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Health Outcomes among Youths and Adults with Spina Bifida

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Objective To describe the health and health-related quality of life (HR-QoL) outcomes of youths and young adults with spina bifida.

Study design One global rating of self-rated health and 2 generic measures of HR-QoL were administered to a group of youths and young adults with spina bifida. HR-QoL was measured using the Health Utilities Index Mark 3 (HUI_3) and the Assessment of Quality of Life version 1 (AQoL).

Results Data was obtained from 40 youth (mean age 16.0 years) and 13 young adults (mean age 26.6 years). Most youth rated their overall health as either excellent or very good (65%) compared with fewer adults (23%) ($P = .007$). The mean HR-QoL scores for youths versus adults were 0.57 versus 0.36 ($P = .03$) for the HUI_3 and 0.37 versus 0.25 for the AQoL ($P = .09$). HUI_3 and AQoL scores were correlated with level of anatomic lesion (rho = 0.64 and rho = 0.42, respectively).

Conclusions The HR-QoL of youths and young adults with spina bifida was low on measures that are aggregated using societal values (the HUI_3 and AQoL). This is in contrast to their single global self-ratings of health, which were more favorable. These findings underscore the distinction between ratings of HR-QoL based on societal values versus the personal lived experiences of adults with childhood-onset disability. *(J Pediatr 2012;■:■-■)*.

See editorial, p •••

Spina bifida occurs in approximately 20/100 000 live births in North America,[1] and results in complex physical disability affecting the brain, spinal cord, bowel, bladder, and sensation. The care and survival of children with spina bifida has improved markedly, and approximately 80% now reach adulthood.[2] As these children age, information on outcomes is increasingly important.[3] Health-related quality of life (HR-QoL) is recognized as an important outcome and is arguably the most salient from the perspective of patients.[4] Data on the HR-QoL of people with spina bifida has emerged in the past decade.[5-11] The most detailed HR-QoL data for people with spina bifida is presented in 3 key papers that explore the impact of 1 or more key factors.[8-10]

In 2002, Sawin et al reported on 60 American youth with spina bifida (12-21 years) and showed mean Spina Bifida HR-QoL to be 4.0, parent ratings were similar to youths self-report, and neither age nor lesion level was related to spina bifida HR-QoL scores.[8] In 2007, Verhoef et al reported on 179 Dutch young adults with spina bifida (16-25 years) compared with the general population using the Short Form 36-item health survey (SF-36).[9] They found lower scores in physical functioning (mean = 46.1 for spina bifida sample vs 94.2 for the general population), which were inversely related to the lesion level.[9] In 2009, Buffart et al reported on 51 Dutch young adults with myelomeningocele (16-30 years) and described the impact of factors on SF-36 scores.[10] The mean Physical Component Summary score was 43.1 (SD 9.0) and the mean Mental Component Summary score was 55.0 (SD 9.7). Better SF-36 scores were related to age, sex (male), and lesion level (ambulators) based on logistic regressions.[10]

In summary, the picture of health and HR-QoL outcomes is becoming clearer. The changes in HR-QoL that may be expected as youths enter adulthood is not yet clear, nor is the impact of sex. However, lesion level appears to be a key factor explaining HR-QoL outcomes. This article provides a comparative data on the health and HR-QoL outcomes of youths and young adults with spina bifida and explores the relationship of age, sex, and lesion level on HR-QoL.

Methods

Participants in this study were recruited as part of a larger descriptive study that was representative of the graduates from children's rehabilitation centers in the province of Ontario, Canada. The study

AQoL	Assessment of Quality of Life
CP	Cerebral palsy
CTC	Children's Treatment Center
HAQ	Health Assessment Questionnaire
HR-QoL	Health-related quality of life
HUI_3	Health Utilities Index Mark 3
SF-36	36-Item Short Form Health Survey
SRH	Self-rated health

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was approved by the Research Ethics Boards at the lead institutions and the participating rehabilitation centers. Participants in the larger study included youths (13.0-17.9 years of age) and young adults (23.0-32.9 years of age) who had either acquired brain injury, cerebral palsy (CP), or spina bifida.[12] The quality of life outcomes for the youths and adults with CP have been previously reported,[13] as have the results from interviews with some of these participants.[14]

