table 2). A large proportion of mothers of SB children had higher GHQ-12 scores compared to those of the controls. These mothers also had higher mean total PSI/SF and subscale scores. However, between groups differences of clinical significance (taken as scores ≥ 90 th percentile) were only for P-CDI subscale. There were no statistical differences in life stress scores between the two groups. Children with SB had lower mean composite VABS and all 4 subdomain scores, and a larger proportion of them had inadequate adaptive skills than the controls.

The factors associated with high GHQ-12 scores are shown in Table 3. Univariate analysis revealed having a SB child, higher life stress, PD and DC scores from the PSI/SF, and lower motor scores from the VABS were statistically significant. When these variables were subjected to a forward logistic regression analysis, the only significant factors were the presence of a SB child (odds ratio [OR] 4.3, 95% confidence interval [CI] 1.30-14.23), higher DC scores (OR 1.1, 95% CI 1.00-1.16), and life stress scores (OR 1.1, 95% CI 1.01-1.71).

Table 1. Sociodemographic differences between children with spina bifida and the controls

Variables	Spina bifida, n=81 (%)	Control, n=69 (%)	P value
Age, y, mean (SD)	6.8 (3.9)	6.6 (3.0)	0.717
Male gender	44 (54.3)	35 (50.7)	0.660
Ethnicity			
Malay	41 (50.6)	39 (56.5)	0.726*
Chinese	21 (25.9)	17 (24.6)	
Indian	19 (23.5)	13 (18.9)	
Maternal age, y, mean (SD)	35.7 (7.17)	35.3 (6.61)	0.633
Single parent	6 (7.4)	2 (2.9)	0.289
Socioeconomic status			
Maternal education, y, mean (SD)	10.4 (2.82)	11.4 (2.21)	0.025
Paternal education, y, mean (SD)	11.1 (2.72)	11.6 (2.40)	0.295
Maternal occupation			
Professional	13 (16.0)	19 (27.5)	0.004*
Skilled/semiskilled	15 (18.5)	25 (36.3)	
Unskilled	10 (12.3)	3 (4.3)	
Unemployed	43 (53.2)	22 (31.9)	
Paternal occupation			
Professional	20 (24.7)	18 (26.1)	0.294*
Skilled/semiskilled	44 (54.3)	44 (63.8)	
Unskilled	16 (19.8)	6 (8.7)	
Unemployed	1 (1.2)	1 (1.4)	
Monthly family income, median (IQR)	2000 (1250-3750)	3000 (2000-4000)	0.005
Sibship size, median (IQR)	2 (0.5-3.5)	3 (2-4)	0.034
Primary caregiver			
Mother alone	48 (59.3)	18 (26.1)	0.001*
Mother with help	18 (22.2)	20 (29.0)	
Others	15 (18.5)	31 (44.9)	

SD: standard deviation; IQR: inter quartile range; *: Chi-square test with (df-1) degrees of freedom. Figures in parentheses indicate percentages unless indicated otherwise.

Table 2. Comparison of mean scores for the General Health Questionnaire-12 (GHQ), Parenting Stress Index/Short Form (PSI) and Vineland Adaptive Behavior Scales (VABS) between spina bifida and control groups

Domain	Spina bifida, n=81	Control, n=69	P value
GHQ score, median (IQR)	0 (0-1)	0 (0-0)	0.005
No. with score ≥ 3	19 (23.5)	5 (7.2)	0.007*
PSI [§]			
Total PSI score, mean (SD)	86.2 (13.60)	75.2 (18.85)	<0.001
No. with score ≥ 90 th percentile (%)	28 (34.6)	11 (15.9)	0.018
PD score, mean (SD)	29.4 (6.82)	25.7 (7.87)	0.003
No. with score ≥ 90 th percentile (%)	15 (18.5)	9 (13.0)	0.362
DC score, mean (SD)	28.6 (5.78)	24.6 (7.80)	<0.001
No. with score ≥ 90 th percentile (%)	9 (11.1)	7 (10.1)	0.926
P-CDI score, mean (SD)	28.0 (5.60)	24.8 (5.45)	0.001
No. with score ≥ 90 th percentile (%)	41 (50.6)	13 (18.8)	<0.001 [†]
Life stress score, median (IQR)	1 (0-2.5)	1 (0-2)	0.091
VABS [§]			
Composite score, mean (SD)	82.5 (17.48)	96.9 (6.26)	<0.001
No. with score <85 (%)	33 (41.3)	2 (2.9)	<0.001 [‡]
Communication score, mean (SD)	88.8 (20.31)	99.1 (7.18)	<0.001
No. with score <85 (%)	19 (23.5)	2 (2.9)	<0.001 [‡]
Daily living skills score, mean (SD)	82.3 (20.26)	99.0 (9.91)	<0.001
No. with score <85 (%)	33 (40.7)	0 (0)	<0.001 [‡]
Socialization score, mean (SD)	85.7 (19.47)	93.1 (5.06)	<0.001
No. with score <85 (%)	29 (35.8)	3 (4.3)	<0.001 [‡]
Motor skills score, mean (SD)	78.8 (26.14)	105.7 (9.05)	<0.001
No. with score <85 (%)	43 (53.1)	0 (0)	<0.001 [‡]

