Health-related quality of life in paediatric spina bifida

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ABSTRACT

Introduction: The average incidence of spina bifida (SB) in Malaysia is 0.43 among 1,000 live births. The burden of the disease and its impact on the overall development and health though tremendously improved, remains significant. Therefore, current patient management strategies must include quality of life (QOL) measures.

Methods: This was a prospective, cross-sectional study on spina bifida children aged 5-20 years, attending the paediatric spina bifida clinics of Universiti Kebangsaan Malaysia Medical Centre Kuala Lumpur and Hospital Tuanku Jaanku Seremban. Scores were obtained using the validated disease specific Parkin QOL questionnaire. Univariate and multivariate analysis were used to investigate factors that were determinants for these outcomes. Results were expressed as beta coefficient and 95% confidence intervals (95%CI).

Results: A total of 54 children and adolescents aged between 5-20 years completed the questionnaires. Presence of neurogenic bowel (p=0.003), neurogenic bladder (p=0.041), shunt (p=0.044), non-ambulators (p=0.007) and being the only child in the family (p=0.037) were associated with lower QOL scores. Multivariate analysis showed presence of neurogenic bowel (β=0.375, 95%CI: 0.00, 0.15) and being the only child in the family (β=0.250, 95%CI: 0.04, 0.17) explained 22.1% of the variance in the QOL mean percentage scores.

Conclusion: Being a single child in the family was the only socio-demographic variable associated with lower QOL scores. Although several clinical factors appeared to contribute significantly to QOL in spina bifida children, the presence of neurogenic bowel had the greatest impact.

KEY WORDS:
Health-related quality of life, spina bifida, neurogenic bowel

INTRODUCTION

Spina bifida is a complex congenital disorder and is the most common of the neural tube defects in the World.[1] The average incidence of spina bifida in Malaysia is 0.43 among 1,000 live births.[2] A modest decrease in the prevalence was seen following an increase in maternal pre-conceptual consumption of folic acid supplements. Despite the decrease in the incidence of spina bifida, there is still an appreciable population of children with this condition. While the prevention of disabling malformations is an important goal, individuals with spina bifida often experience considerable medical and psychosocial morbidity.[3] Long term complications related to the disorder include ambulatory difficulties, bladder and bowel incontinence, hydrocephalus and its associated complications, pressure sores and neuro-cognitive dysfunction. The burden of the disease and its impact on the overall development and health remains significant. Therefore, current patient management strategies must now be focused on their quality of life (QOL).[4]

The World Health Organization (WHO) defines QOL as "A state of complete physical, mental and social well-being and not merely the absence of disease or infirmity". It is important to assess health-related QOL (HRQOL) when dealing with chronic health illnesses. This provides a measure of patient's perception of his or her overall level of functioning with the aim to optimize clinical care.

Existing reports on HRQOL is mainly based on Western population of spina bifida. A descriptive correlational study has shown that stress from spina bifida, communication efficacy and attitude towards spina bifida explained 60% of the variance in the HRQOL. No clinical or demographic variables were significant.[5] Studies performed previously highlighted the negative impact of mobility limitation, urinary and bowel incontinence in spina bifida patients.[6-8] Unfortunately, information from developing countries are scarce. Differences in healthcare practices, social and cultural acceptance may indicate a different pattern of QOL determinants among spina bifida patients in Malaysia. Identifying these factors would provide useful information to improve interventions and provide targeted management strategies to bring about better knowledge and understanding regarding QOL among the spina bifida population in Malaysia and possibly among other similar populations.

MATERIALS AND METHODS

Patient selection

This was a hospital based cross-sectional study. The participants were spina bifida children and adolescents aged 5-20 years, attending the paediatric spina bifida clinics at the Universiti Kebangsaan Malaysia Medical Centre Kuala

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Lumpur (UKMMC) and Hospital Tuanku Ja'afar Seremban (HTJS) between 2013 and 2016. These children with spina bifida have regular follow-up examinations every 3-6 months in these two centres. All patients aged between 5-20 years with written consent from parents or patient were included in the study. Exclusion criteria included children with other pre-existing chronic illnesses or co-morbids, behavioural problems, e.g. attention deficit hyperactive disorder, parents who were unable to read, comprehend or complete the questionnaire and those who declined to participate.