Potential survey participants were identified by reviewing all health records for clients with a diagnosis of spina bifida, at 6 Children's Treatment Centers (CTCs) in Ontario, Canada. In this region, CTCs are the primary source of children's rehabilitation services and are regionally organized. The vast majority of children with spina bifida receive services from a CTC, with all clinical services paid for through provincial health insurance. Thus, this was a representative sample of the population. Centers are located in urban communities and provide outreach services to northern and rural regions. Participants were recruited by a mail invitation sent from the CTCs. Severity information was obtained from chart review and reflected the lesion level noted at the time of surgical closure. We classified respondents' lesion level into 4 categories: thoracic, high-lumbar (L1-L3), low-lumbar (L4-L5), and sacral. These categories are based on Molnar's classification.[15]

Health Outcomes

The primary outcomes were health and HR-QoL. Health is defined by the World Health Organization's *International Classification of Functioning, Disability, and Health*[16] and goes beyond the presence or absence of disease to include information on the nature and extent of health limitations. HR-QoL is defined as a multidimensional construct that refers to the specific impact of health conditions on physical, mental, emotional, and social well-being as perceived by the person experiencing the health condition.[17,18] Information on health was gathered using a single self-rated health (SRH) question that has been used in many population health surveys: In general, would you say your health is excellent, very good, good, fair, or poor?

HR-QoL was measured using 2 generic scales: the Health Utilities Index Mark 3 (HUI_3)[19,20] and the Assessment of Quality of Life version 1 (AQoL).[21] These scales were chosen because of their strong psychometric properties[21-24] and ability to span the age range of 13-33 years. There were no disease-specific measures that met both of these criteria.[5] The HUI_3 is a multi-attribute health classification system that is valid and reliable[19] for both youth and adult populations. We used the 15-item version that describes 8 attributes or domains of health.[20] The aggregate scores for the HUI_3 have a mathematical minimum of -0.341 and a maximum of 1.0. The AQoL is a 15-item HR-QoL questionnaire that has 5 scales or domains of health and strong psychometric properties.[21,24] The aggregate scores for the AQoL have a mathematical minimum of 0.0 and a maximum of 1.0 and were used to provide additional information on the HR-QoL. The Health Assessment Questionnaire (HAQ) is a 20-item measure of physical function that was selected as secondary outcome to compliment the health and HR-QoL data.[25] HAQ scores range from 0-3. However, we rescaled the HAQ to a 0-1 scale, similar to the HUI_3 and AQoL, to simplify interpretation. Thus, 1.0 indicates the best outcome for all measures used.

All health and HR-QoL outcomes data were collected by mail-administered survey, according to the Dillman method,[26] with reminder post-cards and repeat mailings at 3 and 6 weeks. Participants were encouraged to self-report if possible but were permitted to have assistance to complete the questionnaires or have it completed by a primary care giver (proxy-report) if necessary. Additional details on the methods of this study have been previously reported.[12,27]

Statistical Analyses

All analyses were performed using Stata v. 11 (StataCorp, College Station, Texas). We generated frequency distributions by age group for lesion level and SRH scores. The internal consistency of the HUI_3 and the AQoL were assessed using Cronbach alpha and their summary scores were computed using the multi-attribute utility functions specified by the developers. Means and SDs were computed by age group for the HUI_3 and AQoL. The relationship between the HUI_3 and AQoL was assessed using Pearson correlation coefficient, and their relationship to SRH was assessed using Spearman rho. We also prepared box plots of the HUI_3 and AQoL domain scores by age group for each of these measures. Similar analyses were done on the HAQ to put the HR-QoL scores into context relative to physical function scores.