SD: standard deviation; IQR: inter quartile range; *: defined as having poor mental health (GHQ-12 1988); †: defined as clinically significant levels of stress (Abidin 1990); ‡: defined as having inadequate skills (VABS 1984); §: using Bonferroni correction, $P < 0.013$ is considered statistically significant.

Table 3. Factors associated with poor mental health (GHQ score ≥ 3) in mothers of children with spina bifida and the controls

Variables	GHQ score, ≥ 3 , <i>n</i> =24 (%)	GHQ score ≤ 2 , <i>n</i> =126 (%)	<i>P</i> value
Child's age, y, mean (SD)	7.4 (4.02)	6.6 (3.40)	0.310
Male gender	11 (45.8)	68 (54.0)	0.464
Ethnicity			
Malay	9 (37.5)	71 (56.3)	0.089*
Chinese	6 (25)	32 (25.4)	
Indian	9 (37.5)	23 (18.3)	
Maternal age, y, mean (SD)	36.4 (7.10)	35.3 (6.88)	0.613
Single parent	3 (12.5)	5 (4.0)	0.117
Socioeconomic status			
Maternal education, y, mean (SD)	9.9 (2.42)	11.1 (2.60)	0.057
Paternal education, y, mean (SD)	10.5 (2.62)	11.5 (2.55)	0.098
Paternal occupation			
Professional	7 (29.2)	31 (24.6)	0.330*
Skilled/semiskilled	11 (45.8)	77 (61.1)	
Unskilled	5 (20.8)	17 (13.5)	
Unemployed	1 (4.2)	1 (0.08)	
Mother not working	11 (45.8)	55 (43.7)	0.518
Monthly family income, median (IQR)	2250 (950-3550)	2500 (1250-3750)	0.711
Sibship size, median (IQR)	2 (1-3)	3 (2-4)	0.254
Mother sole caregiver	12 (50.0)	54 (42.6)	0.518
Maternal PSI/SF scores			
PD score, mean (SD)	32.1 (8.88)	26.9 (6.96)	0.012†
DC score, mean (SD)	31.3 (8.91)	26.0 (6.39)	0.009†
PCDI score, mean (SD)	28.3 (4.60)	26.2 (5.88)	0.100
Life stress score, median (IQR)	2 (1-3)	1 (0-2)	0.009†
Child's VABS scores			
Communication score, mean (SD)	87.9 (18.92)	94.8 (15.72)	0.105
Daily Living Skills score, mean (SD)	82.2 (23.62)	91.6 (16.30)	0.018
Socialisation score, mean (SD)	87.8 (12.47)	89.4 (12.67)	0.537
Motor score, median (IQR)	81 (58-104)	99.5 (84.5-114.5)	0.010†

SD: standard deviation; IQR: inter quartile range; *: Chi-square test with (df-1) degrees of freedom; †: statistically significant ($P < 0.013$) following Bonferroni correction. Figures in parentheses indicate percentages unless indicated otherwise.

Discussion

In this study, more mothers of SB children experienced poor mental health (as measured by GHQ-12) than those of children with acute illnesses. The proportion of SB mothers with poor mental health was 23.5%, compared to the proportion of those with psychopathological disorders (19% to 52%).^[8-12] However these figures are not comparable, partly because of different measurement instruments used. Earlier studies used semi-structured interviews, which were later replaced by standardized measures of psychological symptoms. Reliability and validity studies have been conducted in recent years.^[2] Although GHQ-12 has not been widely used in SB study, it is the only standardized measure of psychological symptoms that has been translated and validated for

local use. Nevertheless, a control group is necessary when GHQ-12 is used in the clinical setting locally. The difference in effect size between SB and controls in this and other studies need to be interpreted with care, as it would vary depending on whether the control group comprises children with chronic illnesses,^[10] normal means^[12-16] or normal children from the same community.^[8,11,17] Another confounding factor preventing direct comparison is the variability in the characteristics of the SB population among studies. In our study, less than 10% of SB children were from single-parent families as reported elsewhere,^[9,11] but it was much lower than 15%-52%.^[8,12-17] Our SB children showed less severe physical disability, which was comparable to those recruited from hospitals and registries^[8,11,16] and those performed in the 1970s.^[9,15,18]

