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CME Objectives:
On completion of this article, the reader should be able to: (1) List factors that influence quality-of-life in emerging adults with spina bifida; (2) Describe how severity of spina bifida relates to quality-of-life; (3) Discuss areas of potential intervention to protect against deterioration in quality-of-life in patients with spina bifida.

Level: Advanced

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Family Satisfaction, Pain, and Quality-of-Life in Emerging Adults with Spina Bifida

A Longitudinal Analysis

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ABSTRACT

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Objective: This study uses the Life Course Model for Spina Bifida (SB) to advance knowledge of factors associated with change in quality-of-life (QOL) among emerging adults with SB.

Design: Forty-eight participants (mean [SD], 22.04 [2.16] yrs) completed self-report questionnaires at two time points, 15 mos apart. Four QOL domains (physical health, psychological, social relationships, and environment) were measured using the World Health Organization QOL–BREF version. SB clinical data were collected via chart reviews. Paired *t* tests and reliable change indices evaluated group- and individual-level QOL change, respectively. Multiple regression analyses tested the contributions of the Life Course variables in explaining change in QOL over time.

Results: No significant group-level differences in the QOL domains were found between time 1 and time 2, but there was substantial individual variation in QOL over time. SB severity was related to a decline only in psychological QOL ($B = -0.68$, $P = 0.02$). Increased pain was associated with reduced physical health ($B = -0.29$, $P = 0.049$) and psychological ($B = -0.29$, $P = 0.03$) QOL at time 2, whereas greater family satisfaction was related to improved QOL in several domains.

Conclusions: Clinicians should be aware of the negative impact of pain and the protective influence of family satisfaction on QOL in emerging adults with SB.

Key Words: Quality-of-Life, Spina Bifida, Young Adults, Family Satisfaction, Pain

Disclosures:

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Advancements in health care and interdisciplinary management of spina bifida (SB), a congenital birth defect with multisystem involvement, have resulted in an unprecedented number of affected individuals surviving into adulthood.[1] The increased life expectancy has created new challenges to promote health functioning and quality-of-life (QOL) across the life span.[2,3] Young adults with SB experience a multitude of health issues requiring coordinated medical and rehabilitation management, including shunt malfunction, syringomyelia, tethered cord syndrome, pyelonephritis, lymphedema, pressure ulcers, obesity, hypertension, and musculoskeletal pain.[4,5] A systematic review of health problems encountered by adults with SB highlighted additional health concerns stemming from the longitudinal effects of hydrocephalus, neuromuscular weakness, neurogenic bowel and bladder, and bone and joint deformity.[6] Young adults with SB also encounter difficulties with full community participation, as evidenced by poor rates of employment and independent living[7] and restricted peer interactions.[8,9] Collectively, the ongoing health problems and deficits in psychosocial functioning may place young adults with SB at risk for impaired QOL.

Verhoef and colleagues[10] observed significant differences in QOL scores between young adults with SB and typically developing comparison peers, with the SB group reporting lower levels of QOL across six of eight domains. Buffart et al.[11] similarly found a majority of individuals with SB (mean age, 21 yrs) to endorse low physical QOL and experience difficulties in performing daily life activities. Further, young adults with SB report lower health-related QOL than do peers with cerebral palsy or an acquired brain intjury.[12] Other findings refute an elevated risk status for this group, with self-rated QOL scores falling within the range of the general population[13] or even higher than their peers.[14] Research findings for the influence of SB impairment indicators on QOL are likewise inconsistent. Some studies suggest that a higher lesion level[15] and mobility limitations[16,17] predispose individuals with SB to reduced QOL, whereas other data fail to support a significant effect of lesion level,[18] hydrocephalus,[19] or mobility status[20] on QOL. The inconsistency of QOL findings in SB research may, in part, be caused by the use of different study methods (e.g., measures of QOL) and samples, has been an ongoing puzzle for researchers and clinicians alike, and underscores the need to clarify contributing variables to help structure and direct intervention efforts.

The Life Course Model for SB, developed by the National Spina Bifida Program at the Centers for Disease Control and Prevention and in collaboration with interdisciplinary SB experts, proposes that three functional domains contribute to positive QOL outcomes in this population.[21] The self-management and health domain pertains to developing independence in SB management activities including the prevention of secondary conditions (e.g., pain). The social and personal relationships domain delineates how social competence with peers and healthy family functioning promote QOL, whereas the education and income support component suggests that prevocational skill development and employment for income security enhance QOL outcomes. Consistent with the definition of QOL advanced by the World Health Organization,[22] the Life Course Model for SB treats QOL as a broad construct that extends beyond condition symptoms to include physical, mental, and social well-being.

