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Psychosocial needs and outcomes of adults with spina bifida: A scoping review, 1974-2023

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ABSTRACT

Limited evidence exists that serves to guide the field of practice and research pertaining to the long-term issues and needs of adults with spina bifida. Understanding the lived experience of adults with spina bifida has lagged behind considerably resulting in limited evidence-based guidance for individuals with spina bifida and their families and the health care professionals who provide services to this population. Given the paucity of knowledge of the lived experience as it pertains to adulthood, this scoping review was undertaken. More than 10,000 records from 1974 through 2023 were screened based upon the search criteria whose purpose was to examine the research conducted the psychosocial outcomes and needs of adults with spina bifida. A total of 81 articles were included in this review. The findings of this review revealed significant gaps were apparent. There was limited data on adulthood benchmarks of employment, education, community living and social relationships. Limitations associated with the investigations of this review included underpowered samples, lack of longitudinal designs, use of instruments with insufficient psychometrics, and the use of clinical and administrative data sets not designed for research purposes. As the survival rates of individuals with spina bifida continue to improve with medical advances more robust psychosocial research pertaining to this population is needed.

Improvements in the diagnosis, treatments, and ongoing condition management of individuals with congenital and acquired childhood chronic conditions have contributed to improved rates of survival. The survival rates have progressively improved wherein it is now estimated that one million adolescents with chronic conditions enter adulthood annually.[1–6] A similar survival trajectory applies to individuals born with spina bifida (SB), although survival rates are closely associated with the level of SB severity that ranges from 60% to 85%.[7–10] The longevity patterns for individuals with SB have resulted in greater numbers of adults than children living with SB.[11–15] Studies have explored age-related longevity patterns in later decades of adulthood with respect to SB. One investigation reported a 36% survival rate to the mid-40 s, with a mean age of 40 years[16], whereas another study cited a 33% survival rate with a mean age of 46 years.[17] Researchers found that the extent of condition severity significantly affected survival rates, which ranged from 17% to 61%.[16]

Understandably, the SB literature has focused on medical treatments, biophysical functional outcomes, and metrics associated with condition stability and instability. Individuals with SB have increased morbidity risks as evidenced by aging and rates of accessing urgent and emergent care and hospitalizations.[18–29] Hospitalization-admitting diagnoses included treatment for neurosurgical complications (i.e., shunt revisions), urologic issues (urinary tract infections), and orthopedic and skin care problems (i.e., pressure injuries).[19,22,25,28,29,31] Lifestyle challenges uniquely affecting individuals with SB compared to the general population are the emergence of long-term secondary conditions, functional deterioration associated with aging, and the need to deal with unanticipated medical and surgical complications.[18,20,21,24,26,28,30–35] The needs for health services with declining health status were found to accompany the aging process more often as compared to typical

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populations.[12,26,28] The emphasis on biophysical findings is evidenced by the United States National Spina Bifida Patient Registry, funded by the Centers for Disease Control and Prevention. All studies ($n = 24$) in this registry are focused on clinical topics such as bowel and bladder continence, mobility, and other related clinical outcomes.[36,37]

Researchers have suggested that the transition from pediatric to adult health care services is a particularly vulnerable period of increased health risks related to hospitalization and emergent care.[19,22,23,25,27,38,39] Improvements in the provision of health care transition services to facilitate transfer of care to primary and specialty care providers could reduce health risks, preventable illnesses, and health care expenditures.[19,22,23,25,27,28] Others have advocated for a comprehensive health care transition model of care that includes the provision of self-management and system navigation training and support to foster independence as a health care consumer.[23,40,41,42] Ideally, as has been suggested, comparable interdisciplinary models of pediatric care might be replicated in adult health system of care or via an adult medical home.[22,28] Experts have acknowledged transition planning needs to address individualized, specialized needs that require additional focus for individuals with intellectual disabilities based on neurocognitive testing for the appropriate assessment of cognitive functioning.[40,41,42] Transition service accommodations may be needed for cognitive issues associated with executive functioning and memory that can affect the acquisition of skills and knowledge.[40]

By comparison, less is known about the long-term psychosocial needs and outcomes of adults with SB as they proceed into the second decade and beyond. Despite advances in medical treatments that have contributed to improved survival rates for individuals with SB, the achievement of developmental milestones associated with adulthood has lagged behind that of the general population. Rates of postsecondary enrollment, degree attainment, and employment for both full-time and part-time positions are lower for individuals with SB as compared to the general population.[8,12] Social relationship with significant others, peers, and co-workers have been rarely reported; investigations exploring community living options report that high percentages of adults with SB continue to live with parents and experience challenges with establishing social relationships.[8,12] These metrics are associated with higher risks for diminished quality of life and depression.[12,25,33,43] The literature on relevant issues regarding adult management pertaining to long-term medical and surgical care focuses on functional outcomes, with less emphasis on the psychosocial impact on the lived experience. There has been a lack of attention to the integration of the psychosocial impact of these complex medical needs on lifestyle issues associated with the lived experience for adults with SB.[38] For example, practice recommendations on bowel and bladder incontinence on activities of daily living such as employment status and community living outcomes are relevant topics for discussion. Experts have suggested that an integrated and comprehensive approach is needed to address the disparities that currently exist for adults with SB as it applies to long-term psychosocial outcomes.[21]

There is limited evidence to inform and guide practice on the lived experience of adults with SB associated with employment, education, community living, social relationships, and emotional well-being. Limitations associated with the research conducted as reviewed in this scoping review are constrained by underpowered samples, lack of longitudinal designs, use of instruments with insufficient psychometrics, and the use of clinical and administrative data sets not designed for research purposes[43].

Purpose Statement

The purpose of this scoping review is to examine the research that has been conducted to explore the psychosocial needs and related outcomes for adults with SB. It sought to map the body and evolution of research literature pertaining to this medically complex population, spanning fifty years, so as to identify the available evidence, clarify key concepts, and analyze knowledge gaps associated with psychosocial needs and outcomes of adulthood.

Scoping Review Question

What research on the psychosocial needs and outcomes of adults with SB has been published?

Methods

This review adhered to the JBI protocol for conducting a scoping review and followed the PRISMA-ScR checklist reporting guidelines.[44] An a priori protocol was previously published in JBI Evidence Synthesis.[43] The Covidence systematic review screening and data extraction platform was utilized to manage the data and document team member efforts.

Study Inclusion

The following bibliographic databases were used in this study: MEDLINE (Ovid), Embase (Elsevier), Cochrane CENTRAL (Wiley), CINAHL (EBSCO), Web of Science (Clarivate Analytics), PsycINFO (ProQuest), and ERIC (ProQuest). A medical librarian (LK) initially created a Medline search strategy using a combination of Medical Subject Headings (MeSH) and keywords for the concepts of spina bifida and adulthood (18 years and older). All team members reviewed the strategy and results to modify and improve the search strategy. With the approval of the team, the librarian customized the search using controlled vocabulary and keywords in the databases listed above (Appendix A displays the search strategy for this study). On February 20, 2018, all resulting citations were exported into an EndNote library (Clarivate Analytics) and duplicates were removed. No additional efforts were made to seek out grey literature. On March 20, 2023, the search was repeated in the same bibliographic databases to identify recently published studies.

Results

A total of 15,558 records were retrieved, 4923 of which were immediately identified as duplicates and removed. An additional 8117 records were removed after the title and abstract screening, leaving 2156, of which 2067 full-text articles were retrieved and reviewed. Following a full-text review, an additional 1986 articles were excluded due to the research outcome not being psychosocial ($n = 1190$), sample age not being adults, sample containing too few adults, or no separate analysis on the adult sample ($n = 255$), sample diagnosis not SB, sample composed of too few with SB diagnosis, or no separate analysis performed on those with SB ($n = 162$), articles written in language other than English ($n = 181$), the discovery of additional duplicate articles ($n = 114$), and others as grey literature or non-research studies ($n = 84$). This resulted in 81 research studies included in the review. Fig. 1 shows the adaptation of the PRISMA 2020 flow diagram for new systematic reviews, which included searches of databases and registers only.

Characteristics of included studies

All research studies (N = 81) were published between 1974 and 2023 in peer-reviewed journals from seven databases from the time of their inception as noted previously (Appendix A). As Fig. 2 shows, most studies ($n = 65$, 78%) were published in the past seventeen years of this review (2006–2023).

Prior to a 1993 United States study, all research ($n = 5$, 6%) in the first nineteen years of this review was conducted in the United Kingdom. In all years combined, over half ($n = 46$, 57%) of studies in this scoping review originated in the United States ($n = 34$, 42%) and the United Kingdom ($n = 11$, 14%) combined, with the plurality of psychosocial

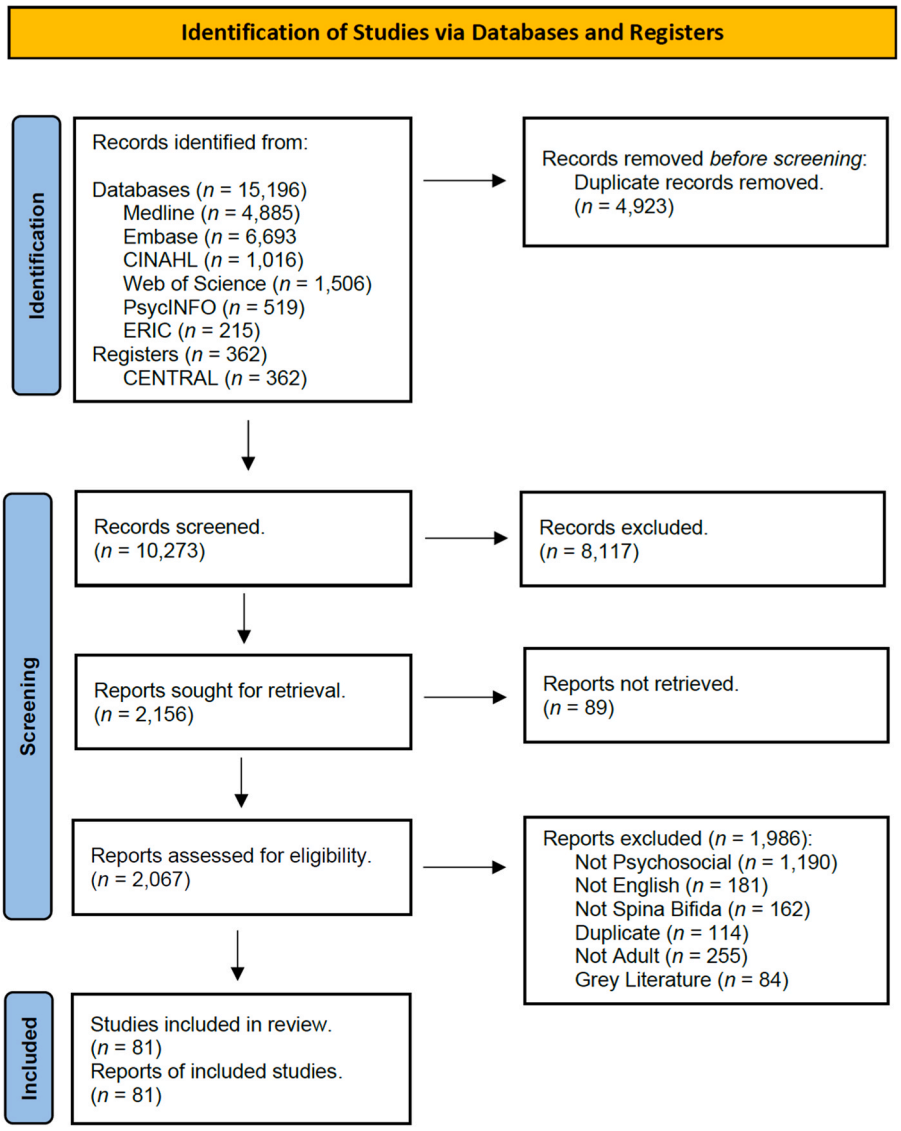

Fig. 1. (a) Selection of Sources of Evidence Flow Diagram created from PRISMA-ScR Checklist. (b) Adapted from Page et al.[45].