There are limitations in generalizing the results in the community as more severe patients were recruited from tertiary centers. In the present study, child's functional skills (communication, daily living skills, socialization or motor domains) were not associated with poor maternal mental health. Tew et al^[15] reported that child's physical disability was a source of maternal stress, our study as well as other studies did not find any association between physical disability and parental adjustment,^[9,11-13,17] which may be due to improved care and psychosocial support for SB children and their families over the decade. Moreover lack of standardization in measuring disease severity in SB children is another factor. We used a standardized measure of adaptive skills,^[17] but other studies used the level of lesion,^[12-14,16] functional limitations such as ambulatory status, bowel and bladder incontinence,^[9,14-15] treatment intensity in terms of hospitalization and number of shunt revision,^[12,14] or standardized measures of physical^[16] or cognitive skills.^[9,15,17] There have been attempts to quantify severity using scoring systems based on some of the mentioned physical characteristics.^[9,12,14,16,17] However, when the level of the lesion is thought to be correlated well with ambulatory status, there is little correlation with other parameters.^[11]

Other than SB, the DC subscale of PSI/SF is associated with poor maternal mental health. DC focuses on the mother's perception of the behaviour and temperament of her child, which make them difficult to manage. Since there was no difference in the proportion of SB mothers and controls with clinically significant high DC stress scores in our study, we speculated that stress was related to child's perceived behaviour problems that affect maternal mental health, irrespective of the duration or nature of the illness. Nevertheless, there is evidence that child behavior problems are associated with parental psychological symptoms

in the SB population.^[14,16,17] Appraised child-related stress rather than the actual severity of child's medical condition has been shown to be associated with maternal psychological well-being in SB cases.^[12] Friedman et al^[19] reported that the longitudinal relationship between parent functioning and child adjustment (especially behavior) in SB tended to be in the direction of parent to child. Hence intervention targeting maternal perception of child behaviour may not only improve mental health of the mother, but also positively affect child's adjustment to disease.

This study also demonstrated the relationship between non-illness life stressors and mental health. Not surprisingly, the original scoring method of GHQ-12 as used in this study detects acute psychological distress better than chronic distress.^[4] Dorner et al^[9] reported that SB mothers attributed their depression more to acute events (which could occur in any family) than child's disability, while Weigner and Donders^[10] found that the presence of a significant new stressor within the 6 months of a study predicted the level of parental distress. The mothers of SB children in our study did not report more stressful life events than the controls, suggesting that factors other than the events alone mediated poor mental health of the mothers. A longitudinal study is needed to determine if the chronic care of SB children, especially during critical periods of the child's life (e.g., surgery in early life, school entry, adolescence), render SB mothers more vulnerable to poor mental health than controls when faced with a similar non-illness related stressful event.

Variations in parents' psychological adjustment are linked to many factors: child (age, behavior and cognitive problems), parent (socio-economic status, appraised stress, coping resources), family (family income, partner relationship, family climate), and environmental factors (social support).^[2] Since the present study has explored some of "risk" factors, it is necessary to look at "protective" factors maintaining good mental health. We found that there is no relationship between maternal mental health, ethnicity, socioeconomic status and the child's adaptive skills. Future studies should therefore focus on parental coping styles and resources,^[8,11-12] family climate^[10,13,16,19] and social support networks.^[13,16-17]

There are also other limitations in our study. PSI/SF and VABS was not validated for the local population, and the control group was not evenly matched for possible confounding socioeconomic factors. Cross-sectional associations limited our inferences on causality. The patient age range was wide, and disease awareness and treatment varied because each SB child was not diagnosed and treated at birth. A longitudinal

study may yield more accurate information on the psychosocial functioning of the families.