Measures

The validated English version of Parkin SB HRQOL questionnaire was used.[3] Questionnaires were administered in the clinic during the follow-up visits and completed by the primary caregiver or the patient. Permission to use and to translate the questionnaire to Bahasa Malaysia was obtained from the author. Parkin HRQOL questionnaire was developed in 1997 to assess the HRQOL of spina bifida children and adolescents from the viewpoint of the individual themselves as well as their parents. This instrument is disease specific and has good measurement properties, taking into consideration all of the challenges faced by the spina bifida patient. The questionnaire uses 5-point Likert scale to assess the HRQOL. It consists of either 44 questions for the five to12 years old children, or 47 questions for the13 to 20 years old adolescents. Questionnaires were completed by the parents for all patients who are 12 years old and less. The scores ranged from 44-220 and 47-235, respectively. The final score is obtained by adding up individual items, and scoring is reversed for negative questions. To enable cross comparison between the two age groups, these scores were expressed as a percentage of total score with higher percentage indicating a superior quality of life. Social-demographic data and medical history were obtained from a combination of case-note reviews and direct interviews.

Statistical analysis

Descriptive analysis was used to characterise the study population using the Statistical Package for Social Sciences (SPSS) version 22 for Windows (SPSS, Inc., Chicago, IL, USA) software. Univariate analysis was performed using independent t-test to test the association between socio-demographic and medical factors (independent variables) and the Parkin HRQOL mean percentage scores (dependent variable). Factors of clinical interest found to be significant in the univariate analysis (p-value <0.05) were then entered into a stepwise multivariate linear regression analysis to determine the most significant predictors of the HRQOL. Mean and standard deviation were calculated for continuous variables, frequency and percentages for dichotomous variables. A p-value of less than 0.05 was considered statistically significant. All applicable institutional regulations concerning ethical use of human volunteers was followed during the period of this research.

RESULTS

The study population consisted of 61 patients attending the hospital facilities in UKMMC and HTJS who were available for review. Of these, two were lost to follow-up, two were transferred to other hospitals for logistic reasons and three caregivers were unable to answer the questionnaire due to poor comprehension of English and Bahasa Malaysia language. Of the remaining 54 patients with spina bifida, medical records were retrieved and reviewed. Results were presented for all 54 patients (Figure 1).

Table I shows the socio-demographic and clinical characteristics of the study group. There were almost equal numbers of male and female patients. Of those enrolled, 30 were Malays, 11 were Chinese and 13 were Indians. Thirty-two patients (59%) were children aged 5-12 years, and 22 were adolescents 13-20 years old. In all, 12 (22%) children were the only child in the family. Majority of patients (68%) had low level lesions, L5 and below. Of the total 54 patients, 22 (41%) had ventriculo-peritoneal shunt inserted for hydrocephalus. Sixteen (30%) were non-ambulators and wheelchair dependent. Most of our patients had neurogenic bladder and neurogenic bowel. Only six children had recent hospital admission within the past one year for the following reasons: urinary tract infection and elective admissions for imaging purposes.

Our study revealed the overall HRQOL scores as perceived by parents of children and adolescents were 77.1 and 80.5 percent respectively. Univariate analysis found that having ventriculo-peritoneal shunt, neurogenic bladder and bowel, non-ambulators and being the only child in the family were associated with lower mean score for HRQOL (Table II and III). A stepwise multivariate linear regression analysis was conducted to determine the overall combined predictive value of these independent variables for HRQOL. Presence of neurogenic bowel (β=0.375, 95%CI 0.00, 0.15) showed the largest unique and statistically significant contribution to explain the HRQOL scores (Table IV). Results of our analysis indicated that 22.1% of the variance in QOL was contributed by the presence of neurogenic bowel and being the only child in the family (R^2=0.221).