We hypothesized a priori that health scores would be lower in young adults than in youths. We compared the proportion of youths versus adults who reported their SRH as "excellent" or "very good" using a 2-sample test of proportions. We then compared youth and adult summary scores for the HUI_3 and AQoL using unpaired t tests. Subsequent exploratory analyses were conducted using logistic regression for SRH. Note that the SRH variable was converted to a binary variable, to facilitate logistic regression, by combining the excellent and very good responses into 1 category and combining the good, fair, or poor responses into a second category. Exploratory analyses were conducted by linear regression for the HUI_3 and AQoL (with all variables entered concurrently) to determine the importance of 3 key factors identified from the literature: age (as a continuous variable),[9,10] sex (a binary variable),[9,10] and lesion level (4 categories based on anatomic lesion level).[8-11,28-30]

Note that because we expected to obtain self-report data for some participants and proxy-report for others, we specified a priori that we would merge these data to enable the inclusion of full spectrum of severity. We conducted a sensitivity analysis of the key findings to assess the impact of this decision on our conclusions.

Results

A group of 210 youths and 145 young adults with spina bifida were identified from 6 CTCs in Ontario. Address information was not available for 63 of the 355 members of the target population. Thus, information packages were sent out to 292 potential participants and 116 responded (39.7% response rate). Of these 116 respondents, 61 (52.6%) consented to participate. Questionnaire packages were sent to all of these followed by reminder postcards as well as phone follow-up when questionnaires were not received back.

Questionnaires were received from 40 youths and 13 adult participants (overall survey completion rate of 87%). The mean ages were 16.0 (SD = 1.30; range 13.0-17.9) years among youths and 26.6 (SD = 3.10; range 23.0-32.9) years among adults. Of the responding group, 65% of the youths and 77% of the adults were female. The breakdown in terms of severity was based on the anatomic lesion level reported in the clinical chart. The severity distribution of the youth group was 25% sacral, 30% low-lumbar (L3-5), 18% high-lumbar, and 28% thoracic. The severity breakdown in the adult group was 15% sacral, 31% low-lumbar (L3-5), 23% high-lumbar, and 15% thoracic. Note that 2 of the adults did not have a lesion level recorded on their chart (both had survey responses that suggest they may belong to the thoracic group).

In our sample, 29 (55%) of the participants completed their survey by self-report, with an additional 16 (30%) completing it by self-report with assistance. Eight (15%) surveys were completed by a proxy-respondent. There is some inconsistency in the literature about the impact of respondent on the measurement of HR-QoL.[31] In our study, parental report was infrequent (n = 8), thus, we have presented the results together.

SRH

From this sample, we generated a description of the health of youths and adults who have spina bifida as measured by SRH. The responses to the SRH question showed a wide distribution. Overall, 54.7% reported their health to be “excellent” or “very good.” However, 65% of youths reported “excellent” or “very good” health compared with 23% of young adults. This difference was statistically significant ($P = .007$). Furthermore, 30% of youths rated their health as “good” and 5% as either “fair” or “poor.” Among adults, 38.5% rated their health as “good” and 38.5% as either “fair” or “poor.”

HR-QoL

We also generated a description of HR-QoL as measured by the HUI_3 and AQoL. The internal consistency of these measures in this sample was 0.71 and 0.75, respectively. The mean HUI_3 score for our combined sample was 0.52 (SD = 0.28). The mean AQoL score was 0.34 (SD = 0.24). The average HUI_3 scores by age group were 0.577 (SD = 0.270) for youths and 0.36 (SD = 0.27) for adults. The average AQoL scores by age group were 0.37 (SD = 0.26) for youths and 0.25 (SD = 0.17) for adults. The adult scores were lower than scores for the youths. This difference was statistically significant based on an unpaired *t* test of the HUI_3 (mean difference of 0.22, $P = .016$). Although, the same trend was observed for the AQoL, the difference did not reach statistical significance (mean difference of 0.13, $P = .096$). The means and SDs for the HUI_3 and AQoL scores were calculated by lesion level (Table I).

Table I. QoL scores by lesion level

Lesion level	Subsample size*	HUI_3		AQoL	
		Mean score	SD	Mean score	SD
Thoracic	13	0.29	0.14	0.22	0.14
High-lumbar	10	0.44	0.31	0.28	0.23
Low-lumbar	16	0.63	0.23	0.39	0.21
Sacral	12	0.76	0.20	0.51	0.30
Unknown	2	0.22	0.02	0.09	0.04

QoL, quality of life

*Youth and adult age groups were combined due to small cell sizes.