To date, research on correlates of QOL has primarily focused on the impairment indicators of SB (e.g., lesion level and hydrocephalus), but empirical support is building for the relationships proposed by the Life Course Model for SB. For example, in a small sample of adolescents with myelomeningocle, youths who were more independent in daily skills endorsed higher QOL.[23] The importance of healthy family functioning in promoting QOL in this population has also been described. Sawin and colleagues[24] found greater satisfaction with family functioning to predict higher QOL scores among adolescents and young adults with SB. A protective influence of adaptive family functioning on QOL was similarly observed in a sample of adolescents with mobility impairment including SB.[25]

Finally, the associations between self-reported physical pain and poor QOL are especially robust. In an assessment of health problems in youths and young adults with SB, chronic pain was associated with significantly lower QOL scores,[14] and Verhoef and colleagues,[10] using the 36-item Short-Form Health Survey measure of QOL, specifically identified pain as a risk factor of poor social functioning and general vitality. The experience of pain in adolescents and young adults has also been shown to interfere with school, job, and recreation activities[26] and to be

associated with psychologic distress in this population.[27] In summary, although measuring and supporting QOL in individuals with SB are acknowledged as an important clinical outcome,[3] modifiable risk and protective influences on QOL in emerging adults are an understudied area, and longitudinal data for this population are especially limited. This exploratory longitudinal study aimed to advance knowledge of QOL in emerging adults with SB and to identify the contributions of select variables from the Life Course Model for SB (self-management, SB severity, pain, family satisfaction, and employment status) in explaining change in QOL. Specifically, it was hypothesized that employment and increases in self-management and satisfaction with family functioning from time 1 to time 2 would be associated with improved QOL over time but that greater pain and a higher degree of SB severity would be related to decreased QOL from time 1 to time 2.

METHODS

Participants

Participants were part of a larger investigation of health outcomes, healthcare use, and psychosocial adaptation in emerging adults with SB.[16,27] The data presented in this study focus on QOL outcomes collected at two time points, approximately 15 mos apart. Sixty-one participants were originally enrolled in this study, which drew its sample from five SB clinic sites across the United States. Of these geographically diverse sites, three operate from a life span approach and provide services to individuals with SB from birth through adulthood, whereas two serve only adults with SB. Eligibility criteria included a primary diagnosis of SB; being 18–25 yrs of age, based on current theory of emerging adulthood[28]; and documented capacity to understand the study measures, as assessed by the MacArthur Competence Assessment Tool (see study by Bellin et al.[27]).

The study procedures were reviewed and approved by institutional review boards at each participating site and by the Professional Advisory Council of the Spina Bifida Association. Subsequently, the participants were recruited through face-to-face contact during SB clinic visits and by mailed invitations that included an opt-out postcard declining contact by the research team. Consent was obtained from interested individuals who subsequently completed a self-report questionnaire inclusive of demographic questions and standardized measures of satisfaction with family functioning, self-management, and QOL, described below. The study staff completed a chart review to record clinical data such as lesion level and history of shunted hydrocephalus. The participants received a $40 gift card as compensation for their time completing the time 2 questionnaires, which took approximately 45 mins to complete.

Of the original 168 eligible individuals who received recruitment materials, 64 (38%) agreed to study enrollment. Face-to-face recruitment yielded a higher enrollment rate than did mailed invitations (76% *vs.* 29%). The discrepancy reflects challenges in reaching the emerging adults by telephone, as well as a potential impact of executive functioning difficulties that impede follow-through.[29] Three failed the competency screening, resulting in the final time 1 sample of 61 emerging adults with SB. Data collection for time 1 and time 2 occurred approximately 15 mos apart for 48 of the original 61 participants (79% retention rate). Three participants withdrew from this study because of the length of the procedures involved with data collection, and the others either moved or were lost to follow-up. Attrition analysis revealed no significant differences in SB clinical features, key demographics, or study outcomes between the individuals who remained in this study through time 2 *vs.* those who completed time 1 only.