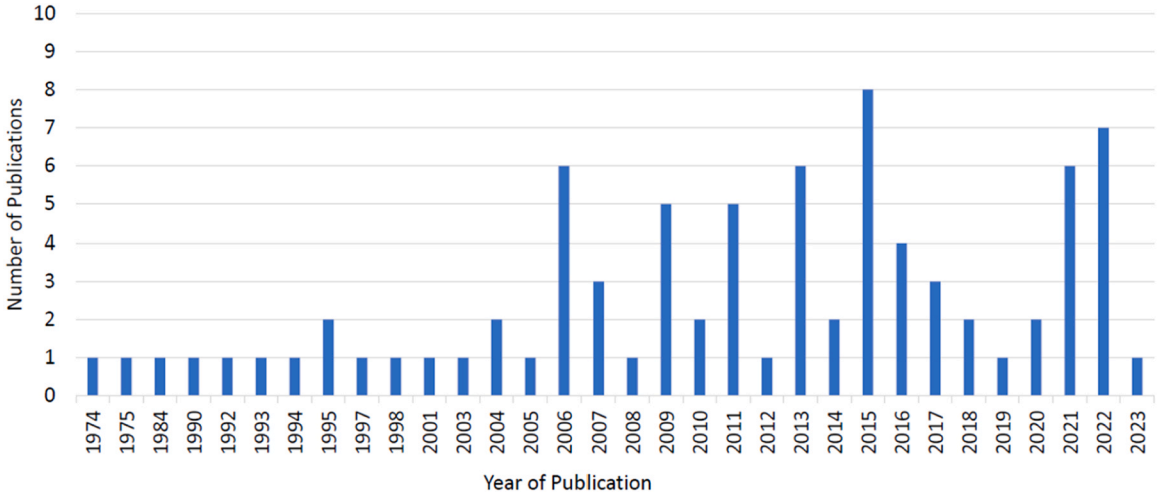

Fig. 2. Publications by Year (N = 81).

investigations still conducted in the United States. Several studies were produced in the Netherlands ($n = 9$, 11%), followed by Sweden ($n = 8$, 10%), Canada ($n = 6$, 7%), and Norway ($n = 5$, 6%). To a lesser extent, studies from South Korea ($n = 3$, 4%) and Australia ($n = 2$, 2%) were represented, and just one study each originated from Japan, France, and India (Appendix B). Of interest, the prevalence rate of SB in Korea is reported as 8.1 per 10,000[46] and 4.6 per 10,000 in Australia as compared to 3.6 per 10,000 in the United States.[33,47]

Over half ($n = 46$, 57%) of the reviewed research was written by interdisciplinary authorship. Just over half ($n = 41$, 51%) of the reviewed studies were funded. Of those funded, predominant support was by foundations ($n = 17$, 61%), followed by university and/or medical center funding ($n = 14$, 50%), and the remainder received government funding ($n = 9$, 32%).

The predominant research methods evident in the scoping review were quantitative ($n = 63$, 78%); notably, very few ($n = 4$, 6%) were intervention studies. Comparable small percentages of other research methods were qualitative ($n = 11$, 13%) and mixed methods ($n = 7$, 9%) (Appendix C). The majority were cross-sectional ($n = 70$, 86%), with the remainder being longitudinal designs ($n = 11$, 14%) (Appendix B).

Individuals with SB were the primary sources of information in 85% ($n = 69$) of the research studies. Other sources of information consisted of combinations of individuals with SB, along with their parents, caregivers, guardians, and/or their healthcare providers ($n = 11$, 14%); and two studies contained secondary data analyses of health care records (Appendix B).

Participants were predominantly recruited from a hospital, medical center, and clinic-type setting ($n = 54$, 67%). Other recruitment sites included community-based settings ($n = 5$, 6%), social media ($n = 4$, 5%), and condition-specific advocacy groups ($n = 3$, 4%). The remaining studies ($n = 15$, 19%) recruited participants from a combination of the previously mentioned sources (Appendix B).

Purpose Statements

Purpose statements of all retrieved studies were extracted and analyzed qualitatively using a content analysis approach.[48] Terms and related terms were coded as one concept. For example, purpose statements pertaining to participating in social activities, socializing, and/or socialization were coded as socialization. Similarly, purpose statements pertaining to independence and independent living were coded as independent living. Terms must have appeared in at least two purpose statements to be included. Statements were analyzed independently and then compared among team members. Results are graphically represented as a word cloud in Fig. 3.

Characteristics of Research Samples

All studies ($n = 81$) included in this review contained samples representing those with SB. Among them were forty-three studies in which the largest percent of the sample was identified with myelomeningocele. Other forms of SB identified were occulta, meningocele, and lipomeningocele. Additional relevant diagnostic information on hydrocephalus ($n = 42$, 52%) and shunt status (n = 51, 63%) was reported in many studies included in this scoping review (Appendix C; Table 1).

Apart from one study[49] gender was identified in the remaining investigations ($n = 80$, 99%). The race/ethnicity of the sample was reported in just about two thirds of the studies ($n = 50$, 62%); among these, 31% ($n = 16$) reported racially/ethnically diverse samples, meaning more than one race/ethnicity were included in the study

Table 1
Characteristics of Research Samples ($N = 81$).

Sample Characteristics Identified:		
Diagnostic Information		
Diagnosis Myelomeningocele	43	53%
Hydrocephalus Status	42	52%
Shunt Status	51	63%
Gender	80	99%
Race/Ethnicity	50	62%
Physiological Functioning		
Ambulatory Status	55	68%
Continence Status	50	62%
Reproductive Function	20	25%
Psychological Functioning and Disorders	41	51%
Community Integration		
Independence	56	69%
Productivity	35	43%
Inclusion	33	41%
Living Skills	30	37%
Housing Arrangements	41	51%
Employment	51	63%
Education Level	45	56%
Socioeconomic Level	20	25%

Note. Percentages do not equal 100 due to multiple constructs being explored in one or more studies.

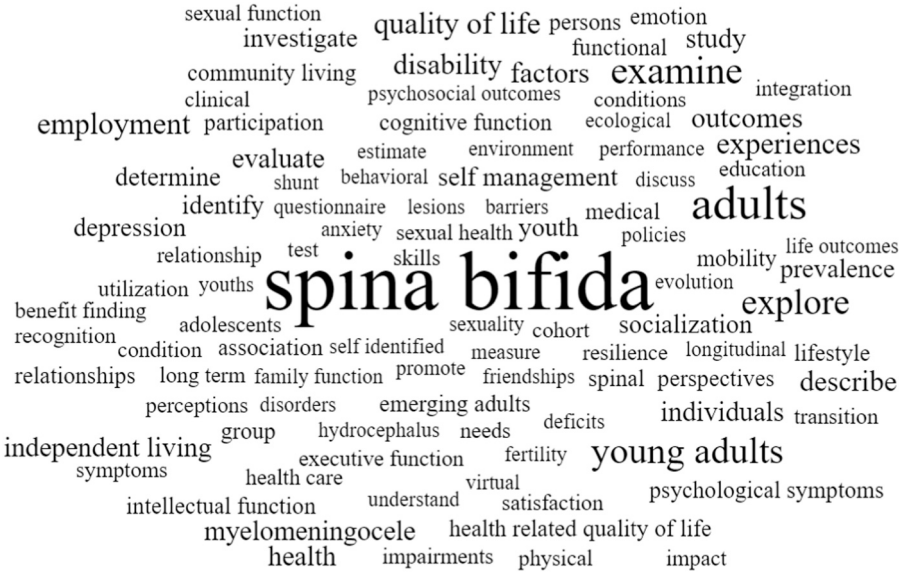

Fig. 3. Word Cloud Depicting Prominent Terms Used in Purpose Statements.

sample (Appendix C; Table 1).

Physiologic Functioning

Ambulatory status was the most reported physiologic characteristic ($n = 55$, 68%) referenced from the studies in this review. Instruments used to measure ambulatory status included the Hoffer Scale of Function Ambulation, which was used in two studies;[50,51] the Motor Independence Scale of the Scales of Independent Behavior–Revised (SIB-R) that was used in one study;[52] and most recently, the Timed Up and Go and Six-Minute Walk Test that were both used by Lidal et al.[53]. Continence status was reported in 62% ($n = 50$) of the investigations; of these, urologic status and management ($n = 45$, 90%) was reported more often than bowel status and management ($n = 38$, 76%). In addition to identifying whether urinary incontinence was present, a study by Khan et al.[54] explored the degree to which urinary incontinence was bothersome and the impact urinary incontinence had on life using the instruments Urogenital Distress Inventory, Incontinence Impact Questionnaire-7, and American Urological Association Symptom Index. Choi et al.[55] used the International Consultation on Incontinence Questionnaire[56] in their study of sexual function and quality of life among young men with SB. (Appendix C; Table 1).

Explorations of reproductive function were reported in 25% ($n = 20$) of the studies. Examples of tools discovered in this review that measured reproductive functioning included the Questionnaire for Screening Sexual Dysfunctions used by Vroege et al.;[57] the Watts Sexual Function Questionnaire used by Lassmann et al.;[58] the Female Sexual Function Index used by Lee et al.;[59] the International Index of Erectile Function used by Lee et al.[59] and Rosen et al.;[60] and the Sexual Health Inventory for Men, also known as the IIEF-5 questionnaire, used by Alwaal et al.,[61] Lee at al.,[59] and Rosen et al.[62] (Appendix C; Table 1).