We explored the impact of sex on HR-QoL scores, and found a slight trend toward higher HR-QoL among females in both the HUI_3 (0.05 points higher) and the AQoL (0.08 points higher), but these differences were small and nonsignificant (HUI_3 $P = .515$ and AQoL $P = .283$).

The HUI_3 and AQoL domain scores were explored visually. The 8 domains of the HUI_3 and 5 domains of the AQoL were graphed by age group (Figures 1 and 2). It is important to note that the physical ability domain of the AQoL is based on vision, speech, and hearing, thus, it is not surprising to find many perfect scores in this domain in a sample with spina bifida. Mobility limitations are captured within the independent living domain of the AQoL. Thus, the AQoL’s independent living domain shows a similar distribution to that seen on the HUI_3’s ambulation domain.

The box plots in Figures 1 and 2 confirm that the overall summary score distributions are lower among the young adults, and that this is due to some subtle differences in domains. Most notably, HUI_3 scores were lower among the adults in the domains of vision (youth mean = 0.98 vs adult mean = 0.90), ambulation (0.47 vs 0.30), and pain (0.93 vs 0.74). The greatest difference in AQoL scores between youths and young adults occurred in the domains of illness (youth mean = 0.42 vs adult mean = 0.24) and psychological well-being (0.93 vs 0.84). What may not be apparent on the box plots is that none of the domain scores were higher in the adult group when compared with the youth group.

To put these HR-QoL scores into context, we computed the mean physical function scores based on the HAQ. Mean HAQ scores were 0.65 (SD 0.29) for youths and 0.56 (SD 0.30) for adults. These scores indicate that our sample had significant physical impairments ($P < .0001$ in comparison with a population sample).[32] The HAQ scores were slightly worse in the adult group.

The relationships between the main outcomes were examined using correlations. SRH was moderately correlated with the HUI_3 (rho = −0.45, $P < .001$), the AQoL (rho = −0.58, $P < .001$) and the HAQ (rho = −0.56, $P < .001$). The HUI_3 was strongly correlated with the AQoL (r = 0.73, $P < .001$). The HUI_3 was also strongly correlated with the HAQ (r = 0.79, $P < .001$). The AQoL also had a strong correlation with the HAQ (r = 0.70, $P < .001$). These correlations suggest

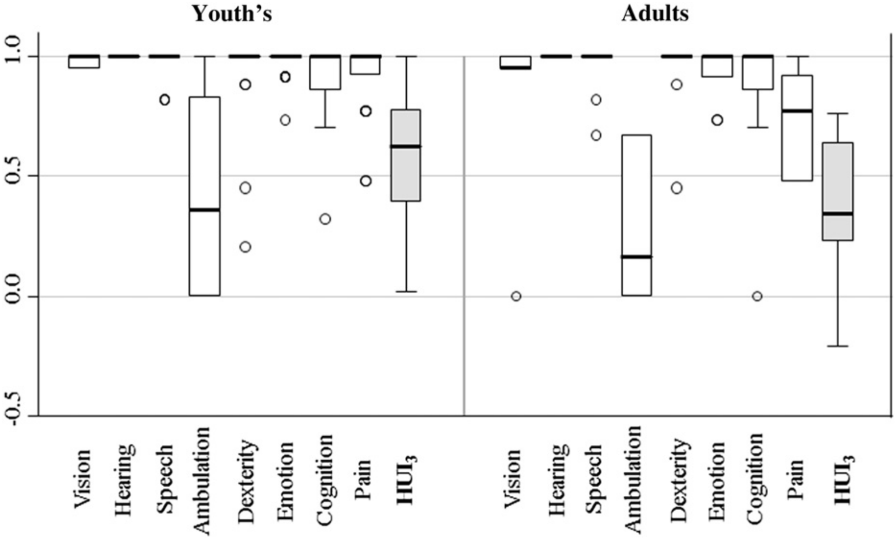

Figure 1. HUI_3 domain scores by age group.

that the overall concepts measured by the HUI_3 and AQoL are consistent (ie, both measure HR-QoL) and that in this population, HR-QoL is strongly associated with physical function (as measured by the HAQ). However, this is not unexpected, given that physical disability is a characteristic that all of the participants share. Figures 1 and 2 provide more detailed information on the relative contribution of the various domains within HR-QoL. It is important to note that some of the domains measured by the HUI_3 and AQoL are different, and thus, each measure provides unique information.