Measures

SB Clinical Factors: SB Severity and Pain

The small sample size limited the number of SB impairment indicators entering the regression models. To maximize power for the proposed analysis, an SB severity index was created using the guidelines established by Hommeyer et al.[30]: (1) shunt status (no, 1; yes, 2), (2) myelomeningocele (no, 1; yes, 2), (3) lesion level (sacral, 1; lumbar, 2; thoracic, 3), and (4) ambulation status (no assistance, 1; needs assistive devices to walk, 2; wheelchair use, 3). Scores range from 4 to 10, with higher scores reflecting greater severity. A significant association between the SB severity index and health professionals' rating of SB severity ($r = 0.60$, $P < 0.001$) supports its validity (Hommeyer et al.,[30] 1999). Internal consistency of the composite in this sample (Cronbach's $\alpha = 0.70$ at time 1 and 0.69 at time 2) is consistent with previous research.[30] The SB severity scores in this sample ranged from 4 to 10 (mean [SD], 7.70 [1.68]).

As an index of self-reported pain, the participants rated their worst pain in the last week using a 10-cm horizontal visual analog scale (1, no pain, to 10, extreme amount of pain). Previous research on the experience of pain in individuals with SB found the worst pain in the last week, but not the current level of pain, to correlate with psychologic distress[31] ($r = 0.51$, $P < 0.01$).

Satisfaction with Family Functioning

The Family APGAR provided an assessment of how satisfied the participants were with family interactions.[32] The self-report scale measures five dimensions of family functioning: adaptation, partnership, growth, affection, and resolve (five items; e.g., "I am satisfied that I can turn to my family for help when something is troubling me"), with higher scores (items range from 1, never, to 5, always) reflecting greater levels of family satisfaction. The measure has been psychometrically established for use with individuals with SB.[18,24] The Cronbach's α values in the current sample were 0.91 at time 1 and 0.94 at time 2, and test-retest reliability was 0.66.

Self-management

The Adolescent Self-Management and Independence Scale II, a structured clinical interview, measured the participants' level of self-management.[33] The 17-item instrument gathers information about SB knowledge and self-care skills (e.g., managing SB medication, SB complication prevention, personal safety) as well as skills for general activities of daily living (e.g., managing money, transportation, making money, managing insurance, household skills, social communication, and general problem solving). The study staff rated the participants' descriptions of their self-management activities on a 7-point response category (1, total assistance, to 7, complete independence). Interrater reliability was established with standard case scoring by data collectors on two cases created by one of the Adolescent Self-Management and Independence Scale II developers before beginning time 1 data collection and again before time 2 data collection ($r = 0.90$). Strong internal reliability ($\alpha = 0.93$ at time 1 and 0.89 at time 2) and acceptable test-retest reliability ($r = 0.79$) were observed in this sample.

Quality-of-Life

The 26-item World Health Organization QOL–BREF instrument[34] provided an assessment of four related domains of QOL: physical health (seven items; e.g., pain, energy, mobility, and activities), psychological (six items; e.g., self-esteem, body image, and cognitions), social relationships (three items; e.g., personal relations, social support, and physical intimacy), and environment (eight items; e.g., safety and security, health/social care, finances, and home environment). The items range from 1, low, to 5, high, with higher scores reflecting greater levels of perceived QOL. The World Health Organization QOL–BREF is a psychometrically strong, cross-culturally valid measure of QOL in individuals with SB.[35] The Cronbach's α and test-retest reliability for the domains were the following: physical health domain (time 1, $\alpha = 0.78$; time 2, $\alpha = 0.80$; $r = 0.69$), psychological domain (time 1, $\alpha = 0.69$; time 2, $\alpha = 0.73$; $r = 0.58$), social relationships domain (time 1, $\alpha = 0.62$; time 2, $\alpha = 0.62$; $r = 0.63$), and environment domain (time 1, $\alpha = 0.67$; time 2, $\alpha = 0.68$; $r = 0.53$).

Statistical Analysis

Statistical analysis included examination of group and individual differences in the QOL domains across time using the Predictive Analytics SoftWare (version 18). The use of change scores is one method for analyzing individual-level difference across multiple time points, and these scores are derived by subtracting time 1 scores from time 2 scores. Considered an "unbiased estimate of true change" (Rogosa,[36] p 180), the analysis of change scores is best suited for examining individual change when the direction of change is not consistent for all individuals.[37] Positive change scores indicate an increase in scores over time, and negative values indicate a decrease in scores over time. The significance of change scores can be tested using reliable change indices (RCIs), which allow the researcher to determine whether the magnitude of observed differences from time 1 to time 2 is statistically significant or caused by chance. RCIs are calculated as the difference in scores between time points divided by the standard error of the difference in scores between time points.[38] Similar to other standardized distributions (e.g., t scores and z scores), RCI values in excess of ±1.96 indicate statistically significant differences at $\alpha = 0.05$. The reliability of change scores is maximized when (1) reliability is high at each time point the measure is used and (2) the correlation between scores at different time points is moderate.[39] As reported above, all measures had acceptable internal consistency at both time points, and correlations between time 1 and time 2 scores (i.e., test-retest reliability) fell in the moderate-to-high range.