Psychological Functioning and Disorders

Psychological functioning and disorders were explored in just over half ($n = 41$, 51%) of the studies in this review. Among these, 47% ($n = 24$) reported a mental health diagnosis; depression was the most common ($n = 15$, 63%), followed by anxiety ($n = 10$, 42%). Tools used to measure depression and anxiety were the Beck Depression Inventory,[63] the Beck Depression Inventory–II,[64] Patient Health Questionnaire-9,[65] and the Depression Anxiety Stress Scales-21 item that had been used in two studies.[54,66] Less frequently reported mental and emotional health concerns ($n = 15$) included feelings of dysphoria, sense of panic and helplessness, stress and emotional pressure, hostility, night-time fantasies of assaultive behavior, psychoticism, paranoid ideation, obsessive-compulsive disorder, and substance use disorder. The functional status of adults with intellectual or developmental disabilities was measured by Liptak et al.[67] using the Rochester Health Status Survey–IV,[68] and emotion recognition was measured by Stubberud[69] using the Reading the Mind in the Eyes Test (Appendix C; Table 1).

Community Integration

In the community context, independence was explored most often ($n = 56$, 69%), followed by feelings of productivity ($n = 35$, 43%), inclusion ($n = 33$, 41%), living skills ($n = 30$, 37%), and housing arrangements ($n = 41$, 51%; Appendix C, Table 1). The Craig Handicap Assessment and Reporting Technique[70] was utilized in four studies[63,64,66,71] to measure the domains of physical and cognitive independence, mobility, occupation, social integration, and economic self-sufficiency in adults 18 to 65 years. The Functional Independence Measure was used to assess functional independence within the domains of self-care, sphincter control, transfers, locomotion, communication, and social cognition in one study.[72] The Spinal Cord Independence Measure[73] was used by Coco et al.[74] to measure level of independence in three areas: self-care, respiration, and sphincter management mobility (need for external respiratory support and bowel and bladder management). Khan et al.[54] explored the extent of neurological disability involvement using Guy's Neurological Disability Scale as operationalized using 12 categories of functioning (i.e., swallowing, speech, cognition, mood, vision, upper limb function, lower limb function, bowel function, bladder function, sexual function, fatigue, and others). Four studies[75–78]—three of which were part of a larger longitudinal investigation—used the Adolescent Self-Management and Independence Scale II[79] to measure SB condition self-management and independence with community living skills (i.e., money management and the use of transportation).

Employment ($n = 51$) and education level ($n = 45$) were each assessed in more than half of the studies (63% and 56%, respectively). The least assessed characteristic was socioeconomic level ($n = 20$), having been assessed in only 26% of the studies in this review. Of the studies that explored education level, the majority identified post-secondary education ($n = 36$, 80%) and training ($n = 25$, 56%). (Appendix C; Table 1).

Scoping Review Psychosocial Findings

Characteristics of Psychosocial Outcomes

Psychosocial outcomes that were either quantitively measured or qualitatively explored across all 81 research studies included in this review were organized thematically into three domains: social, psychological, and cognitive (Appendix D; Table 2).

Social

Various types of relationships were reported in half the studies, ranging from those involving significant others, families, and peers, to those associated with the community, employment, and access to health care. The studies in this scoping review that explored social relationships most often reported relationships with significant others ($n = 39$, 48%); of note, 20% ($n = 16$) included the topic of parenthood. Exploration of relationships with co-workers and health care providers were reported the least often: $n = 9$ (11%) and $n = 10$ (12%), respectively. The construct of relationships was assessed using various methodologies. In one instance, it was a self-report on the number of friends.[71] In another,[80] social participation was assessed using the Assessment of Life Habits Questionnaire.[81] Sixteen studies explored the association of quality of life with satisfaction with social relationships.[50,52,58,71,75,76,82–91] Several different instruments were used to measure quality of life as described below, so the construct of type of relationship varied as

Table 2
Domains of Psychosocial Outcomes Explored Thematically ($n = 81$).

Social		
Significant Others	39	48%
Family Members	26	32%
Friends/Peers	25	31%
Community	20	25%
Parenthood	16	20%
Health Care Providers	10	12%
Coworkers	9	11%
Psychological		
Quality of Life	30	37%
Attitudes/Beliefs	15	18%
Coping	12	15%
Self-Determination	8	10%
Self-Efficacy	6	7%
Self-Esteem	3	4%
Cognitive		
Cognition/Intelligence	36	44%
Executive Function	14	17%

Note. Percentages do not equal 100 due to multiple constructs being explored in one or more studies.

well. For example, the Quality-of-Life Questionnaire contains items that provide scores on marital relations, parent–child relations, extended family relations, and extra-marital relations.[52] The World Health Organization Quality of Life Group's[92] Medical Outcomes Study 26-item Short Form questionnaire measures quality of life as it pertains to several domains, including social relationships. Sixteen studies examined the influence of bladder and bowel continence on a range of social activities, relationships, and sexual activity.[53,83,90,93–104] In all studies, continence was an obviously related, often inhibitory factor with the development of relationships. Marital status was reported in eighteen studies.[52,71,83–85,88,90,97,98,103–111] A few studies reported on sports involvement/exercise,[80,93,103] owning a car,[53,109] and church attendance and community cultural activity (i.e., museum, $n = 1$).[103]

Psychological

The extent to which psychological constructs were measured in studies of this scoping review was examined. The construct, quality of life, was explored in 37% of the studies ($n = 30$). This included all measures of quality of life, those broadly encompassing all aspects of life, as well as health-related quality of life that focuses on the impact of illness and treatment on one's quality of life. Quality of life as the construct of health-related quality of life was predominately quantified by the World Health Organization Quality of Life questionnaire,[71,75,76,78,89,112,130] and the Medical Outcomes Study Health Survey[55,58,80,83,87,91,113] in seven studies each ($n = 14$). These, and other quality of life tools used less often are cited in Appendix E. A survey questionnaire was used to collect quality of life data in two studies;[82,85] one study reported quality of life, but the measure was not identified.[114] Four studies used qualitative methods to collect data on quality of life (i.e., semi-structured questionnaire, focus group).[53,90,94,115]

A total of 15 (18%) studies explored participant attitudes and beliefs, 8 of which used validated instruments to explore participants' attitudes on a variety of topics,[80,86,87,95,108,109,116,117] and the remaining studies[53,90,91,94,97,118] used qualitative methods (i.e., semi-structured interviews and focus groups) to gather data on participants' attitudes. Other psychosocial constructs included coping ($n = 12$, 15%), wherein it was explored with the use of validated instruments in six studies.[54,66,80,86,102,113] The remaining studies used qualitative methods,[90,94,97,115] case study approach,[119] and a survey.[103] The other concepts included self-determination ($n = 8$, 10%), self-efficacy ($n = 6$, 7%), and self-esteem ($n = 3$, 4%); these concepts were reported in 10% and less of the studies reviewed. (Appendix D and E; Table 2).

Cognitive

Thirty-six (44%) studies reported level of cognition/intelligence; far fewer studies reported executive functioning ($n = 14$, 17%; Appendix D and E; Table 2). Twenty-one (26%) studies used validated instruments to measure intelligence and cognition. The Wechsler scales were most often used: the Wechsler Intelligence Scale for Adults–Revised[120,121] in four studies,[52,111,122,123] the Wechsler Abbreviated Scale of Intelligence[120,121,124] in three studies;[69,113,125] and the Wechsler Memory Scale[121,126] in one study.[128] The Raven (1996) Standard Progressive Matrices test was used in five studies;[50,104,127-129] the CHART-SF[70] was used in two studies.[64,130] Other tools used in individual studies were the Guy's Neurological Disability Scale,[131] Health Utilities Index–Mark III,[132] Mini Mental State Examination (for screening),[133] National Adult Reading Test,[134] Peabody Picture Vocabulary Test–Revised Edition,[135] Mann-Whitney U Test,[136] and Repeatable Battery for the Assessment of Neuropsychological Status.[137] Other measures of emotional and social intelligence used in individual studies included the Eyes Test of Emotional Judgment/Intelligence,[138] and the Functional Independence Measure[139] measured social cognition. Eleven studies reported using intelligence and competency scales to screen for study inclusion and exclusion criteria, although the tools used were not consistently identified. Fifteen studies referred to the level of intelligence or cognitive functioning in report of findings or in discussion, but no instruments for assessment were mentioned. The intelligence quotient was reported with the use of Rochester Health Status Survey IV (RHSS-IV)[68,140] used in one study to extract these data from health records.[67] Two studies assessed academic skills[52,122] using the Metaphorical Language Test & Wide Range Achievement Test-R, Reading, Spelling & Arithmetic[141] and the Kaufman Functional Academic Skills Test (K-FAST),[142] respectively. (Appendix E).

Eighteen instruments to measure executive functioning were reported in 11 studies as presented in Appendix E. The Wechsler Adult Intelligence Scale II was used in three studies[93,125,143] and Wisconsin Modified Card Sorting Test,[144] Delis-Kaplan Executive Function System,[145] Trail Making Test A and B,[146,147] Behavior Rating Inventory of Executive Function–Adult Version: BRIEF A[148] and Dysexecutive Questionnaire[149,150] were each used in two studies. The remaining number of validated measures of executive functioning were used in five studies. Executive functioning was referred to in the findings and discussion without identification of the instrumentation used for assessment purposes in two studies. Inclusion and exclusion criteria for study participation were determined with the administration of executive functioning assessment using validated instruments in three studies (Appendices E; Table 2).

Analysis of Scoping Outcomes Examined Over the Decades

Trends in the presentation of articles included in this scoping review over the decades were examined. These trends were investigated by the five decades of articles published that constituted the final sample of eligible articles. The category of trends reviewed includes the following: sample characteristics, mental health, community integration, employment and training, and the psychosocial outcomes in the domains of social, psychological, cognition. (Appendices B – D).