DISCUSSION

The importance of measuring QOL in spina bifida populations has been well established in the literature.[9,10] This is the first study to determine the HRQOL in children and adolescents with spina bifida in Malaysia. Our study showed that the overall HRQOL scores as perceived by parents of children and adolescents were 77.1 and 80.5% respectively. Olesen et al.,[9] studied the quality of life in a paediatric spina bifida population using the Parkin instrument, found the overall HRQOL score was 83%. In paediatric spina bifida population, one must not assume that disease symptoms necessarily correlate with QOL. Although these parents and adolescents face multiple challenges in various life domains (psychosocial, emotional, physical and environmental) and may not be at the level of achievement that either would prefer, they nonetheless view the overall QOL positively. In fact, it has been shown that children with spina bifida are not typically as bothered by their symptoms as health care providers often perceived.[11]

Our study showed clinical factors appeared to contribute a significant impact on the HRQOL. Four variables were

Table I: Socio-demographic and clinical characteristics of the study group

Characteristics	n (%)
Gender	
- Male	28 (52)
- Female	26 (48)
Ethnicity	
- Malay	30 (55.6)
- Chinese	11 (20.4)
- Indian	13 (24.1)
Age (years)	
- 5-12 years old	32 (59)
- 13-20 years old	22 (41)
Child's education level	
- Primary	11 (20)
- Secondary	20 (37)
- None/ Not applicable	10 (19)
- Special Education	13 (24)
Mother's Education level	
- Primary	11 (20)
- Secondary	28 (52)
- Tertiary	15 (28)
Father's Education level	
- Primary	8 (15)
- Secondary	28 (52)
- Tertiary	18 (33)
Monthly family income (RM)	
- 1000-1999	9 (17)
- 2000-2999	15 (28)
- 3000-3999	14 (26)
- >4000	16 (29)
Siblings with Chronic illness/ disability	
- Yes	1 (2)
- No	53 (98)
Number of siblings	
- None	12 (22)
- 1-2	35 (65)
- 3-4	6 (11)
- More than 4	1 (2)
Pathology:	
- Spina Bifida	52 (96)
- Sacral Agenesis	2 (4)
Level of lesion:	
- Thoraco-lumbar (T2-L2)	1 (2)
- Lumbar (L3-L5)	16 (30)
- Lumbo-sacral (L5-S1)	24 (44)
- Sacral (S1-S5)	13 (24)
Shunt:	
- Yes	22 (41)
- No	32 (59)
Ambulatory Status:	
- No aids	19 (35)
- With aids	19 (35)
- Wheelchair dependant	16 (30)
Neurogenic Bladder:	
- Yes	43 (80)
- No	11 (20)
Neurogenic Bowel	
- Yes	36 (67)
- No	18 (33)
Hospitalization last 1year	
- None	48 (89)
- 1 or more	6 (11)

Table II: Demographic variables associated with spinal bifida and health-related quality of life

Independent variable (value label)	n (%)	Mean percentage score	Standard deviation	t-value	P-value*
Gender (n=54)					
- Male	28 (52)	76.9	11.1		
- Female	26 (48)	80.1	13.7	-0.9	0.36
Age (n=54)					
- Children (5-12 years)	32 (59)	77.1	13.5		
- Adolescent (13-20 years)	22 (41)	80.5	10.5	-1.0	0.32
Child's education (n=44)					
- Normal stream	31 (70)	79.1	12.8		
- Special education	13 (30)	75.1	11.3	1.0	0.33
Father's education (n=54)					
- Secondary education and below	36 (67)	77.5	11.2		
- Tertiary education (Diploma/ Degree)	13 (33)	80.5	14.6	-0.8	0.45
Mother's education (n=54)					
- Secondary education and below	39 (72)	77.0	11.5		
- Tertiary education (Diploma/ Degree)	15 (28)	82.2	14.3	-1.4	0.17
Family income (n=54)					
- < RM 3000	24 (45)	75.2	10.4		
- RM 3000 or more	30 (55)	81.1	13.3	-1.8	0.071
Siblings with Chronic Illness/ Disability (n=54)					
- Yes	1 (2)	78.6			
- No	53 (98)	78.5	12.5	0.01	0.99
Single child (n=54)					
- Yes	12 (22)	71.9	11.5		
- No	42 (78)	80.3	12.1	-2.1	0.037*