Differences in QOL domain scores at time 1 and time 2 for the full sample were assessed using paired-samples t tests. Lastly, hierarchical regression models were run to explore the contribution of variables from the Life Course Model for SB in predicting change in QOL domain scores over time. The total variance (model R^2) and the change in explained variance associated with each step of the model (model $R^2\Delta$) were examined. An a priori power analysis conducted used G*Power[40] indicated that a sample size of 36 was required for the proposed analysis based on the following parameters: (1) $\alpha = 0.05$, (2) B = 0.20, (3) three predictors in the model, and (4) a large effect size of $f^2 = 0.35$.

RESULTS

At time 2, the mean age of the sample was 22.04 yrs (SD, 2.16; range, 19–26 yrs). The participants were primarily white (n = 37, 77%), and a slight majority were women (n = 26, 54%). More than half of the sample was unemployed (n = 26, 54.2%), and only 10% of the emerging adults were employed full time (n = 5). The participants who were employed generally held low-wage positions (for example, guest services, sheltered workshop). Two-thirds (n = 31, 64.6%) resided in a supervised living setting, such as at the home of their parents/guardians or in a group home. Similar to other SB cohort studies,[41,42] chart reviews indicated that the participants primarily had a diagnosis of myelomeningocele, which is the most severe form of SB (n = 41, 85.5). Other types of SB included lipomyelomeningocele (n = 5, 10.5%) and meningocele (n = 1, 2.1%). More than two-thirds had shunted hydrocephalus (n = 33, 68.8%), with a mean (SD) of 2.95 (2.68) surgical revisions to the shunt. The lesion level spanned all levels of the spinal cord, but lumbar level of lesion was most frequently reported in the medical chart (n = 27, 56.3%), followed by sacral level of lesion (n = 15, 31.3%) and thoracic level of lesion (n = 6, 12.5%).

A summary of descriptive statistics for the study variables is provided in Table 1. Positive change score values indicate an increase in the scores from time 1 to time 2, and negative values reflect a decrease in scores over time. The group mean score for satisfaction with family functioning remained consistent across time points (t_{47} = 0.26, P = 0.79), but examination of individual change scores reflected some variation. On the basis of RCI values, 16 individuals (33%) demonstrated significant variation in family satisfaction from time 1 to time 2 (7 decreased and 9 increased). Time 2 scores for pain were significantly lower (t_{47} = 5.01, P < 0.001) than what was observed at time 1, reflecting a decrease in pain over time for the full sample. Significant variation was also detected at the individual level for six emerging adults (12.5%), with significant decreases in pain found in four of these participants. The most commonly reported sources of pain were the shoulder, the back, and headaches. Self-management scores for the full sample significantly improved (t_{47} = −4.76, P < 0.001) from time 1 to time 2. On the basis of RCI values, 18 participants (37.5%) had a significant change in self-management over time, with 17 of these emerging adults reporting improvement from time 1 to time 2.

Individual- and Group-Level Change in QOL

On the basis of tests of normality, skewness, and kurtosis, change scores for each QOL domain were normally distributed. The distribution of significant differences in QOL for the individuals is summarized

TABLE 1 Descriptive statistics for the study variables

Variable	Mean (SD)			Sample Range (Scale Range)		Change Scores	
	Time 1	Time 2	Change	Time 1	Time 2	RCI Range	Sig. RCI
SB severity	7.70 (1.68)	NA	NA	4 to 10	NA	NA	NA
Family satisfaction	19.97 (4.48)	20.02 (4.88)	0.14 (3.88)	6 to 25 (5 to 25)	8 to 25 (5 to 25)	−13 to +7	7 (−), 9 (+)
Pain	5.17 (3.19)	4.06 (3.02)	−1.11 (3.53)	1 to 10 (1 to 10)	1 to 10 (1 to 10)	−9 to +9	4 (−), 2 (+)
Self-management	76.06 (24.17)	86.19 (19.42)	1.41 (2.06)	35 to 118 (17 to 119)	41 to 116 (17 to 119)	−19 to +53	1 (−), 17 (+)
Physical health QOL	26.39 (4.84)	26.33 (4.64)	−0.06 (3.81)	9 to 35 (7 to 35)	13 to 34 (7 to 35)	−6 to +9	11 (−), 9 (+)
Psychological QOL	21.58 (3.91)	21.98 (4.26)	0.39 (3.74)	11 to 29 (6 to 30)	11 to 28 (6 to 30)	−11 to +7	3 (−), 6 (+)
Social Relationships QOL	10.79 (2.58)	10.47 (2.58)	−0.32 (2.19)	5 to 15 (3 to 15)	3 to 15 (3 to 15)	−6 to +6	8 (−), 2 (+)
Environment QOL	31.00 (4.45)	31.04 (4.94)	0.04 (4.58)	23 to 39 (8 to 40)	20 to 40 (8 to 40)	−14 to +9	4 (−), 6 (+)