Selected Sample Characteristics

The trends in selected sample characteristics in over five decades of the review were examined. With two exceptions, males and females were identified in all studies of this review. Only males were investigated in studies published in 1992[119] and in 2017;[55] female-only samples were reported in 2018[83] and in 2021 studies;[88,90] gender was not identified in a 1984 study.[49] Other gender classifications were not reported. The ethnicity and race of samples were not identified in the first three decades of this review (1970–1999). Later, from 2000 to 2023, racial and ethnic characteristics of samples were presented in 16 (22%) of the studies (Appendix C). Sample characteristics, such as evidence of hydrocephalus and shunt, were identified infrequently in the first three decades of this scoping review. In studies published from 2000 onward, the trend revealed higher numbers and percentages of studies reporting diagnosis of hydrocephalus and presence of shunt; the reporting of these data was found in 52% ($n = 42$) and 63% ($n = 50$), respectively, of all the studies reviewed. The presence of shunt was reported more often than the diagnosis of hydrocephalus. As presented in Appendix C, ambulatory status was the most often identified sample characteristic. Urologic and bowel status and continence status were rarely identified in the earlier years of the review (1970–1999); the number/percentages of studies reporting this clinical profile increased from 2000 onward. Continence status and urologic status and management were identified in approximately half of the studies; bowel management was reported in slightly less than half of the studies of this review, as depicted in Appendix C.

Mental Health

Twenty-four (30%) of the investigations included in the scoping review presented findings pertaining to a specific mental health

symptomology of study participants, with just over half (51%) reporting on psychological functioning overall. Prior to 2000, very few studies reported issues related to mental health. During the period after 2000, that number increased to about one third of the studies included in this review. (Appendix C).

As this scoping review demonstrated, mental health metrics have generally been overlooked in research designs. Given the unique lifestyle challenges that individuals with spina bifida experience in their lived experience, exploration of their mental health concerns warrant inclusion in research designs.

Community Integration

Concepts pertaining to community integration and living were explored, including independence, productivity, inclusion, and living skills. The extent to which sample participants functioned independently in the community was reported more often ($n = 56$, 69%) compared to the other indicators of community integration. The other indices of community integration, productivity, and inclusion were reported in less than 50% of the studies. Living skills ($n = 30$, 37%) were detailed the least. The frequency was miniscule between 1970 and 1989, as less than 4% of the studies reported any indices of community integration. The trend was more evident from 2000 onward wherein more studies reported one or more elements of community integration (Appendix C).

Understanding of the extent to which individuals with spina bifida are integrated into the community is an essential psychosocial outcome associated with adulthood and warrant consideration in research designs. Concepts of independence, productivity and inclusion can be operationalized with a number of metrics such as living arrangements and community involvement. Recommendations for measurements are provided in the discussion.

Employment and Training

Employment ($n = 51$, 63%) was reported the most often compared to the other outcomes examined. Involvement in postsecondary training ($n = 25$, 31%) was reported least often in these studies. The trend of reports on these outcomes demonstrated that few studies explored these outcomes from 1970 to 1989. Starting in the 1990 s through 2023, there was a steady increase in the number of studies exploring adult outcomes (Appendix C). Although an increase in the exploration of employment is evidenced in later decades, the definition of employment was less clear. Further explorations of employment among adults with SB earning a customary rate received by similarly employed adults without disability is warranted.

Social

Over the decades, significant others were explored in 39 (48%) of studies reviewed. Social relationships that involved emotionally engaged relationships were reported in 30% or more in studies (i.e., significant others, family members, and friends/peers). Relationships that were associated with community, work, and health care settings were cited less often in the reviewed studies. Nine studies explored relationships with co-workers and ten with health care providers. The trend of reporting any type of relationships increased over the years from less than 10% in the 1970 s to just less than 30% of nearly all of studies from 2000 to 2023 (Appendix D).

Future research designs would benefit with an expanded framework to explore more in-depth information about acquisition of adult outcomes that include not only employment but other benchmarks of adulthood. Suggestions for expanding investigating exploration of adult psychosocial outcomes are provided in the discussion.

Psychological

Examination of psychological constructs revealed that quality of life was reported in 32 (39%) of the studies in this scoping review. Coping, attitudes and beliefs, and self-esteem were reported in less than 20% of the studies reported. A few studies investigated self-efficacy and self-determination; these studies were conducted from 2010 to 2023. A small number of studies from 1970 to 1999 used measures to explore coping, attitudes and beliefs, and self-esteem. It was not until 2000 that quality of life was examined in the studies of this scoping review. As demonstrated by the frequencies and percentages of the studies published from 2000 to 2023, relatively few studies explored psychological constructs (Appendix D).

It is imperative that studies examining psychosocial outcomes include measurements with strong psychometric properties. Use of measures lacking measures of validity and reliability call into question the trustworthiness of the study findings. Furthermore, use of valid and reliable measurements enable comparison and contrasts with not only investigations conducted with individuals with spina bifida but with other adult populations with disabilities. Use of psychological constructs in research designs serve to strengthen the quality of evidence to inform practice and future research.

Cognition

A total of 36 (44%) investigations reported the cognition/intelligence level of the sample. Just two studies published from 1970 to 1989 explored cognitive functioning. Cognitive status was reported more often from 1990 onward. Relatively few studies explored executive functioning. It was not reported until the third decade (1990–1999) of this review when only two investigations presented findings on executive function (Appendix D).

Studies would benefit with the inclusion of measures of cognitive functioning. Measurement of this important variable would enable additional analysis of the association of level of cognitive function and acquisition of adult psychosocial outcomes. Enlarged insights associated with cognitive function would provide potential prescriptive for long-term service planning to optimize outcomes for adults with spina bifida.

Discussion

As the findings of this scoping review revealed, psychosocial outcomes were rarely investigated from the earliest article published in 1974 through the decades of the 1990 s. Beginning in 2000, as evidenced in this review, psychosocial constructs are slowly being introduced into research investigations. A modest collection of studies has reported the consequential metrics associated with adult outcomes of employment, housing, postsecondary education and training and social relationships despite the ongoing reported advances in survival rates of individuals with SB. The disproportionate ratio of studies exploring psychosocial outcomes compared to clinical investigations can be attributed to several factors. The survival rates during the early period of this review were grim compared to currently reported rates of long-term survival.[151,153] Technological improvements and advances in treatment approaches have been critical to improving the survival rates of individuals with SB. Heretofore, survival in the 1970 s and 1980 s were limited to late adolescence and early adulthood.[154] Since then, the survival rates have continued to improve; current estimates report survival rates, depending on condition severity and involvement, to the mid-40 s[110,111,155] Hence, the earlier literature was aligned with fewer feasible areas of study pertaining to psychosocial outcomes. Nevertheless, despite improvements in survival rates, psychosocial literature has lagged behind the progress reported with medical treatments and scientific developments. Relevant to conducting investigations of psychosocial outcomes would be the necessity of access to adults with SB, which would be both a challenge in terms of actual sample sizes

powered sufficiently for analysis and ability to track individuals with SB who entered systems of adult care with assignment of new identifiers that are not matched with prior identifiers used in pediatric care.

Research sample and sample characteristics

One interesting finding was that symptomology fundamentally associated with SB was inconsistently reported in the studies of this scoping review. Over half of the studies reported the accompanying diagnosis of hydrocephalus and the presence of a shunt. Coupled with inconsistent reporting, less than half of studies reported the level of cognitive functioning, and relatively few studies disclosed information about executive functioning. In addition, a limited number of studies included the use of well-established instruments to measure the level of intelligence; eleven investigations referred to the IQ levels of participants without reference to the measurement used. Nevertheless, there is extensive literature that individuals with SB, hydrocephalus, and shunts are at greater risk for cognitive challenges that include lower IQ levels and problems with executive functioning. The variables as predictors of psychosocial outcomes were not extensively reported; however, more often, the presence of hydrocephalus and shunt status were presented as part of the descriptive profile of the sample.

Increased attention in health care in the United States and other countries around the world has focused on social determinants of health and their impact on health equity. The World Health Organization[152] defined *social determinants of health* as "the conditions in which people are born, grow, work, live, and age or the non-medical factors that influence health outcomes." Healthy People 2030[156] has grouped these social determinants of health into five domains: economic stability, education access and quality, health care access and quality, neighborhood and built environment, and social and community content. These social determinants of health have direct links to health inequities, health disparities, and poorer health outcomes for those underserved or marginalized populations. Organizations, including Healthy People 2030, Centers for Medicare and Medicaid Services, Robert Wood Johnson Foundation, Institute for Healthcare Improvement, among many others, are developing health equity frameworks as guides for healthcare providers and organizations to assist in achieving health equity for all clients. Braveman et al.[157] defined *health equity* in the following way:

> Everyone has a fair and just opportunity to be as healthy as possible. This requires removing obstacles to health such as poverty, discrimination, and their consequences, including powerlessness and lack of access to good jobs with fair pay, quality education and housing, safe environments, and health care.

Included in the definition of *health equity* is the requirement of addressing the social determinants of health to achieve quality health care services and health for all. Many of the recommendations from these organizations include incorporating diversity in research.

The National Institute on Minority Health and Health Disparities[158] discussed in depth the importance of including the social determinants of health in research, with emphasis on the inclusion in clinical trials. They published the following statement:

> To account for the diverse lived experiences and exposures of various populations, clinical trials must be appropriately inclusive of racial and ethnic minority groups, as well as other populations experiencing health disparities, including sexual and gender minority or socioeconomically disadvantaged populations.

In this scoping review, race or ethnicity of the sample was reported in over half of the studies; however, among these, only a handful reported racially or ethnically diverse samples. The research sample characteristics from this scoping review demonstrated little focus on social determinants of health, including post-secondary education, post-secondary training, employment, and housing arrangements. The least assessed social determinant of health characteristic was socioeconomic levels, which was reported in only a few studies.

The predominant research method evident in the scoping review was quantitative. Just over 86% of the studies used cross-sectional designs; apart from one study published in 1995, longitudinal designs were not evident until the last decade of this review. Of these eleven studies, data reported in three, were generated from the same longitudinal data set. The relatively few longitudinal studies of this scoping review likely reflect the difficulties associated with long-term tracking of individuals with SB who exit the pediatric system of care and enter the adult system of care in the United States. Although seven of the eleven studies originated in the United States, few states assign an identifier to enrollees that enable long-term follow-up across systems of care. The data infrastructure of countries with national health care systems lends itself to more feasible longitudinal tracking. Equally compelling, of the quantitative studies, only four (6%) introduced an intervention, which were published in the last years of this review (2009–2017). The need for experimental designs is warranted, allowing for increased understanding of strategies and efficacy of treatments that work best to improve outcomes for this population. Lacking evidence to improve psychosocial outcomes results in delays in changes necessary to improve education, policy, and practice.

As has been demonstrated in this scoping review, there is a dearth of evidence on psychosocial outcomes of adults with SB. It is relevant, if not of the utmost importance, to conduct studies that test interventions and investigate the lived experience outcomes of adults with SB pertaining to long-term outcomes in postsecondary education, employment, community living, social relationships, and quality of life that will inform practice, research, and policy making leading to improved psychosocial outcomes.