Maternal psychological well-being is important not only for child adjustment, but also for maintenance of family cohesion and marital stability.^[10,12,13,16] Psychosocial services are necessitated for families with SB children but this will invariably impose further demands on the healthcare system. It is worth noting that in our study the majority of mothers with SB children (76.5%) did not report poor mental health, implying many were coping with their child's disability. This together with other findings from the present study suggest a more pragmatic approach be given to target mothers who perceive their children as "difficult" and to offer crisis counselling for those facing an acute stressful event.

Acknowledgements

We wish to thank the doctors and medical staff of the Spina Bifida clinics in the Pediatric Institute Kuala Lumpur and UKM Medical Centre for allowing us to interview the patients.

Funding: None.

Ethical approval: Not needed.

Competing interest: None.

Contributors: Ong LC conceptualized the study design, analyzed data and revised the manuscript. Norshireen NAR designed the protocol, collected and analyzed data and drafted the article. Chandran V designed the protocol, collected data and revised the manuscript.

References

- Ong LC, Lim YN, Sofiah A. Malaysian children with spina bifida: relationship between functional outcome and level of lesion. *Singapore Med J* 2002;43:12-17.
- Vermaes IP, Janssens JM, Bosman AM, Gerris JR. Parents' psychological adjustment in families of children with spina bifida: a meta-analysis. *BMC Pediatrics* 2005;5:32.
- Holmbeck GN, Greenley RN, Coakley RM, Greco J, Hagstrom J. Family functioning in children and adolescents with spina bifida: an evidence-based review of research and interventions. *J Dev Behav Pediatr* 2006;27:249-277.
- Goldberg D, Williams P. A user's guide to the General Health Questionnaire. Windsor: NFER-Nelson Publishing Co. Ltd, 1988.
- Abidin RR. Parenting stress Index/Short Form. New York: Psychological Assessment Resources Inc., 1990.
- Maniam T, Ding LM, Lim TO, Toh CL, Aziz A, Sararak S, et al. Psychiatric morbidity in adults. In: National Health and Morbidity Survey. Kuala Lumpur: Public Health Institute, Ministry of Health Malaysia, 1999: Vol. 6.
- Sparrow SS, Balla DA, Cichetti DV. Vineland Adaptive Behavior Scales. Interview Edition - Survey Form Manual. Circle Pines, MN: American Guidance Service, 1984.

- 8 Holmbeck GN, Gorey-Ferguson L, Hudson T, Seefeldt T, Shapera W, Turner T, et al. Maternal, paternal and marital functioning in families of preadolescents with spina bifida. *J Pediatr Psychol* 1997;22:167-181.
- 9 Dorner S. The relationship of physical handicap to stress in families with an adolescent with spina bifida. *Dev Med Child Neurol* 1975;17:765-776.
- 10 Wiegner S, Donders J. Predictors of parental distress after congenital disabilities. *J Dev Behav Pediatr* 2000;21:271-277.
- 11 Grosse SD, Flores AL, Ouyang L, Robbins JM, Tilford JM. Impact of spina bifida on parental caregivers: findings from a survey of Arkansas families. *J Fam Child Stud* 2009;18:574-581.
- 12 Kronenberger WG, Thompson RJ Jr. Medical stress, appraised stress, and the psychological adjustment of mothers of children with myelomeningocele. *J Dev Behav Pediatr* 1992;13:405-411.
- 13 Fagan J, Schor D. Mothers of children with spina bifida: factors related to maternal psychosocial functioning. *Am J Orthopsychiatry* 1993;63:146-152.
- 14 Lemanek KL, Jones ML, Liebermann B. Mothers of children with spina bifida: adaptational and stress processing. *Child Health Care* 2000;29:19-35.
- 15 Tew B, Laurence KM. Some sources of stress found in mothers of spina bifida children. *Br J Prev Soc Med* 1975;29:27-30.
- 16 King G, King S, Rosenbaum P, Goffin R. Family-centered caregiving and well-being of parents of children with disabilities: linking process with outcome. *J Pediatr Psychol* 1999;24:41-53.
- 17 Barakat LP, Linney JA. Optimism, appraisals, and coping in the adjustment of mothers and their children with spina bifida. *J Child Fam Stud* 1995;4:303-320.
- 18 Shehu BB, Ameh EA, Ismail NJ. Spina bifida cystica: selective management in Zaria, Nigeria. *Ann Trop Paediatr* 2000;20:239-242.
- 19 Friedman D, Holmbeck GN, Jandasek B, Zukerman J, Abad M. Parent functioning in families with preadolescents with spina bifida. *J Family Psychol* 2004;18:609-619.

Received December 15, 2009

Accepted after revision April 15, 2010