The symbol + indicates a significant positive RCI and − indicates a significant negative RCI; Sig, significance, P < 0.05; NA, not applicable, as that variable could not change over time.

in Table 1. Of the 20 emerging adults (41.6%) demonstrating significant changes in physical health QOL scores over time, 9 were in a positive direction. Significant individual-level variation in psychological QOL scores was generally positive, with six of nine participants displaying increases. Assessment of each individual's change in social relationship QOL revealed that 20% of those with substantial variation (two of ten) were positive changes. RCI analysis of environment QOL scores identified ten individuals (20.8%) with significant variation over time and six of these emerging adults having higher scores at time 2. Figure 1 depicts variation in individual change scores for each QOL domain. No group-level differences in QOL scores were observed over time (physical health QOL, $t_{47} = 0.11$ [$P = 0.91$]; psychological QOL, $t_{47} = -0.73$ [$P = 0.47$]; social relationships QOL, $t_{47} = 1.06$ [$P = 0.29$]; and environment QOL, $t_{47} = -0.06$ [$P = 0.95$]).

Bivariate Relationships

Because of the small sample size and the need to maximize statistical power for hypothesis testing, significant bivariate relationships among variables from the Life Course Model for SB and the QOL domains were assessed before entry into the regression models. No sex differences were found in the change scores for any of the four QOL domains ($P > 0.05$), nor did SB type (myelomeningocele *vs.* other types of SB) differentiate change in QOL domains from time 1 to time 2 ($P > 0.05$). Employment status and change in self-management skills similarly failed to predict change in QOL across time ($P > 0.05$). To maximize statistical power, these variables were not included as covariates in the regression models. However, change in physical health QOL from time 1 to time 2 was significantly associated with change in satisfaction with family functioning ($r = 0.40$, $P = 0.005$), with increased family satisfaction over time associated with greater physical health QOL at time 2. An increase in family satisfaction from time 1 to time 2 was similarly related to improved psychological QOL at time 2 ($r = 0.42$, $P = 0.003$), but a higher level of SB severity ($r = -0.35$, $P = 0.015$) was associated with reduced psychological QOL over time. Unexpectedly, change in social relationships QOL was positively correlated with change in pain ($r = 0.34$, $P = 0.02$), suggesting that the social QOL scores increased as self-reported pain increased. Lastly, increased satisfaction with family functioning

