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REVIEW

Long-Term Quality of Life for Patients with Spina Bifida

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Abstract

Purpose of Review This review explores the literature that currently exists regarding quality of life for patients with spina bifida and assesses screening tools available for practitioners to assess these outcomes. Domains addressed include health related quality of life, relationships, bowel and bladder dysfunction, sexuality, pain, and mood.

Recent Findings Several recent papers have assessed available questionnaires and reviewed findings of key quality of life domains, with varying results.

Summary Historically, the literature has focused on surgical outcomes for patients with spina bifida. Though some authors have begun to explore quality of life for this patient population, particularly in the short-term, a paucity of quality, long-term studies exist that describe quality of life domains or offer potential targets for improvements in outcomes.

Keywords Spina bifida · Quality of life outcomes · Quality of life surveys · Neurogenic bladder

Introduction

Spina bifida (SB) is a group of congenital disorders caused by the incomplete closure of the developing neural tube. Myelomeningocele (MMC), the most common type of SB lesion, is defined by extrusion of the spinal cord outside of the canal. Compared to the closed type (non-myelomeningocele), MMC is associated with more severe neurologic consequences. All types of SB have the potential to lead to a wide variety of clinical manifestations including lower extremity weakness, decreased sensation, bladder and bowel dysfunction, bone and joint abnormalities, and sexual dysfunction [1•].

Globally, SB impacts approximately 40 per 100,000 live births per year [2]. Early emphasis in SB care was primarily focused on surgical and medical efforts to extend life expectancy and reduce morbidity. Due to rapid and continuous improvements in surgical and medical management, the life expectancy for these patients has increased dramatically in the past several decades, increasing from about 15 years in 1956 to nearly as high as that of the general population today [3]. In the United States, it is conservatively estimated that 166,000 or more individuals are currently living with SB and the majority are adults [4–6].

With more patients surviving into adulthood, it is important to expand academic focus to include long-term quality of life (QOL) considerations. While efforts have begun to investigate QOL outcomes, minimal research exists examining long-term patient outcomes, nor addressing potential solutions to improve QOL. The limited studies available focus more on function, rather than perception and lived experience of SB patients [7, 8••]. A shift away from immediate short-term surgical successes to long-term QOL metrics will be vital. Accurate assessment and use of these metrics have been shown to aid clinical decision making, improve outcomes, and patient satisfaction [9]. Additionally, a focus on QOL is particularly important for urologists, given many symptoms treated by the specialty are chronic and can impact patient psychosocial condition (i.e. bladder, self-esteem, and sexuality). As such, many in the field have called for QOL measures to better individualize and tailor patient care [10].

This review outlines the literature describing QOL survey instruments and current long-term studies assessing QOL across multiple domains.

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Quality of Life Survey Instruments

In 1995, the World Health Organization (WHO) published a definition of QOL as "a state of complete physical, mental and social well-being and not merely the absence of disease," noting that QOL is inherently subjective [11]. To our knowledge, the first study about QOL metrics in patients with SB was published in 1997 and, since then, only a handful of studies have become available [12].

In the 2020s, a strong shift towards focusing on patient experience developed, which led to a 2021 review by Raveendran et al. evaluating the available literature on available QOL survey instruments in pediatric urology [8••].

Survey instruments can be broken down into those that assess function (i.e. both physical and social activities), quality of life (i.e. subjective perception of a respondent's life and goals), or a combination of both. Raveendran identified 9 instruments (38 studies) assessing function, 4 instruments (9 studies) assessing QOL, and 9 instruments (17 studies) assessing a combination of function and QOL [8••]. The authors go on to discuss the strengths and weaknesses of each survey instrument and provide a summary of all available surveys.

Overall, Raveendran et al. found that 88.4% of studies used an instrument assessing function while only 9% of studies used an instrument exclusively designed to measure QOL, highlighting the continued need to improve strategies for measuring and studying QOL [8••]. The most used survey in pediatric urology was the Pediatric Quality of Life Inventory (PedsQL), a survey designed to measure function. Quality of Life Assessment in Spina Bifida (QUALAS), KINDL, KIDSCREEN, and DISABKIDS are surveys that are designed to measure quality and should be considered over more traditional questionnaires when selecting an instrument [8••].