Based upon the aforementioned analysis of psychosocial outcomes, future research would be enhanced with more in-depth exploration of the psychosocial outcomes beyond the limitation range of outcome queries reported in this scoping review. An improved understanding of employment status would include items that gather data on hours worked per week (part-time, full time); accommodations needed in the worksite; type of employment; type of workplace setting (i.e., inclusive, supported employment); employment supports/services accessed (i.e., vocational rehabilitation, job corps).

Educational outcomes investigated can be expanded to explore types of postsecondary program enrollment (i.e., vocational training, college); highest level of education achieved, degree type/focus; access to Disabled Student Services; types of academic and health-related accommodations received; and financial assistance received. Exploration of community living circumstances would provide additional context as to fuller understanding of long-term adult outcomes and quality of life indices. Gathering data of community mobility would provide insights as to inclusive community access such as driving a car, use of paratransit and public transportation; and reliance on friends/family for transportation needs. Expanding the options available to have more informed perspectives of living arrangements would include a broader range such as living independently, living with spouse/significant other, living with parents/family member/roommate. To better understand the profile of social relationships warrants gathering data on social networks that includes number of friends, frequency of social contacts and characteristics of social contacts (i.e., in person, social media, via phone). Lastly recreational involvement is an important metric that reflects community engagement. The barriers the are encountered and experienced in the achievement of psychosocial outcomes need to be explored as well.

Expanding the scope of long-term data on adults of individuals with spina bifida will serve to better inform the spina bifida community on implementation of service models that enhance the acquisition of psychosocial benchmarks of adulthood. This scoping review provides foundational insights and understanding that will serve to provide direction for spina bifida practice and serve as an impetus to conduct studies that focus on psychosocial outcomes for adults.

Conclusion

In the period 1974 through 2023, over 10,000 records meeting the search criteria were screened, leaving 81 research studies that met the inclusion criteria. As the research was examined, gaps were evident in the presentation of information, and a lack of clarity was noted as it pertained to concepts studied. Absence of information pertaining to benchmarks of adulthood for adults with SB as it relates to education, employment, community living, and social relationships were evident. These indices lag behind the psychosocial literature in general. As medical advances continue to improve the survival rates of individuals with SB, more research pertaining to this population, their psychosocial needs, and related outcomes is requisite to capturing the progress of this adult population.

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This paper follows the ethical guidelines put forth by this journal. It is a product of the coauthors' original work and reflects their own review and analysis of the literature in a truthful and complete manner. All authors have been substantially involved in the work leading to the creation of this paper and take responsibility for the content. This paper has not been submitted elsewhere for publication.

CRediT authorship contribution statement

Jennifer Emilie Mannino: Conceptualization, Methodology, Investigation, Formal Analysis, Data Curation, Writing - Original Draft, Writing - Review & Editing. **Heather Reens:** Investigation, Data curation, Visualization, Writing- Original Draft, Writing – Review & Editing. **Kathryn Smith:** Conceptualization, Investigation, Writing – Review & Editing. **Lynn Kysh:** Investigation, Formal Analysis, Data Curation, Writing - Original Draft. **Serge' R. Nelson:** Investigation, Writing – Review & Editing. **Yuding Wang:** Investigation, Writing – Review & Editing. **Manu Raam:** Writing – Review & Editing. **Michele Roland:** Investigation, Writing – Review & Editing. **Alexander Van Speybroeck:** Investigation, Writing – Review & Editing. **Cecily L. Betz:** Supervision, Conceptualization, Methodology, Investigation, Formal Analysis, Data Curation, Writing - Original Draft, Writing - Review & Editing

Declaration of Competing Interest

The authors declare the following financial interests/personal relationships which may be considered potential competing interests. Cecily L. Betz, PhD, RN, FAAN, coauthor of this article and the Editor-in-Chief of Health Care Transitions, has not been involved in the management or review of this article.

Data availability

Data will be made available on request.

Appendices A-E. Supporting information

Supplementary data associated with this article can be found in the online version at doi:10.1016/j.hctj.2024.100041.

References

*Indicates studies included in this review

1. Cheng L, Shaewitz D. *The 2021 Youth Transition Report: Outcomes for Youth and Young Adults with Disabilities*. Washington, DC: Institute for Educational Leadership; 2021.
2. Child and Adolescent Health Measurement Initiative. *National Survey of Children's Health 2016-2017 data query*. Baltimore, MD: Data Resource Center for Child and Adolescent Health; 2019.
3. Erickson W. *Estimate of disability statistics for youth and young adults based on analysis of 2019 American Community Survey Public Use Microdata Sample. Special tabulations prepared by William Erickson, Yang-Tan Institute*. Cornell University; 2022.
4. Park S. *Estimate of young adult chronic condition prevalence based on analysis of the 2017 Behavioral Health Risk Surveillance System. Special tabulations prepared by Sally Park*. San Francisco: University of California; 2019.
5. Census Bureau US. *Annual estimates of the resident population by single year of age and sex for the United States*. April 1, 2010, to July 2018. Washington, DC: US Census Bureau,; 2019.
6. Census Bureau US. *Current Population Survey, Annual Social and Economic Supplement, 2019*. Washington, DC: US Census Bureau; 2020.
7. Glinianaia SV, Morris JK, Best KE, et al. Long-term survival of children born with congenital anomalies: A systematic review and meta-analysis of population-based studies. e1003356. 2020 Sep 28 *PLoS Med*. 2020;17(9). https://doi.org/10.1371/journal.pmed.1003356.
8. Mukherjee S. Transition to adulthood in spina bifida: changing roles and expectations. Published 2007 Nov 26. doi *Sci World J*. 2007;7:1890–1895. https://doi.org/10.1100/tsw.2007.179.
9. Shin M, Kucik JE, Siffel C, et al. Improved survival among children with spina bifida in the United States. *J Pedia*. 2012;161(6):1132–1137. https://doi.org/10.1016/j.jpeds.2012.05.040.
10. Tennant PW, Pearce MS, Bythell M, Rankin J. 20-year survival of children born with congenital anomalies: a population-based study. *Lancet*. 2010;375(9715):649–656. https://doi.org/10.1016/S0140-6736(09)61922-X.
11. Bamer AM, Connell FA, Dudgeon BJ, Johnson KL. Frequency of purchase and associated costs of assistive technology for Washington State Medicaid program enrollees with spina bifida by age. *Disabil Health J*. 2010;3(3):155–161. https://doi.org/10.1016/j.dhjo.2009.10.009.
12. Liptak GS, Garver K, Dosa NP. Spina bifida grown up. *J Dev Behav Pedia*. 2013;34(3):206–215. https://doi.org/10.1097/DBP.0b013e31828c5f88.
13. Ouyang L, Grosse SD, Armour BS, Waitzman NJ. Health care expenditures of children and adults with spina bifida in a privately insured U.S. population. *Birth Defects Res A Clin Mol Teratol*. 2007;79(7):552–558. https://doi.org/10.1002/bdra.20360.
14. Ouyang L, Grosse SD, Thibadeau J, Swanson M, Campbell VA. Outpatient medical conditions among children and adults with spina bifida in the United States: frequency and expenditures. *J Pedia Rehabil Med*. 2010;3(3):177–185. https://doi.org/10.3233/PRM-2010-0127.
15. Webb TS. Optimizing health care for adults with spina bifida. *Dev Disabil Res Rev*. 2010;16(1):76–81. https://doi.org/10.1002/ddrr.99.
16. Oakeshott P, Hunt GM, Poulton A, Reid F. Expectation of life and unexpected death in open spina bifida: a 40-year complete, non-selective, longitudinal cohort study. *Dev Med Child Neurol*. 2010;52(8):749–753. https://doi.org/10.1111/j.1469-8749.2009.03543.x.
17. Oakeshott P, Reid F, Poulton A, Markus H, Whitaker RH, Hunt GM. Neurological level at birth predicts survival to the mid-40s and urological deaths in open spina bifida: a complete prospective cohort study. *Dev Med Child Neurol*. 2015;57(7):634–638. https://doi.org/10.1111/dmcn.12698.
18. Armour BS, Ouyang L, Thibadeau J, Grosse SD, Campbell VA, Joseph D. Hospitalization for urinary tract infections and the quality of preventive health care received by people with spina bifida. *Disabil Health J*. 2009;2(3):145–152. https://doi.org/10.1016/j.dhjo.2009.02.001.
19. Caterino JM, Scheatzle MD, D'Antonio JA. Descriptive analysis of 258 emergency department visits by spina bifida patients. *J Emerg Med*. 2006;31(1):17–22. https://doi.org/10.1016/j.jemermed.2005.09.005.
20. Dicianno BE, Wilson R. Hospitalizations of adults with spina bifida and congenital spinal cord anomalies. *Arch Phys Med Rehabil*. 2010;91(4):529–535. https://doi.org/10.1016/j.apmr.2009.11.023.
21. Domino JS, Lundy P, Glynn EF, Partington M. Estimating the prevalence of neurosurgical interventions in adults with spina bifida using the Health Facts data set: implications for transition planning and the development of adult clinics, 2021 Dec 24 *J Neurosurg Pediatr*. 2021;29(4):371–378. https://doi.org/10.3171/2021.10.PEDS21293.
22. Dunbar P, Hall M, Gay JC, et al. Hospital readmission of adolescents and young adults with complex chronic disease. Published 2019 Jul 3 *JAMA Netw Open*. 2019;2(7), e197613. https://doi.org/10.1001/jamanetworkopen.2019.7613.
23. Inouye BM, Jiang R, Alkazemi MH, et al. Hospital and ED charges for spina bifida care in the United States between 2006 and 2014: Over $2 billion annually. *Disabil Health J*. 2019;12(3):431–436. https://doi.org/10.1016/j.dhjo.2019.01.007.
24. Kinsman SL, Doehring MC. The cost of preventable conditions in adults with spina bifida. *Eur J Pedia Surg*. 1996;6(Suppl 1):17–20. https://doi.org/10.1055/s-2008-1071031.
25. Lundberg Larsen K, Maalen-Johansen IK, Rennie L, Lidal IB. Gait function in adults aged 50 years and older with Spina Bifida. *Arch Phys Med Rehabil*. 2021;102(4):702–708. https://doi.org/10.1016/j.apmr.2020.10.118.
26. Young NL, Anselmo LA, Burke TA, McCormick A, Mukherjee S. Youth and young adults with spina bifida: their utilization of physician and hospital services. *Arch Phys Med Rehabil*. 2014;95(3):466–471. https://doi.org/10.1016/j.apmr.2013.09.015.