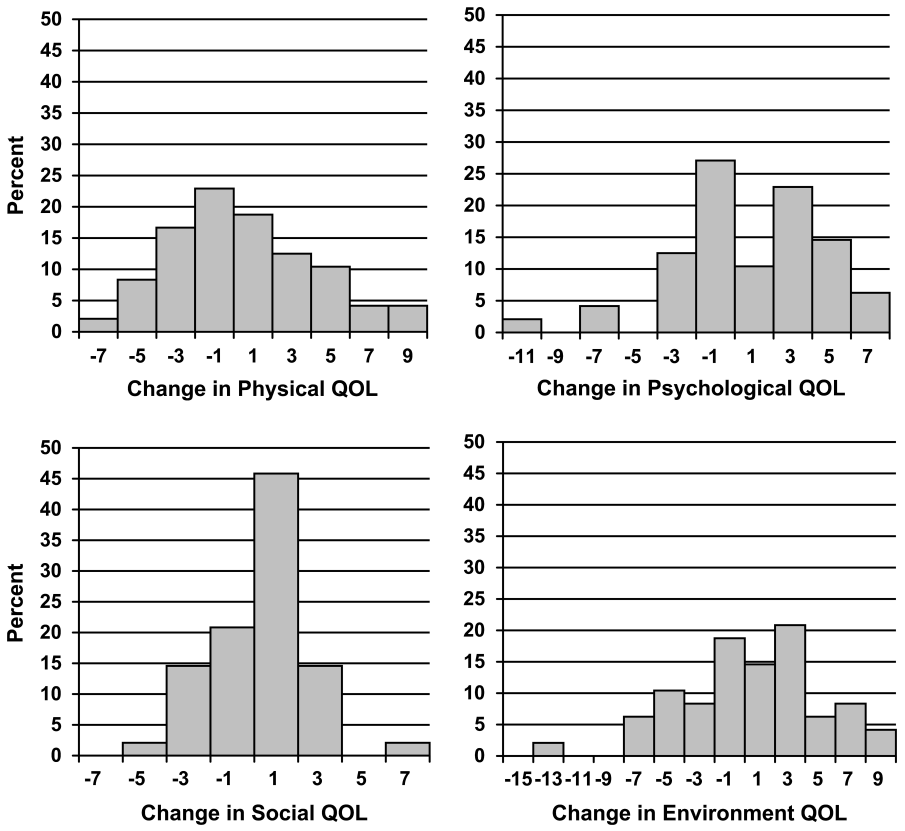

FIGURE 1 *Individual QOL domain change scores across time. Positive change scores indicate an increase in QOL from time 1 to time 2, and negative values indicate a decrease in QOL over time.*

over time was associated with improved environment QOL scores from time 1 to time 2 ($r = 0.32$, $P = 0.03$).

QOL Multiple Regression Models

Change in Physical Health QOL

As indicated in Table 2, the inclusion of SB severity in step 1 of the model did not yield a significant R^2 ($F_{1,45} = 0.29$, $P = 0.59$), but the addition of change scores for satisfaction with family functioning and pain in step 2 resulted in a significant $R^2\Delta$ and final model R^2 of 0.23 ($F_{3,43} = 4.25$; $P = 0.009$; 95% confidence interval [CI], 0.03–0.42), reflecting a medium to large effect size[43] ($f^2 = 0.30$). In the final model, change in satisfaction with family functioning was a significant predictor (B = 0.38, $t = 2.85$, $P = 0.007$), as was change in pain (B = −0.29, $t = -2.03$, $P = 0.049$). Specifically, increases in family satisfaction scores over time were predictive of higher physical health QOL at time 2, whereas increases in pain were associated with lower physical health QOL over time. SB severity was nonsignificant ($P = 0.36$).

Change in Psychological QOL

Each step in the hierarchical regression had a significant $R^2\Delta$. SB severity on step 1 yielded an R^2 of 0.12 ($F_{1,45} = 6.09$; $P = 0.017$; 95% CI, 0.01–0.28). Inclusion of the family satisfaction and pain change variables in step 2 resulted in a significant $R^2\Delta$ of 0.26 ($F_{2,43} = 9.26$; $P < 0.001$; 95% CI, 0.06–0.25). The overall model R^2 of 0.38 ($F_{3,43} = 8.95$; $P < 0.001$; 95% CI, 0.18–0.58) was consistent with a large effect size ($f^2 = 0.61$). SB severity remained significant in the final model (B = −0.68, $t = -2.46$, $P = 0.02$) and indicated that a higher degree of SB severity was associated with decreased psychological QOL at time 2. Change in family satisfaction and pain over time were also significant predictors, with greater satisfaction with family functioning associated with improved psychological QOL at time 2 (B = 0.43, $t = 3.61$, $P = 0.001$) and increased pain over time (B = −0.29, $t = -2.21$, $P = 0.03$) related to decreased psychological QOL.

Change in Social Relationships QOL

R^2 for step 1 with SB severity was not statistically significant ($F_{1,45} = 1.01$, $P = 0.32$), but inclusion of change scores for satisfaction with family functioning and pain in step 2 yielded a significant $R^2\Delta$ of 0.14 for step 2 ($F_{2,43} = 3.66$, $P = 0.03$). However, the test of the overall model R^2 (0.16) was not statistically significant ($F_{3,43} = 2.82$, $P = 0.050$), thereby precluding interpretation of individual variable parameter estimates.

Change in Environment QOL

Although $R^2\Delta$ for step 1 and step 2 were not statistically significant ($F_{1,45} = 2.25$, $P = 0.14$, and $F_{2,43} = 3.07$, $P = 0.06$, respectively), the overall model R^2 (0.17) was statistically significant ($F_{3,43} = 2.87$; $P = 0.048$; 95% CI, 0.01–0.35) and within the range of a medium effect size ($f^2 = 0.20$). Change in satisfaction with family functioning was the only significant predictor in the final model (B =0.39, $t = 2.44$, $P = 0.02$), with increased family satisfaction over time associated with improved environment QOL scores at time 2.