In response to Raveendran et al., Tasian and Ellison (2021) published a commentary in which they emphasize the importance of intentionality in instrument selection. They propose that it is not only vital to consider patient experience, but to select instruments that focus on "clinically relevant and actionable aspects of physical, social, and emotional health [13]." In addition, the continuous inclusion of patients and caregivers is particularly important when considering lived experiences, given goals and expectations readily change throughout life. Finally, most studies use instruments that rely on typically developing individuals or measures designed for a different age group or disability [14]. Szymanski et al. emphasize that questionnaires must be validated not only using statistical analyses, but also with qualitative analyses of face validity to ensure they measure what they purport to measure. They write, "QOL is a subjective experience… Therefore, QOL questionnaires should answer if what is happening matters to the child." [15••].

In addition to selecting a survey that focuses on QOL, it is vital to use one that is validated and specific to the age and condition of the patient in question. QUALAS as well as PROMIS, a group of NIH-sponsored assessment tools developed specifically for patient populations in question [16], may be two such measures.

Self-Report v. Proxy

One additional important consideration in selecting a survey instrument is the concept of self- versus proxy report. In this population, particularly when patients are still children, it is common for caregivers or family members to represent the patient. Though some surveys have been validated specifically for use by a caregiver, self-report is the superior choice [15••]. Historically, the literature has demonstrated that patients are not as bothered by symptoms as parents and providers may perceive [17]. With some variation [18], this trend has largely remained true in the last several decades [19, 20]. These findings provide support for the "disability paradox," a concept in which observers (aka "proxies"), such as family members, caregivers or healthcare professionals, underestimate the QOL of people under their care.

Furthermore, many QOL surveys equate function with high quality, which inherently discriminates against individuals with disabilities as it assumes people with disabilities automatically cannot have as high of quality of life. Sawin et al. write "this conceptual equation devalues the lives of people with disabilities by automatically declaring that a person with a disability cannot have as good a quality of life as someone without disabilities." [20].

To limit this bias and appropriately prioritize and remedy treatable symptoms, it is vital to ask patients directly about their quality of life and to select a survey instrument designed to assess quality and not function.

Quality of Life Domains

Domains of QOL measured in surveys are varied, reflecting broad areas of concern for these patients. Starowicz et al. (2021) report the most common chief complaints in this patient population are care coordination, neurogenic bladder, medications, assistive devices, and neurogenic bowel. Additional concerns include pain, sexuality, and mental health [21].

As such, we will review available literature across the domains of health-related quality of life, relationships, bowel and bladder dysfunction, sexuality, pain, and mood.

Health Related Quality of Life

Health related quality of life (HRQOL) refers to the patient's subjective opinion of the impact of a condition or health-status on their day-to-day life [22].

Several studies directly examine this sub-domain for patients with SB. The PROMIS Global Health questionnaire (PGH-7) is a questionnaire that asks individuals to rate their overall health, quality of life, and their physical, mental, and social health. The median PGH-7 score in the U.S. population is 50 (± 10) [23]. A 2022 study used the PGH-7 to examine the association between health literacy and self-reported quality of life in adolescents and adults with SB aged 12–31 years. The authors found that mean PGH-7 score was 47 (± 8) with a positive association between health literacy and patient reported outcomes [24•]. These data suggest a limited reduction in HRQOL for patients with SB exists when compared to the general population.

Conversely, Rocque et al. reported that patients with MMC had significantly lower QOL overall than other forms of SB. The authors evaluated children and adolescents with SB aged 5–20 using the Health Utilities Index-3 (HUI3) [25]. The HUI3 is not specific to SB and overrepresents ambulation, which could explain this finding and not necessarily be a true representation of HRQOL.

Though additional long-term studies for this domain are needed, available data of HRQOL in SB patients does not show strong evidence of differences compared to healthy controls.

Importance of Relationships

As for any person, close interpersonal relationships are vital for patients with spina bifida and are an important consideration in assessment of quality of life. Thus, several studies have examined the role of relationships and their impact on QOL. In 2019, Ridosh et al. asked the question "what makes a good quality of life?" to 209 families and patients with SB. The most frequently cited response was an engaged family [14]. Further, in a 2021 retrospective review evaluating health concerns amongst 94 adults with SB, Starowicz et al. reported social and multidisciplinary care as common concerns [21].

Fremion (2021) used QUALAS, a survey tool validated specifically for patients with SB, to examine a wide variety of domains including family/independence. They found that in a group of 88 young adults with SB, they had a score of 73.8 (± 19.9) [26••]. This value is lower than the mean for adolescents without SB of 86.1 (± 18.8) [27].