Exploration of the Impact of Contributing Factors

We were particularly interested in understanding the relative contribution of various factors to health status and HR-QoL in youth and adulthood for this population. The only candidate factor that was a strong predictor of excellent or very good SRH, using logistic regression, was age (OR: 0.80, $P = .007$), with younger participants having better SRH.

Linear regression analysis showed that the most important single factor contributing to HR-QoL outcomes was the surgical lesion level at birth, which was responsible for 40% of the variance in the HUI_3 scores and 18% of variance in the AQoL scores. Because of the importance of lesion level in determining HR-QoL, the mean scores for the HUI_3 and AQoL are presented by lesion level in Table I. When age and sex were added to this regression model, we found that both lesion level and age were important, and together they explained 48% of the variance in HUI_3 scores and 22% of the variance in AQoL scores (Table II).

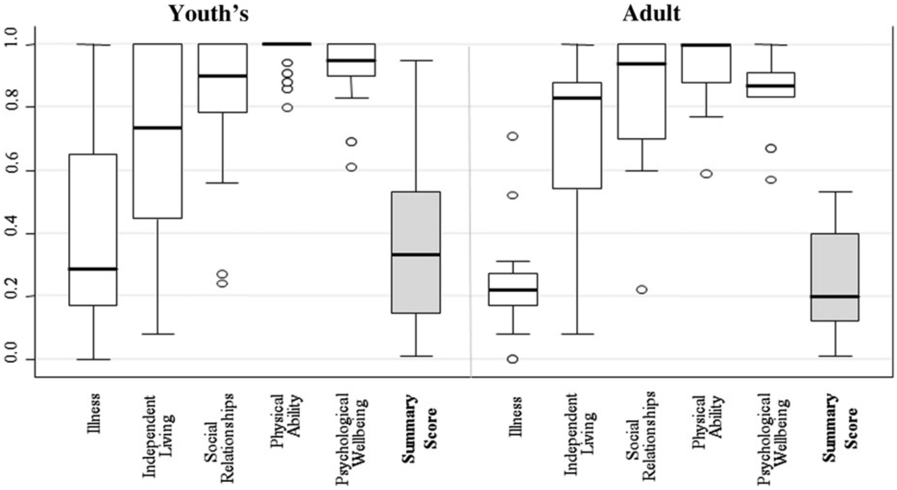

Figure 2. AQoL domain scores by age group.

Table II. Factors associated with QoL outcomes

	HUI_3		AQoL	
Factor	**Coefficient**	**Significance**	**Coefficient**	**Significance**
Age	−0.018	$P = .003$	−0.023	$P = .042$
Sex (female)	0.017	$P = .789$	0.062	$P = .350$
Lesion level				
Thoracic	Reference group		Reference group	
High-lumbar	0.014	$P = .106$	0.051	$P = .569$
Low-lumbar	0.319	$P < .001$	0.151	$P = .063$
Sacral	0.451	$P < .001$	0.274	$P = .002$
Constant	0.629	$P < .001$	0.422	$P = .002$
% of variance explained	48%		22%	

Sensitivity analyses were completed to examine the impact of the 8 participants whose data came from proxy-reports. These analyses determined that although the mean scores were slightly higher in the self-report group (HUI_3 mean + 0.04, AQoL mean + 0.03, HAQ mean + 0.04), the conclusions based on the comparison of youth and adult data and the regression models remained unchanged.

Discussion

The previous literature describes average HR-QoL scores predominantly based on data from youths. The 3 papers that present the most detail on HR-QoL were those led by Verhoef,[9] Buffart,[10] and Sawin[8] who presented mean HR-QoL scores of 0.46, 0.86, and 0.75, respectively, when rescaled to a 0-1 score. Our results and those of others[9,29] have demonstrated the importance of lesion level in determining HR-QoL scores. Detailed mean scores by lesion level were not presented in previous papers. This article presents information on the HR-QoL of both youths and young adults with spina bifida, based on a representative Canadian sample of 40 youths and 13 young adults. This sample size is small in the context of the medical literature but is similar to other health survey reports in the spina bifida population. Interpretation of the results must take into account that the sample of adults was small.