DISCUSSION

The purposes of this study were to advance knowledge of QOL in individuals with SB during emerging adulthood and to offer a preliminary understanding of how variables from the Life Course Model for SB[19] may contribute to change in QOL over time. No significant changes in QOL were found from time 1 to time 2 for the full group. However, the lack of group-level differences in QOL may, in part, be explained by the substantial bidirectional variation observed at the individual level. For each QOL domain, some participants reported increases over time, whereas others demonstrated a decline in scores from time 1 to time 2. When these positive and negative changes are similar in magnitude, mean scores for the full sample do not vary across time. Divergent patterns of association among the Life Course variables and QOL also emerged. Consistent with previous findings of a limited association between SB impairment indicators and QOL, the relationship between SB severity and QOL was modest in this sample of emerging adults with SB. Interestingly, SB severity was not associated with physical health QOL and instead predicted change in only psychological QOL, with a higher degree of SB severity related to a decrease in psychological QOL from time 1 to time 2. These findings indicate that SB severity is not a driving force of QOL in these emerging adults. However, rehabilitation-focused research involving other medical populations (e.g., spinal cord injury) suggests that the relationship between impairment severity and QOL may be indirect, with a mediating role played by the disruptive impact of severity on activities, participation, and related variables.[44,45]

Self-reported pain may be a potential component of this indirect role of SB severity insofar as an increase in pain across time was associated with decreases in both physical health and psychological QOL. As individuals with SB age, secondary musculoskeletal problems such as shoulder pain or

TABLE 2 Hierarchical regression results

	DV = Changes in Physical QOL						
	Model	**B**	**SE**	**β**	**95% CI**	R^2	$R^2\Delta$
1	(Constant)	−1.53	2.62		−6.81 to 3.74	0.006	0.006
	SB severity	0.18	0.32	0.08	−0.49 to 0.85		
2	(Constant)	−2.69	2.46		−7.65 to 2.27	0.23[b]	0.23[b]
	SB severity	0.28	0.31	0.13	−0.34 to 0.90		
	Δ Family satisfaction	0.37[a]	0.13	0.38	0.11 to 0.63		
	Δ Pain	−0.29[b]	0.15	−0.28	−0.60 to −0.002		
	DV = Changes in Psychological QOL						
	Model	**B**	**SE**	**β**	**95% CI**	R^2	$R^2\Delta$
1	(Constant)	6.33[a]	2.45		1.39 to 11.26	0.12[a]	0.12[a]
	SB severity	−0.77[a]	0.31	0.29[a]	−1.39 to −0.14		
2	(Constant)	5.25[a]	2.19		0.83 to 9.67	0.38[c]	0.26[c]
	SB severity	−0.68[a]	0.27	−0.30	−1.22 to −0.12		
	Δ Family satisfaction	0.42[b]	0.12	0.43	0.18 to 0.65		
	Δ Pain	−0.29[a]	0.13	−0.27	−0.55 to −0.02		
	DV = Changes in Social QOL						
	Model	**B**	**SE**	**β**	**95% CI**	R^2	$R^2\Delta$
1	(Constant)	−1.62	1.40		−4.45 to 1.21	0.02	0.02
	SB severity	0.18	0.18	0.15	−0.18 to 0.54		
2	(Constant)	−0.62	1.39		−3.45 to 2.18	0.16	0.14[a]
	SB severity	0.08	0.17	0.06	−0.28 to 0.43		
	Δ Family satisfaction	0.11	0.07	0.21	−0.04 to 0.26		
	Δ Pain	0.19	0.08	0.33	0.02 to 0.36		
	DV = Changes in Environmental QOL						
	Model	**B**	**SE**	**β**	**95% CI**	R^2	$R^2\Delta$
1	(Constant)	−4.21	3.02		−20.39 to 2.87	0.05	0.05
	SB severity	0.58	0.38	0.22	−0.19 to 1.35		
2	(Constant)	−4.25	3.02		−10.34 to 1.84	0.17[a]	0.12
	SB severity	0.57	0.38	0.21	−0.29 to 1.33		
	Δ Family satisfaction	0.39	0.16	0.34[a]	0.07 to 0.71		
	Δ Pain	−0.07	0.18	−0.05	−0.43 to 0.29		

[a] $P < 0.05$.
[b] $P < 0.01$.
[c] $P < 0.001$.