*Significant P-value <0.05

Table III: Clinical variables associated with spinal bifida and health-related quality of life

Independent variable (Value label)	n (%)	Mean percentage score	Standard deviation	t-value	P-value*
Pathology:					
- Spina bifida	52 (96)	78.2	12.5		
- Sacral agenesis	2 (4)	84.5	6.9	-0.7	0.49
Level of lesion:					
- High level lesion (T2-L5)	17 (32)	76.7	11.5		
- Low level lesion (L5-S5)	37 (68)	79.3	12.9	-0.7	0.48
Shunt:					
- Yes	22 (41)	74.4	11.9		
- No	32 (59)	81.3	12.1	-2.1	0.044*
Ambulatory status:					
- Ambulating (with or without aids)	38 (70)	81.4	11.9		
- Wheelchair bound	16 (30)	71.6	10.9	-2.8	0.007*
Neurogenic Bladder:					
- Yes	43 (80)	76.7	12.5		
- No	11 (20)	85.3	9.6	-2.1	0.041*
Neurogenic Bowel:					
- Yes	36 (67)	75.0	11.6		
- No	18 (33)	85.4	11.1	-3.1	0.003*
Hospitalization last 1 year:					
- None	48 (89)	78.4	12.7		
- 1 or more	6 (11)	78.7	10.9	-0.05	0.96

*Significant P-value < 0.05

Table IV: Predictors of quality of life scores in SB patients

Variables associated with quality of life scores+	Unstandardized Coefficients		Standardized Coefficients	P- value	t-value	95% Confidence Interval	
	B	Std. Error	Beta			Lower bound	Upper bound
No of siblings	7.375	3.669	0.250	0.05*	2.01	0.037	0.17
Presence of neurogenic bowel	9.770	3.236	0.375	0.004*	3.02	0.000	0.147