In general, though representing an important contributor to overall QOL, relationships and familial support are understudied in this patient population. The few studies that exist suggest that this population has lower QOL in this domain, pointing to relationships as a key target for intervention and creative problem solving.

Impact of Bladder and Bowel Dysfunction

Bladder and bowel incontinence have a strong impact on QOL, a finding that has been well supported in recent years. In 2015, Wagner et al. surveyed 72 adults with SB aged 18–68 using the validated Spina Bifida Secondary Conditions (SBSC) tool and found that 54% experienced stool incontinence and 36% experienced bladder incontinence. 90% of those with bowel incontinence and 81% of those with bladder incontinence felt this symptom impacted their daily life [28]. Likewise, achieving continence has been demonstrated to improve outcomes. One study found that a bowel management strategy improved QOL across all domains for SB patients [18]. Another identified a positive association between continence and employment [29], further bolstering continence as a modifiable target to improve long-term QOL.

Data suggests that the impact of bladder and bowel continence is age dependent. Szymanski et al. examined the impact of bladder and bowel incontinence on QOL in a population of children and adolescents with SB. The authors found an inverse relationship between urinary incontinence and QOL, with an increasing impact as patients age [30]. In other words, social continence becomes increasingly important as this population grows into adolescence and adulthood, expanding on a previous study by the same research group that focused on incontinence in adults [31]. The findings by Szymanski et al. are supported by a recent study from Gilbertson et al. that retrospectively reviewed continence and outcomes in 7,669 patients with SB using data gathered from the National Spina Bifida Patient Registry (NSBPR) [1•]. The authors reported higher continence for patients with non-myelomeningocele lesions (bowel: 72.5%, bladder: 63.2%) when compared to patients with MMC (bowel: 53.1%, bladder: 40.4%). Additionally, the authors concluded that continence is inversely associated with age [1•]. As children grow, continence improves, but incontinence becomes more detrimental to patient QOL.

The impact of bladder and bowel interventions on QOL has also been studied in recent years. Bladder augmentation in neurogenic bladder is performed for renal protection and improvement in continence, often with both indications being present. The presumption is that such surgical interventions also improve QOL, and several studies have examined this question. Romero-Maroto et al. performed a study with a 20 year follow up demonstrating that bladder augmentation both improved kidney function and improved QOL, with 94% experiencing diurnal continence postoperativley [32]. Coco et al. examined a similar question in a group of 54 patients with a median age of 30 years.

The authors found that patients with a history of bladder augmentation had lower independence than those without (49 v. 68) [33]. This finding, however, did not control for lesion-level or pre-operative QOL. Fremion et al. examined whether creation of a continent catheterizable channel for clean intermittent catheterization (CIC) impacted independence using the Adolescent/Young Adult Self-management and Independence Scale II Self-Report/SB (AMIS II-SR/SB) and QUALAS-T questionnaires. The study showed that that urethral CIC, but not abdominal CIC, was associated with a higher independence score [26••]. In evaluating the impact of a bladder training video on QOL in a randomized controlled trial, Brownrigg et al. found that self-confidence related to bladder and bowel was poor both pre and post intervention [34]. Finally, using a modified Peristeen® Neurogenic Bladder Dysfunction NBD questionnaire and the PROMIS-PGH scale, Halstead et al. demonstrated that, though no association exists between bowel program and QOL, more severe bowel symptoms are associated with a reduced QOL [35]. In summary, these studies indicate that regardless of intervention strategy, bladder and bowel continue to be key considerations for QOL and merit special attention by both providers and researchers, alike.

When examining these studies critically, several limitations emerged. Many studies were limited to children. Of the studies with long-term follow-up, most had a small sample size. Furthermore, the distinction between function and QOL is often less clear. As an example, though they included a large sample size, Gilbertson et al. did not report on the impact, or lack thereof, of continence on lived experience or QOL. Additional investigation is merited into how continence plays or does not play a role for these patients.

Sexual Function and Sexuality

As patients with SB begin to live longer lives, it is incumbent on urologists to have a better understanding sexuality and sexual function. In recent studies, data indicate that many patients with SB are sexually active or interested in sexual activity, with over 50% of patients reporting that they engage in sexual activity [36–39]. Through a series of structured interviews, Rague et al. found that most young adults with SB are interested in talking about sex and sexuality. Though conversations with providers are commonly viewed as uncomfortable by patients, suggestions for overcoming barriers to discussions include notifying patients in advance of the topic, creating a safe space where patients can drive the conversation, and addressing their unique physiologic situation as it relates to sexual function [39].