27. Piatt Jr JH. Adults with myelomeningocele and other forms of spinal dysraphism: hospital care in the United States since the turn of the millennium. *J Neurosurg Spine*. 2016;25(1):69–77. https://doi.org/10.3171/2015.9.SPINE15771.
28. Wang Y, Ouyang L, Dicianno BE, et al. Differences in length of stay and costs between comparable hospitalizations of patients with Spina Bifida with or without pressure injuries. *Arch Phys Med Rehabil*. 2019;100(8):1475–1481. https://doi.org/10.1016/j.apmr.2018.12.033.
29. Wang HH, Wiener JS, Ross SS, Routh JC. Emergent care patterns in patients with spina bifida: a case-control study. *J Urol*. 2015;193(1):268–273. https://doi.org/10.1016/j.juro.2014.06.085.
30. Hilberink SR, van der Slot WMA, Klem M. Health and participation problems in older adults with long-term disability. *Disabil Health J*. 2017;10(2):361–366. https://doi.org/10.1016/j.dhjo.2016.12.004.
31. Loftus CJ, Moore DC, Cohn JA, et al. Postoperative complications of patients with Spina Bifida undergoing urologic laparotomy: A multi-institutional analysis. *Urology*. 2017;108:233–236. https://doi.org/10.1016/j.urology.2017.06.019.
32. Mann JR, Royer JA, Turk MA, et al. Inpatient and emergency room visits for adolescents and young adults with spina bifida living in South Carolina. *PM R*. 2015;7(5):499–511. https://doi.org/10.1016/j.pmrj.2014.11.011.
33. National Center on Birth Defects and Developmental Disabilities, Centers for Disease Control and Prevention (September 3, 2020). National Spina Bifida Patient Registry Scientific Publications Retrieved on January 27, 2023, from https://www.cdc.gov/ncbddd/spinabifida/nsbpr-registry-publications.html.
34. Peyronnet B, Gao F, Brochard C, et al. Urologic disorders are still the leading cause of in-hospital death in patients with Spina Bifida. *Urology*. 2020;137:200–204. https://doi.org/10.1016/j.urology.2019.11.006.
35. Werhagen L, Gabrielsson H, Westgren N, Borg K. Medical complication in adults with spina bifida. *Clin Neurol Neurosurg*. 2013;115(8):1226–1229. https://doi.org/10.1016/j.clineuro.2012.11.014.
36. Castillo J, Lupo PJ, Tu DD, Agopian AJ, Castillo H. The National Spina Bifida patient registry: a decade's journey. *Birth Defects Res*. 2019;111(14):947–957. https://doi.org/10.1002/bdr2.1407.
37. Patel SK, Staarmann B, Heilman A, Mains A, Woodward J, Bierbrauer KS. Growing up with spina bifida: bridging the gaps in the transition of care from childhood to adulthood. *Neurosurg Focus*. 2019;47(4), E16. https://doi.org/10.3171/2019.7.FOCUS19441.
38. Bradstreet LE, Ludwig N, Koterba C, Zabel TA, Wilson CS. Supporting the transition to adulthood for youth with Spina Bifida: a call for neuropsychology-informed interventions. *Top Spinal Cord Inj Rehabil*. 2022;28(3):59–62. https://doi.org/10.46292/sci21-00096.
39. Scarberry KA, Gor RA, Kovell RC. Management of the transitional urology patient: the role of the adult reconstructive urologist. *Curr Urol Rep*. 2021;22(3):15. https://doi.org/10.1007/s11934-021-01035-z.
40. Roth JD, Szymanski KM, Ferguson EJ, Cain MP, Misseri R. Transitioning young adults with neurogenic bladder-Are providers asking too much? *J Pedia Urol*. 2019;15(4):384.e1–384.e6. https://doi.org/10.1016/j.jpurol.2019.04.013.
41. Shepard CL, Doerge EJ, Eickmeyer AB, Kraft KH, Wan J, Stoffel JT. Ambulatory care use among patients with Spina Bifida: Change in care from childhood to adulthood. *J Urol*. 2018;199(4):1050–1055. https://doi.org/10.1016/j.juro.2017.10.040.
42. Roth JD, Szymanski KM, Cain MP, Misseri R. Factors impacting transition readiness in young adults with neuropathic bladder. *J Pedia Urol*. 2020;16(1):45.e1–45.e7. https://doi.org/10.1016/j.jpurol.2019.10.017.
43. Betz CL, Smith KA, Kysh L, et al. Psychosocial outcomes for adults with spina bifida: a scoping review protocol. *JBI Evid Synth*. 2020;18(5):1135–1143. https://doi.org/10.11124/JBISRIR-D-19-00072.
44. Peters MDJ, Godfrey C, McInerney P, Baldini Soares C, Khalil H, Parker D. Chapter 11: Scoping Reviews. In: Aromataris E, Munn Z, eds. *Joanna Briggs Institute Reviewer's Manual*. JBI; 2017.
45. Page MJ, McKenzie JE, Bossuyt PM, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *BMJ*. 2021;372:n71. https://doi.org/10.1136/bmj.n71.
46. Ko JK, Lamichhane DK, Kim HC, Leem JH. Trends in the prevalences of selected birth defects in Korea (2008-2014), 2018 May 5 *Int J Environ Res Public Health*. 2018;15(5):923. https://doi.org/10.3390/ijerph15050923.
47. Mai CT, Isenburg JL, Canfield MA, et al. National population-based estimates for major birth defects. *2010-2014 Birth Defects Res*. 2019;111(18):1420–1435. https://doi.org/10.1002/bdr2.1589.
48. Kyngäs H, Mikkonen K, Kääriäinen M. *The application of content analysis in nursing science research*. Springer; 2020.
49. *Lonton AP, Loughlin AM, O'Sullivan AM. The employment of adults with spina bifida. *Z Kinder*. 1984;39(Suppl 2):132–134, 1984.
50. *Barf HA, Post MW, Verhoef M, Jennekens-Schinkel A, Gooskens RH, Prevo AJ. Life satisfaction of young adults with spina bifida. *Dev Med Child Neurol*. 2007;49(6):458–463, 2007.
51. *Barf HA, Post MW, Verhoef M, Jennekens-Schinkel A, Gooskens RH, Prevo AJ. Restrictions in social participation of young adults with spina bifida. *Disabil Rehabil*. 2009;31(11):921–927, 2009.
52. *Hetherington R, Dennis M, Barnes M, Drake J, Gentili F. Functional outcome in young adults with spina bifida and hydrocephalus. *Childs Nerv Syst*. 2006;22(2):117–124. https://doi.org/10.1007/s00381-005-1231-4. Epub 2005 Sep 17.
53. *Lidal IB, Lundberg Larsen K, Hoff M. 50 Years and older - born with spina bifida: participation, health issues and physical function. *Disabil Rehabil*. 2021;43(2):241–250. https://doi.org/10.1080/09638288.2019.1621953.
54. *Khan F, Amatya B, Ng L, Galea M. Rehabilitation outcomes in persons with spina bifida: a randomised controlled trial. *J Rehabil Med*. 2015;47(8):734–740. https://doi.org/10.2340/16501977-1999.
55. *Choi EK, Ji Y, Han SW. Sexual function and quality of life in young men with Spina Bifida: Could it be neglected aspects in clinical practice? *Urology*. 2017;108:225–232. https://doi.org/10.1016/j.urology.2016.11.052. Epub 2017 Mar 31.
56. Avery K, Donovan J, Peters TJ, Shaw C, Gotoh M, Abrams P. ICIQ: a brief and robust measure for evaluating the symptoms and impact of urinary incontinence. *Neurourol Urodyn*. 2004;23(4):322–330. https://doi.org/10.1002/nau.20041.
57. *Vroege JA, Zeijlemaker BYW, Scheers MM. Sexual functioning of adult patients born with meningomyelocele. *Eur Urol*. 1998;34(1):25–29, 1998.
58. *Lassmann J, Garibay Gonzalez F, Melchionni JB, Pasquariello Jr PS, Snyder 3rd HM. Sexual function in adult patients with spina bifida and its impact on quality of life (Oct) *J Urol*. 2007;178(4 Pt 2):1611–1614. https://doi.org/10.1016/j.juro.2007.03.162.
59. *Lee NG, Andrews E, Rosoklija I, et al. The effect of spinal cord level on sexual function in the spina bifida population. *J Pedia Urol*. 2015;11(3):142. e1-6.
60. Rosen R, Riley A, Wagner G, et al. The International Index of Erectile Function (IIEF): A multidimensional scale for assessment of erectile dysfunction. *Urology*. 1997;49:822–830.
61. Alwaal A, Awad M, Boggs N, et al. Sexual health inventory for men questionnaire as a screening method for erectile dysfunction in a general urology clinic. *Sex Med*. 2020;8:660–663.
62. Rosen RC, Cappelleri JC, Smith MD. Development and evaluation of an abridged, 5-item version of the International Index of Erectile Function (IIEF-5) as a diagnostic tool for erectile dysfunction. *Int J Impot Res*. 1999;11:319–326.
63. *Dicianno BE, Gaines A, Collins DM, Lee S. Mobility, assistive technology use, and social integration among adults with spina bifida. *Am J Phys Med Rehabil*. 2009;88(7):533–541. https://doi.org/10.1097/PHM.0b013e3181aa41d4.
64. *Dicianno BE, Kinback N, Bellin MH, et al. Depressive symptoms in adults with spina bifida (Aug) *Rehabil Psychol*. 2015;60(3):246–253. https://doi.org/10.1037/rep0000044.
65. *Bulloch AG, Fiest KM, Williams JV, et al. Depression—a common disorder across a broad spectrum of neurological conditions: a cross-sectional nationally representative survey. *Gen Hosp Psychiatry*. 2015;37(6):507–512, 2015.
66. *Hayter MR, Dorstyn DSResilience. self-esteem and self-compassion in adults with spina bifida. *Spinal Cord*. 2014;52(2):167–171. https://doi.org/10.1038/sc.2013.152.
67. *Liptak GS, Robinson LM, Davidson PW, et al. Life course health and healthcare utilization among adults with spina bifida. *Dev Med Child Neurol*. 2016;58(7):714–720, 2016.
68. Davidson PW, Henderson CM, Janicki MP, et al. Ascertaining health-related information on adults with intellectual disabilities: development and field testing of the Rochester Health Status Survey. *J Policy Pract Intellect Disabil*. 2008;5:12–23. https://doi.org/10.1111/J.1741-1130.2007.00134.
69. *Stubberud J. Theory of mind in spina bifida: relationship with intellectual and executive functioning. *Scand J Psychol*. 2017;58(5):379–388, 2017.
70. Mellick D.: The Craig Handicap Assessment and Reporting Technique—Short Form. The Center for Outcome Measurement in Brain Injury. Available at: http://www.tbims.org/combi/chartsf. 2000. (Accessed 13 May 2009).
71. *Chan WM, Dicianno BE. *Virtual Social adults spina Bifida Pm R*. 2011;3(3):219–225.
72. *Verhoef M, Barf HA, Post MW, van Asbeck FW, Gooskens RH, Prevo AJ. Functional independence among young adults with spina bifida, in relation to hydrocephalus and level of lesion. *Dev Med Child Neurol*. 2006;48(2):114–119, 2006.
73. Catz A, Itzkovich M, Agranov E, Ring H, Tamir A. SCIM–spinal cord independence measure: a new disability scale for patients with spinal cord lesions. *Spinal Cord*. 1997;35(12):850–856. https://doi.org/10.1038/sj.sc.3100504.
74. *Coco CT, Meenakshi-Sundaram B, Eldefrawy A, et al. A cross sectional single institution study of quality of life in adult patients with spina bifida. *Neurourol Urodyn*. 2018;14:14, 2018.
75. *Bellin MH, Dicianno BE, Levey E, et al. Interrelationships of sex, level of lesion, and transition outcomes among young adults with myelomeningocele. *Dev Med Child Neurol*. 2011;53(7):647–652. https://doi.org/10.1111/j.1469-8749.2011.03938.x. Epub 2011 Mar 17.
76. *Bellin MH, Dicianno BE, Osteen P, et al. Family satisfaction, pain, and quality-of-life in emerging adults with spina bifida: a longitudinal analysis. *Am J Phys Med Rehabil*. 2013;92(8):641–655. https://doi.org/10.1097/PHM.0b013e31829b4bc1.
77. *Bellin MH, Dosa N, Zabel TA, Aparicio E, Dicianno BE, Osteen P. Self-management, satisfaction with family functioning, and the course of psychological symptoms in emerging adults with spina bifida. *J Pedia Psychol*. 2013;38(1):50–62, 2013.
78. *Dicianno BE, Fairman AD, McCue M, et al. Feasibility of using mobile health to promote self-management in Spina Bifida. *Am J Phys Med Rehabil*. 2016;95(6):425–437. https://doi.org/10.1097/PHM.0000000000000400.
79. Sawin KJ, Heffelfinger A, Cashin SE, Brei TJ. The development of the adolescent/young adult self-management and independence scale ii: psychometric data. *J Pediatr Rehabil Med*. 2018;11(4):311–322. https://doi.org/10.3233/prm-170479.
80. *Buffart LM, van den Berg-Emons RJ, van Meeteren J, Stam HJ, Roebroeck ME. Lifestyle, participation, and health-related quality of life in adolescents and young adults with myelomeningocele. *Dev Med Child Neurol*. 2009;51(11):886–894, 2009.
81. Fougeyrollas P., Noreau L., St-Michel G. Life Habits measure – shortened version (LIFE-H 3.0). Lac St-Charles, Quebec, Canada: COCIDIH, 2001.