*Significant P-value < 0.05
+ Stepwise linear regression analysis

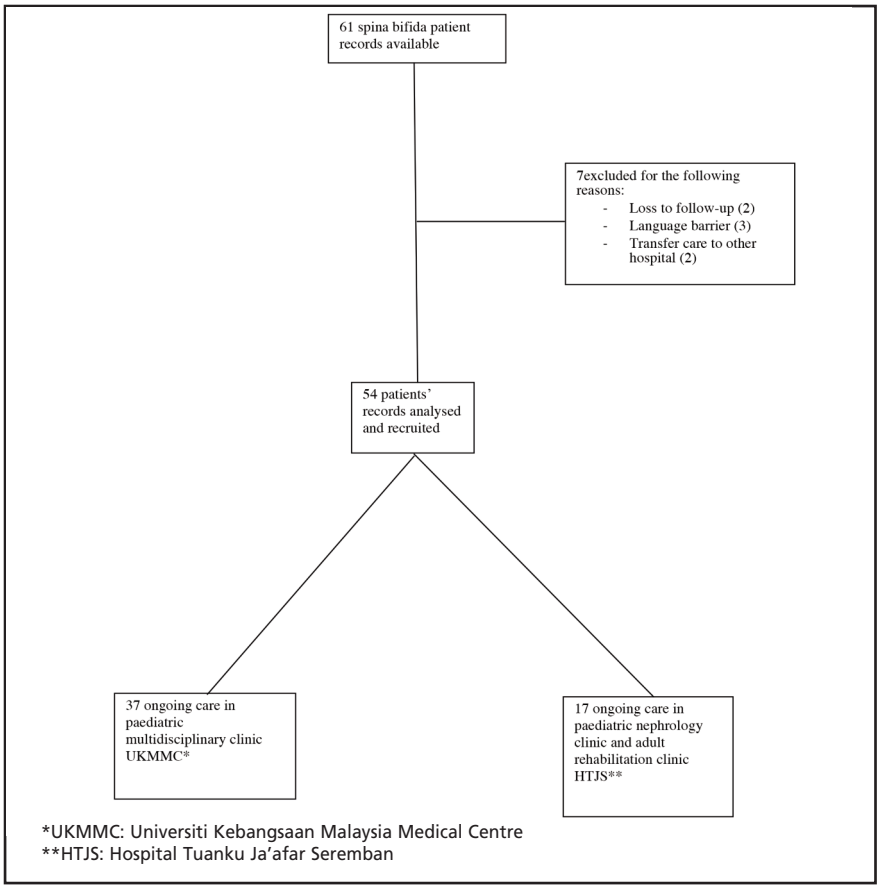

Fig. 1: Patient recruitment from UKMMC and HTJS

significantly associated with lower HRQOL scores: presence of shunt, wheelchair dependent, presence of neurogenic bladder and bowel. Being the only child in the family was the sole significant factor identified to be statistically significant in the socio-demographic variable.

Neurogenic bowel

Children with spina bifida are reported to have a high frequency of neurogenic bowel problems.[12] Hubert et al., highlight the negative impact of bowel dysfunction on QOL in a recent study.8 Clinical problems related to neurogenic bowel (constipation, pain and discomfort, faecal incontinence, anal fissures and haemorrhoids) and socio-psychological problems (low self-esteem, inappropriate and poor social interaction) cause a significant impact on social activities and QOL.[13] Faecal incontinence generates odour, impacts social relations and lowers individual self-esteem causing much distress to both parents and patients. In addition to that, parents and adolescents often encounter major difficulties finding an effective bowel management program as this aspect has often been overlooked and even neglected at times. Mattson et al., found many children and their caregivers were not aware of available treatment options for bowel management.[14] Anti-cholinergic medications used in neurogenic bladder often causes side effects like constipation, which inevitably leads to a compounded effect on the existing bowel problems.[15] Most of our patients were using enema and laxatives as a method of active bowel management. Recent studies supported the evidence of transanal irrigation in spina bifida patients with bowel dysfunction with a success rate reported to be more than 80%.[16,17] Thus, more attention and time should be paid to managing individual bowel problems for our spina bifida patients with an overall aim to achieve regular bowel emptying and continence.

Single child in the family

Although only 22% of our patients were the only child in the family, this factor contributed significant lower HRQOL scores. Prior to this study, the authors had thought that being a single child in the family was linked to less parenting stress scores and focused care from the caregiver or parents, which ultimately results in an overall improvement in HRQOL of spina bifida individuals. Interestingly, the mediating effect of sibling support was an important finding in our study which has thus led to a change in the current perception among medical personnel and society. Perhaps, when strong sibling bonds are built and sustained, reciprocity between siblings will eventually foster an expression of trust, understanding, affection and respect. Relationship quality and the amount of care giving assistance shared between siblings which may lead to better communication efficacy, higher satisfaction within family members, better coping strategies and positive attitude towards spina bifida. This is likely to correlate with fewer psychological symptoms and stress in both parents and children with spina bifida. As parents grow older, having

siblings who are willing to take on a care-giving role is important as many adolescents and adults with spina bifida may eventually turn to their siblings for help in times of need, and this is especially so in our cultural context. Loneliness, lack of physical and emotional support will contribute to lower HRQOL scores in patients who are the only child in the family. Therefore, our findings provide evidence of saliency with regards to sibling relationship in the HRQOL of individuals with SB. For those single child families, health professionals should encourage parents to have another child. With the current recommendations of pre-conception folic acid supplementation, parents should be informed that the risk of recurrence in the future pregnancy is reduced by 72%.[18]