recurrent tethered cord syndrome are common sources of pain and are often associated with impaired mobility such as in wheelchair propulsion, transfers, or ambulation.[1,5,6] In turn, these physical limitations may negatively influence perceptions of body image and self-esteem, especially if the emerging adults with SB perceive themselves as different from peers. Pain has also been linked with disruptions of activity, personal care, and participation in other medical populations, [46,47] and this association could potentially explain the pattern of QOL variability seen in individuals with SB in this study. Lastly, the findings of a negative influence of pain on QOL are consistent with previous research with a mixed sample of adolescents and adults with SB[10] and underscore the critical need to systematically screen for pain in clinical encounters with this population to protect against deterioration in QOL.

Another notable contribution of this study is a preliminary understanding of the role family satisfaction plays in supporting positive QOL in emerging adults with SB. Sawin and colleagues[24] identified the salience of family interactions on QOL in adolescents with SB, and the findings from this study suggest that the family context continues to meaningfully contribute to QOL as individuals with SB transition into early adulthood. Specifically, an increase in satisfaction with family functioning across time was related to improved physical health and psychological QOL at time 2, and family satisfaction was the only significant factor in the environment QOL model. These observed relationships are

consistent with research with comparison populations such as cerebral palsy in which social support in the home was strongly linked to community participation.[46]

The study findings also offer guidance about key components to include in interventions to enhance QOL in emerging adults with SB. A research review on QOL in this population previously identified a gap in science for interventions targeting physical and psychosocial dimensions of QOL, including strategies to reduce the risk for low QOL in individuals with SB.[3] To optimize QOL, the current findings provide preliminary support for screening for self-reported pain; supporting positive interactions in the family system; and guided discussion about negative feelings, low self-esteem, and poor body image, especially for those with greater SB severity.

Limitations

The small sample of convenience and the study methodology present several limitations to the interpretation of key findings. In particular, the limited power restricted the types of analyses run and the number of variables entering the QOL models. There was insufficient power to explore a possible interaction between SB severity and self-reported pain on QOL. SB severity perhaps moderates the observed relationship between pain and psychological QOL, with greater severity exacerbating the negative impact of pain on QOL. It is also important to note that the findings provide a tentative picture of the relationships among the study variables. Future research with a larger sample and more data points for a larger period will allow for a more sophisticated probing of the directionality among significant relationships.

Future research should also explore other individual and contextual factors influencing QOL in emerging adults with SB, especially in light of the lack of support for the social relationships QOL model. For example, the contribution of obesity in explaining variation in QOL was not examined. This is significant because one-third of young adults with SB are considered obese, and obesity rates are even higher in later adulthood.[48,49] The QOL models may have also been improved by including measures of life stress. The work of Alriksoon-Schmidt and colleagues[25] previously identified a significant association between life stress and QOL, with higher stress predicting lower QOL in a sample of adolescents with a physical disability including SB. Lastly, executive functioning was recently identified as a significant predictor of subjective QOL in young adults with SB.[50]

Despite these limitations, the study findings identified factors that were significant and clinically relevant to enhance QOL in emerging adults with SB. The major take-home point from this study is that clinicians caring for individuals with SB should be aware of the negative impact of untreated pain and the protective influence of satisfaction with family functioning. Screening for and addressing pain should be a priority when treating and rehabilitating these patients. This study also supports preventive mental health services that are targeted to individuals with more severe SB and interventions aimed at promoting family function and incorporating family support into care plans for all emerging adults with SB.

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CME Article 2013 Series Number 7: *Bellin et al.*

1. In this study, all of the following were used to determine spina bifida severity except
   A. Age
   B. Lesion level
   C. Shunting status
   D. Ambulation status

2. In the Life Course Model for Spina Bifida, which of the following is NOT a functional domain that contributes to positive quality-of-life outcomes?
   A. Mobility status
   B. Social and personal relationships
   C. Education and income support
   D. Self-management and health

3. In this study increased family satisfaction over time was associated with which of the following?
   A. Greater physical health quality-of-life
   B. Greater psychological quality-of-life
   C. A and B
   D. None of the above

4. According to this study, screening for which of the following may help a physiatrist protect against deterioration in physical and psychological quality-of-life in his/her patients with spina bifida?
   A. Falls
   B. Pain
   C. Alcohol use
   D. Drug use

5. In this study a greater level of spina bifida severity was associated with:
   A. Reduced physical health quality-of-life
   B. Reduced psychological quality-of-life
   C. A and B
   D. None of the above

(Continued next page)

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