82. *Aguilera AM, Wood DL, Keeley C, James HE, Aldana PR. Young adults with spina bifida transitioned to a medical home: A survey of medical care in Jacksonville. *Fla J Neurosurg: Pediatr*. 2016;17(2):203–207.
83. *Choi EK, Kim SW, Ji Y, Lim SW, Han SW. Sexual function and quality of life in women with spina bifida: Are the women with spina bifida satisfied with their sexual activity? *Neurourol Urodyn*. 2018;37(5):1785–1793. https://doi.org/10.1002/nau.23525.
84. *Cope H, McMahon K, Heise E, et al. Outcome and life satisfaction of adults with myelomeningocele. *Disabil Health J*. 2013;6(3):236–243. Disabil Health J 2013;6(3):236-43.
85. *Kasai K, Tomoya U, Fujioka Hiroshi. Sex education needs of Japanese people with Spina Bifida: Relation to participants' demographics. *Sex Disabil*. 2022:40. https://doi.org/10.1007/s11195-022-09763-x.
86. *Kritikos TK, Stiles-Shields C, Shapiro JB, Holmbeck GN. Benefit-finding among young adults with spina bifida. *J Health Psychol*. 2022;27(5):1176–1186. https://doi.org/10.1177/1359105321990804.
87. *Lemelle JL, Guillemin F, Aubert D, et al. Quality of life and continence in patients with spina bifida. *Qual Life Res*. 2006;15(9):1481–1492.
88. *Singh R, Chopra Geeta. Evolution from negative identity to affirmation of 'disability identity': Life story of a woman with Spina Bifida in India. *Disabil CBR Incl Dev*. 2021;Vol 32:81–100. https://doi.org/10.47985/dcidj.409.
89. *Szymanski KM, Misseri R, Whittam B, et al. QUAlity of Life Assessment in Spina bifida for Adults (QUALAS-A): development and international validation of a novel health-related quality of life instrument (Oct) *Qual Life Res*. 2015;24(10):2355–2364. https://doi.org/10.1007/s11136-015-0988-5.
90. *van Buuren AL, O'rinn SE, Lipworth H, Church P, Berndl A. Reproductive health and pregnancy experiences of women with spina bifida: a qualitative study. *J Pedia Rehabil Med*. 2021;14(4):643–654. https://doi.org/10.3233/PRM-200776.
91. *Vu Minh Arnell M, Seljee Svedberg K, Lindehall B, Moller A, Abrahamsson K. Health-related quality of life compared to life situation and incontinence in adults with myelomeningocele: is SF-36 a reliable tool? *J Pedia Urol*. 2013;9(5):559–566.
92. World Health Organization Quality of Life Group. Development of the World Health Organization WHOQOLBREF quality of life assessment. *Psychol Med*. 1998;28:551–558.
93. *Bendt M, Gabrielsson H, Riedel D, et al. Adults with spina bifida: a cross-sectional study of health issues and living conditions. *Brain Behav*. 2020;10(8), e01736. https://doi.org/10.1002/brb3.1736.
94. *Choi EK, Park J, Kim K, et al. Factors affecting the transition to adulthood of Korean young adults with spina bifida: a qualitative study. *BMC Nurs*. 2023;22:46. https://doi.org/10.1186/s12912-023-01194-z.
95. *Heller MK, Gambino S, Church P, Lindsay S, Kaufman M, McPherson AC. Sexuality and relationships in young people with Spina Bifida and their partners (Aug) *J Adolesc Health*. 2016;59(2):182–188. https://doi.org/10.1016/j.jadohealth.2016.03.037.
96. *Kinavey C. Adolescents born with spina bifida: experiential worlds and biopsychosocial developmental challenges (Oct-Dec) *Issues Compr Pedia Nurs*. 2007;30(4):147–164. https://doi.org/10.1080/01460860701728352.
97. *Laurence KM, Beresford A. Continence, friends, marriage, and children in 51 adults with spina bifida. *Dev Med Child Neurol Suppl*. 1975;35:123–128. https://doi.org/10.1111/j.1469-8749.1975.tb03591.x.
98. *Lidal IB, Lundberg Larsen K. Anxiety, depression, and fatigue in middle-aged and older persons with spina bifida: a cross-sectional study. *Disabil Rehabil*. 2022;44(25):7936–7946. https://doi.org/10.1080/09638288.2021.2003453.
99. *Liu T, Ouyang L, Walker WO, et al. Education and employment as young adults living with spina bifida transition to adulthood in the USA: a study of the National Spina Bifida Patient Registry, 10.1111/dmcn.15456 *Dev Med Child Neurol*. 2022. https://doi.org/10.1111/dmcn.15456.
100. *Oakeshott P, Poulton A, Hunt GM, Reid F. Walking and living independently with spina bifida: a 50-year prospective cohort study. *Dev Med Child Neurol*. 2019;61(10):1202–1207. https://doi.org/10.1111/dmcn.14168.
101. *Ridosh M, Braun P, Roux G, Bellin M, Sawin K. Transition in young adults with spina bifida: a qualitative study. *Child Care Health Dev*. 2011;37(6):866–874. https://doi.org/10.1111/j.1365-2214.2011.01329.x.
102. *Showen A, Copp HL, Allen IE, Baradaran N, Liaw A, Hampson LA. Characteristics associated with depression, anxiety, and social isolation in adults with Spina Bifida. *Urology*. 2021;149:255–262. https://doi.org/10.1016/j.urology.2020.11.016.
103. *Veenboer PW, Procee AI, Verheijden JM, Bosch JL, van Asbeck FW, de Kort LM. Medical and psychosocial problems in middle-aged spina bifida patients: survey among members of the Dutch patients' association. *Disabil Rehabil*. 2014;36(7):539–545.
104. *Verhoef M, Barf HA, Vroege JA, et al. Sex education, relationships, and sexuality in young adults with spina bifida. *Arch Phys Med Rehabil*. 2005;86(5):979–987.
105. *Bomalaski MD, Teague JL, Brooks B. The long-term impact of urological management on the quality of life of children with spina bifida. *J Urol*. 1995;154(2 Pt 2):778–781.
106. *Bowman RM, McLone DG, Grant JA, Tomita T, Ito JA. Spina bifida outcome: a 25-year prospective. *Pedia Neurosurg*. 2001;34(3):114–120.
107. *Evans K, Hickman V, Carter CO. Handicap and social status of adults with spina bifida cystica. *Br J Prev Soc Med*. 1974;28(2):85–92. https://doi.org/10.1136/jech.28.2.85.
108. *Hensel DJ, Misseri R, Wiener JS, et al. Solo and partnered sexual behavior among an international sample of adults with Spina Bifida. *J Sex Med*. 2022;19(12):1766–1777. https://doi.org/10.1016/j.jsxm.2022.08.201.
109. *Holmbeck GN, Kritikos TK. Psychosocial adjustment in emerging adults with and without Spina Bifida: A 14-year follow-up study. *J Dev Behav Pedia*. 2022;43(1):e20–e28. https://doi.org/10.1097/DBP.0000000000000956.
110. *Hunt GM, Oakeshott P. Lifestyle in adults aged 35 years who were born with open spina bifida: prospective cohort study. *Cereb Fluid Res*. 2004;1(1):4. https://doi.org/10.1186/1743-8454-1-4.
111. *Roach JW, Short BF, Saltzman HM. Adult consequences of spina bifida: a cohort study. *Clin Orthop*. 2011;469(5):1246–1252.
112. *Szymanski KM, Cain MP, Whittam B, Kaefer M, Rink RC, Misseri R. All incontinence is not created equal: Impact of urinary and fecal incontinence on quality of life in adults with Spina Bifida. *J Urol*. 2017;197(3 Pt 2):885–891.
113. *Stubberud J, Langenbahn D, Levine B, Stanghelle J, Schanke AK. Emotional health and coping in spina bifida after goal management training: a randomized controlled trial. *Rehabil Psychol*. 2015;60(1):1–16.
114. *Showen AE, Copp HL, Allen IE, Hampson LA. Resilience and associated characteristics in adults with spina bifida. *Dev Med Child Neurol*. 2021;63(10):1229–1235. https://doi.org/10.1111/dmcn.14919.
115. *Johnsen V, Skattebu E, Aamot-Andersen A, Thyberg M. Problematic aspects of faecal incontinence according to the experience of adults with spina bifida. *J Rehabil Med*. 2009;41(7):506–511. https://doi.org/10.2340/16501977-0373.
116. *Bellin MH, Zabel TA, Dicianno BE, et al. Correlates of depressive and anxiety symptoms in young adults with spina bifida. *J Pedia Psychol*. 2010;35(7):778–789.
117. *Schriner KF, Roessler RT, Johnson P. Identifying the employment concerns of people with spina bifida. *J Appl Rehabil Couns*. 1993;24(2):32–37. https://connect.springerpub.com/content/sgrjarc/24/2/32.
118. *Gabrielsson H, Hultling C, Cronqvist A, Asaba E. Views on everyday life among adults with spina bifida: an exploration through photovoice. *Int J Qual Stud Health Well-being*. 2020;15(1):1830702. https://doi.org/10.1080/17482631.2020.1830702.
119. *Houston JC. Premeditated assaults on young boys by a man with spina bifida and hydrocephalus–a cognitive-behavioural approach to treatment. *Med Sci Law*. 1992;32(2):133–138.
120. Wechsler D. *WAIS-R Manual*. New York: The Psychological Corporation; 1981.
121. Wechsler D. *WAIS-III/WMSIII technical manual: Updated*. San Antonio, TX: Psychological Corporation,; 2002.
122. *Hurley AD, Bell S. Educational and vocational outcome of adults with spina bifida in relationship to neuropsychological testing. *Eur J Pedia Surg*. 1999;4(Suppl 1):17–18. https://doi.org/10.1055/s-2008-1066146.
123. *Loomis JW, Javornisky JG, Monahan JJ, Burke G, Lindsay A. Relations between family environment and adjustment outcomes in young adults with spina bifida. *Dev Med Child Neurol*. 1997;39(9):620–627. https://doi.org/10.1111/j.1469-8749.1997.tb07498.x.
124. Wechsler D. *Manual for the Wechsler Abbreviated Scale of Intelligence*. San Antonio, TX: Psychological Corporation; 1999.
125. *Stubberud J, Riemer G. Problematic psychosocial adaptation and executive dysfunction in women and men with myelomeningocele. *Disabil Rehabil*. 2012;34(9):740–746.
126. Wechsler D. *Wechsler memory scale*. San Antonio, TX: The Psychological Corporation ; 1974 .
127. Raven JC . *Standard Progressive Matrices* . Oxford: Oxford Psychologists Press ; 1996 .
128. *Barf HA, Post MW, Verhoef M, Gooskens RH, Prevo AJ. Is cognitive functioning associated with subjective quality of life in young adults with spina bifida and hydrocephalus? *J Rehabil Med*. 2010;42(1):56–59.
129. *van Mechelen MC, Verhoef M, van Asbeck FW, Post MW. Work participation among young adults with spina bifida in the Netherlands. *Dev Med Child Neurol*. 2008;50(10):772–777.
130. *Dicianno BE, Bellin MH, Zabel AT. Spina bifida and mobility in the transition years. *Am J Phys Med Rehabil*. 2009;88(12):1002–1006.
131. Sharrack B, Hughes RA. The Guy's neurological disability scale (GNDS): a new disability measure for multiple sclerosis. *MultScler*. 1999;5:223–233.
132. Boyle MH, Furlong W, Feeny D, Torrance GW, Hatcher J. Reliability of the Health Utilities Index–Mark III used in the 1991 cycle 6 Canadian General Social Survey Health Questionnaire. *Qual Life Res*. 1995;4:249–257.
133. Cockrell JR, Folstein MF. Mini-mental state examination (MMSE). *Psychopharmacol Bull*. 1988;24:689–692.
134. Nelson HE. *National Adult Reading. Test. Test manual*. Windsor, UK: NFERNELSON; 1982.
135. Dunn LM, Dunn LM. *Peabody Picture Vocabulary Test—Revised. Circle Pines*. MN: American Guidance Service; 1981.
136. Mann Henry B, Whitney Donald R. On a test of whether one of two random variables is stochastically larger than the other. *Ann Math Stat*. 1947;18(1):50–60. https://doi.org/10.1214/aoms/1177730491. MR 0022058. Zbl 0041.26103.
137. Randolph C, Tierney MC, Mohr E, Chase TN. The Repeatable Battery for the Assessment of Neuropsychological Status (RBANS): preliminary clinical validity. *J Clin Exp Neuropsychol*. 1998;20(3):310–319. https://doi.org/10.1076/jcen.20.3.310.823.
138. Benton AL, Hamshire K de S. *Multilingual Aphasia Examination*. Iowa City, IA: University of Iowa; 1976.
139. Maynard FM, Bracken MB, Creasey G, et al. International standards for neurological and functional classification of spinal cord injury. *Spinal Cord*. 1997;35:266–274.
140. Janicki MP, Davidson PW, Henderson CM, et al. Health characteristics and health services utilization in older adults with intellectual disability living in community residences. *J Intellect Disabil Res: JIDR*. 2002;46(Pt 4):287–298. https://doi.org/10.1046/j.1365-2788.2002.00385.x.
141. Wilkinson GS, Robertson GJ. *Wide Range Achievement Test professional manual*. 5th ed. Bloomington, MN: NCS Pearson, Inc; 2017.