Neurogenic bladder

Toileting problems, nocturnal and daytime urinary incontinence brings about significant social and psychological impact to both patients and parents. Management of neurogenic bladder can represent one of the largest challenges for spina bifida patients. In adolescence, urinary incontinence appears to be the most significant factor associated with a decline in both physical and emotional QOL.[7] The introduction of urinary catheterisation, anticholinergic medications and judicious use of bladder augmentation have resulted in low rates of renal insufficiency.[19,20] However, negative impacts of this treatment on the socio-psychological aspects and schooling remained significant. Kanaheswari et al., studied the predictors of parenting stress in mothers with spina bifida, and found that the need for clean intermittent catheterisation was the only medical factor associated with parenting stress in mothers of children with spina bifida.[21] Responsibility of catheterisation inadvertently falls on the parents or caregiver most of the time. Pain inflicted during insertion of catheter and reluctance of the child to adhere to regular intervals of 4-6 hourly CIC maybe a contributing factor to stress in both parents and spina bifida patients, resulting in an overall lower QOL.

Shunt

In our cohort, presence of ventriculo-peritoneal shunt was associated with lower QOL scores. Metcafe et al., too showed that presence of shunts were associated with greater degrees of incontinence, greater urinary symptoms and an associated decrease in QOL.[19] Long term impact on intellectual disability and neuro-cognitive function can be determined by the presence of shunt and its possible related complications (e.g., shunt dysfunction and infection causing ventriculitis). Hunt et al., who studied the relation between shunted status and outcome, concluded that revisions of shunt, particularly after two years of age were associated with poor long term achievement in adults with spina bifida.[22]

Ambulatory status

The relationship between independent ambulation and QOL in spina bifida is well documented. We grouped our patients into non-ambulators (completely wheelchair-dependent or walking in therapeutic situations) and functional ambulators (household or community walkers). Our study found non-ambulators had significantly lower HRQOL, a finding which is consistent with other documented literature. Schoenmakers et al., found that good muscle strength, mental ability and being independent in mobility appeared to be much more important for daily function and QOL than other medical indicators.[7] Community ambulation is important to enable enrolment to normal mainstream schooling and participation in outdoor activities. Being home bound is often associated with long term physical and psychological impact on children.[23,24] Contrasting management facilities and resources available in other developed countries has resulted in a tremendous improvement and outcome in the ambulatory status of spina bifida patients. Our results speak the importance of planning rehabilitation program for our cohort of spina bifida patients with regards to ambulation. Methods to improve orthopaedic procedures, physiotherapy programs, obtaining high quality orthoses should be prioritised to enable our spina bifida children to achieve early ambulation.

LIMITATIONS

Firstly, our study sample was moderate and we recruited patients from two tertiary centres in the urban cities of Malaysia. Taking into consideration rare conditions like spina bifida, it is difficult to obtain a large cohort of patients. There is possible measurement bias as the assessment of QOL was performed through administration of questionnaire by one single health-care visit. This may not be reflective of the actual quality of life measurement and ideally a repeat measure should have been taken.

Secondly, the questionnaire used was not cross-culturally adapted and thus, some questions may not be applicable to our cohort of patients.

Finally, our Bahasa Malaysia (BM) version was not validated. Majority of our patients were Malays who preferred to answer the questionnaire in BM version. Therefore, there is a need for a reliable and validated BM version specifically designed to accommodate our cohort of patients in Malaysia.

CONCLUSION

Although various socio-demographic and medical characteristics are known to be associated with QOL, we collectively conclude that medical factors appeared to contribute more to HRQOL in our spina bifida population in Malaysia. Bowel management in particular was an important influence in our cohort and thus, emphasis should be paid to bowel dysfunction. Nonetheless, sibling support is also an important finding in the HRQOL and factors associated with and arising from being the only child in the family need to be explored further. With this knowledge we can then recommend appropriate intervention and management strategies for our patients, with an overall aim to optimize QOL among those with spina bifida.

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COMPLIANCE WITH ETHICAL STANDARDS

This study was funded and approved by Ethics and Research Committee Universiti Kebangsaan Malaysia Medical Centre (FF-2013-457). Informed consent was obtained from all individual participants included in this study. All applicable institutional regulations concerning ethical use of human volunteers were followed during the period of this research. The authors declare no conflict of interest.

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