Sexual satisfaction in patients with SB demonstrates significant variability. Romero-Maroto et al. reported 43% of those surveyed using a non-validated quality of life questionnaire were satisfied with their sexual relations, a proportion supported by a review paper by De Win et al. in 2020 who reported 50% sexual satisfaction [32, 40]. After surveying 47 men with SB using the International Index of Erectile Function (IIEF), Choi et al. found that though 67% had normal erectile function and 50% had normal orgasmic function, 87% reported dysfunction or dissatisfaction [38]. In 2015, Akre et al. asked patients aged 13–28 to self-record thoughts or experiences with sexual knowledge and sexuality. Common concerns, questions, and challenges were elicited including romantic relationships, fertility, and lack of communication regarding sex and sexuality [41]. Though the literature reports varying levels of sexual satisfaction, patients with SB appear to be negatively impacted.

Two recent review papers by Hughes et al. and Streur et al. both emphasize that while some work has begun to analyze and understand sexual function and satisfaction for these patients, high quality data is significantly lacking [42•, 43•]. For example, though the IIEF has been used to assess sexual function in patients with SB, it is a survey that was validated in a group of typically developing men. In a qualitative study of 20 men with SB, Rague et al. found serious limitations of the IIEF in assessing this population, including lack of recognition of poor sensation, incontinence, SB-specific mobility limitations, and more [44]. SB-specific sexual function questionnaires are not commonly used to capture the true patient experience in the limited studies that exist examining sexuality in this patient population.

In summary, multi-institutional studies using surveys and questionnaires validated for this specific patient population will be vital to understand, tailor treatment plans, and improve sexual health for patients with spina bifida.

Pain

The unique nature of spina bifida means patients are more likely to experience pain than the general population [45]. The impact of this pain continues to be evaluated.

Pain is a significant factor to patient well-being, especially as these patients age. Lidal et al. found that for older adults with SB (mean age 58), pain was their most notable health concern [46•]. Wagner et al. noted that 90% of patients with SB experience pain, with impact on daily life ranging from 52 to 94% depending on the location of the pain [28]. More conservative estimates of impact are available as well, with rates as low as 20–40% [21, 47]. A very recent prospective study of 51 adults using PROMIS, a survey tool validated in this patient population, found that more women than men reported pain (69% v. 38%, $p=0.003$), but there was no difference in the impact of pain on QOL by gender ($p=0.138$). Higher pain interference was associated with lower QOL ($p=0.042$), but not mood or sleep [48].

The varying reported values of pain underscore the importance of continuing to quantify the extent to which

pain plays a role in the day-to-day life of this population and to tailor treatment to the individual patient.

Mood

Limited literature exists regarding spina bifida and mood. Wagner et al. found that 53% of patients with SB experience depression, with 87% reporting these symptoms somewhat impact or greatly impact daily life [28]. Similarly, Dicianno et al. demonstrated that depressive symptoms are common in this population and undertreated, with 25.8% experiencing depressive symptoms and, of those, only 63% on antidepressants [49]. Screening for mood is vital at patient visits and represents a key consideration in the study of QOL.

Discussion

Wiener and Chaudry (2023) and Sawin et al. (2020) provide guidelines for management of patients with spina bifida and include references to key quality of life targets (psychosocial well-being, continence, pain, sexuality, and independence) [20, 50]. That said, while these newer guidelines often include QOL targets, few studies have described the results of surveys across a large population. Any literature examining QOL is limited, and even more so if exclusively looking at adult populations. Furthermore, the existing literature often uses questionnaires that fail to accurately measure QOL despite attempts or intentions to do so [15••]. In other words, though validated screening tools are now at our disposal, we still do not understand how patients view their QOL. A significant need remains for large-scale studies with long-term follow-up to better understand the perception of SB patient QOL.

The current literature suggests that while health related QOL is equivalent to the general population, the domains of relationships, continence, sexuality, pain, and mood all remain key targets for intervention. In measuring QOL, it is vital to be intentional in survey selection by using questionnaires validated for measuring QOL in patients with spina bifida. It is also important to rely on self-report when possible. Considering QOL in care plans and clinic visits will bolster patient satisfaction and long-term outcomes and continue to be key as this population grows into adulthood.

Conclusions

Interest in studying quality of life for patients with spina bifida has begun, but there continues to be a strong need for long-term studies using validated questionnaires that focus on quality of life, rather than function. Future studies should aim to address this gap and continue to include quality of life in care plans and treatment considerations.

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Declarations

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