QoL in persons with spina bifida is known to be influenced by lesion level,[9,29] thus, it is important to begin by reviewing the distribution according to lesion level. In total, 44% of our sample had thoracic or high-lumbar lesions. This proportion is almost identical to Verhoef et al (43%)[9] but much higher than that of Sawin et al (29%)[8] and Buffart et al (16%).[10] Therefore, the mean scores from our sample would be expected to most closely compare with the mean from Verhoef et al.[9] Our reported HUI_3 mean score for the combined sample of 53 participants was 0.52, and the AQoL mean score in our sample was 0.34. Our results compare favorably with Verhoef's combined sample mean of 0.46 on the physical functioning domain of the SF-36.[9] As expected, the mean physical function domain scores from the Dutch[10] and American[8] populations were higher than those reported here because those samples had fewer participants with high level lesions.

We have documented the means and SDs of HR-QoL scores for both youths and young adults with spina bifida, by lesion level, using 2 different metrics: the HUI_3 and the AQoL. We have shown that the greatest deficits are in the domains of ambulation, pain, and cognition on the HUI_3 and the domains of illness, independent living, and social relationships on the AQoL. The literature suggested that the greatest deficits were in the physical function domain (Buffart 2009) and that HR-QoL outcomes were associated with lesion level (Verhoef 2007). Thus, our article confirmed these findings.

By comparing youths and adults, we identified a statistically significant difference in overall scores for the HUI_3 but not the AQoL. The differences in HR-QoL scores between the youths and young adults were small on both measures, suggesting that the decline in HR-QoL is slower than might be expected based on the Liptak's report of changes in global health ratings over a 4-year period. This is a very positive sign. Our exploration of domain scores showed worse scores among the adults on the ambulation and pain domains of the HUI_3 and the illness domain of the AQoL. These findings support the need for continuing medical care and respite for individuals with complex childhood acquired conditions such as spina bifida as they enter adulthood.

Our results identify a clear discrepancy between SRH and HR-QoL scores. The main differences are the level of detail reported and the values that were implicit in the scores. The SRH scores were based on a single question and used the respondents' values. These were higher than the HR-QoL scores that were based on several questions and scored using societal values. These findings provide new information on how global health ratings differ from HR-QoL measures and may inform us regarding the distinction between HR-QoL and health in this population. The SRH scores from this population were similar to those previously reported for a sample of youths and young adults with CP as part of the larger study.[13] Related qualitative interviews with some participants from the spina bifida sample suggest that their SRH scores reflect acute health issues more than their underlying chronic condition, which was viewed as part of their identity (eg, given that I have spina bifida, my health is good). This result may reflect the impact of living with a condition throughout their lifespan, during which they have become accustomed to their way of functioning. Thus, their overall impression (ie, SRH) may be more positive than an outcome measured by societal values (ie, HUI_3 or AQoL). Finally, this work presents a detailed statistical model of the factors contributing to HR-QoL among a combined sample of youths and young adults with spina bifida. The literature suggested the important factors were age, sex, and lesion level. We have confirmed the impact of age and level of lesion empirically and have shown that approximately 48% of the variation in HUI_3 scores can be explained by lesion level and age in combination, but only 22% of the variance in AQoL can be explained. The difference is likely due, in part, to the fact that the HUI_3 is symptom driven and linked to the underlying level of pathology. Further evidence to support this conclusion comes from the finding that the HUI_3 and HAQ are very highly correlated. Age and lesion level were also the best predictors for 'very good' or 'excellent' SRH (48% of variance), suggesting that the information on lesion level in infancy provides valuable information to youths regarding what they can expect as they age. We found no significant relationships related to sex. However, there remains a large portion of HR-QoL that remains unexplained. There are many other individual factors that contribute to HR-QoL and should be explored in future research.

In conclusion, this study has enriched our understanding of HR-QoL in youths and young adults with spina bifida, based on a representative Canadian cohort, under a system of universal access to health services. This article may form the basis for future research focusing on ways to improve outcomes in spina bifida over time. ■

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