142. Kaufman AS, Kaufman NL. *Functional academic skills test*. 22. Circle Pines: American Guidance Service; 1994.
143. *Riedel D, Hagman G, Green D, Fristedt S. Cognitive function and performance of everyday activities in adults with spina bifida. *J Rehabil Med*. 2021;53(9): jrm00225. https://doi.org/10.2340/16501977-2868.
144. Nelson HE. A modified card sorting test sensitive to frontal lobe defects. *Cortex*. 1976;12:313–324.
145. Delis DC, Kaplan E, Kramer JH. *Delis-Kaplan Executive Functioning System (D-KEFS)*. San Antonio, TX: The Psychological Corporation; 2001.
146. Reitan RM. Validity of the Trail Making Test as an indicator of organic brain damage. *Percept Mot Skill*. 1958;8:271–276.
147. Spreen O, Strauss E. *A compendium of neuropsychological tests*. New York: Oxford University Press; 1998.
148. Gioia GA, Isquith PK, Guy SC, Kenworthy L. *Behavior Rating Inventory of Executive Function: Professional Manual*. Lutz, FL: Psychological Assessment Resources, Inc; 2000.
149. Wilson, B.A., Alderman, N., Burgess, P.W., Emslie, H. & Evans, J.J. (1996). Behavioural assessment of the dysexecutive syndrome. Bury St Edmunds: Thames Valley Test Company.
150. Wilson B, Cockburn J, Baddeley A, Hiorns R. The development and validation of a test battery for detecting and monitoring everyday memory problems. *J Clin Exp Neuropsychol*. 1989;11(6):855–870. https://doi.org/10.1080/01688638908400940.
151. Laurence Km. The natural history of Spina Bifida Cystica: Detailed analysis of 407 cases. *Arch Dis Child*. 1964;39(203):41–57. https://doi.org/10.1136/adc.39.203.41.
152. World Health Organization. (2023). Social Determinants of Health. https://www.who.int/health-topics/social-determinants-of-health#tab=tab_1.
153. Laurence KM. The survival of untreated spina bifida cystica. *Dev Med Child Neurol*. 1966:10–19. https://doi.org/10.1111/j.1469-8749.1966.tb02172.x.
154. *Hunt GM, Poulton A. Open spina bifida: a complete cohort reviewed 25 years after closure. *Dev Med Child Neurol*. 1995;37(1):19–29. https://doi.org/10.1111/j.1469-8749.1995.tb11929.x.
155. *Oakeshott P, Hunt GM. Long-term outcome in open spina bifida. *Br J Gen Pr*. 2003;53(493):632–636. PMID: 14601340; PMCID: PMC1314678.
156. Healthy People 2030, U.S. Department of Health and Human Services, Office of Disease Prevention and Health Promotion. Social Determinants of Health. Retrieved from https://health.gov/healthypeople/priority-areas/social-determinants-health.
157. Braveman P, Arkin E, Orleans T, Proctor D, Plough A. *What is Health Equity? And What Difference Does a Definition Make?* Princeton, NJ: Robert Wood Johnson Foundation; 2017.
158. National Institute of Minority Health and Health Disparities, U.S. Department of Health and Human Services, National Institute of Health. (2023). Diversity & Inclusion in Clinical Trials. Retrieved from https://www.nimhd.nih.gov/resources/understanding-health-disparities/diversity-and-inclusion-in-clinical-trials.html.

References Cited in Appendices

159. *Hunt GM . Open spina bifida: outcome for a complete cohort treated unselectively and followed into adulthood . *Dev Med Child Neurol* . 1990;32 (2) : 108 –118 , 1990 .
160. *Iddon JL , Morgan DJ , Loveday C , Sahakian BJ , Pickard JD . Neuropsychological profile of young adults with spina bifida with or without hydrocephalus . *J Neurol Neurosurg Psychiatry* . 2004;75 (8) : 1112 –1118 , 2004 .
161. *Valtonen K , Karlsson AK , Alaranta H , Viikari-Juntura E . work participation among persons with traumatic spinal cord injury and meningomyelocele . *J Rehabil Med* . 2006;38 (3) : 192 –200 , 2006 .
162. *Valtonen K , Karlsson AK , Siosteen A , Dahlof LG , Viikari-Juntura E . Satisfaction with sexual life among persons with traumatic spinal cord injury and meningomyelocele . *Disabil Rehabil* . 2006;28 (16) : 965 –976 , 2006 .
163. *Young N , McCormick A , Mills W , Barden W , Boydell K , Law M , Wedge J , Fehlings D , Mukherjee S , Rumney P , Williams JI . The transition study: a look at youth and adults with cerebral palsy, spina bifida and acquired brain injury . *Phys Occup Ther Pediatr* . 2006;26 (4) : 25 –45 , 2006 .
164. *Jenkinson MD , Campbell S , Hayhurst C , Clark S , Kandasamy J , Lee MK , Flynn A , Murphy P , Mallucci CL . Cognitive and functional outcome in spina bifida-Chiari II malformation . *Child's nervous system: ChNS: official journal of the International Society for Pediatric Neurosurgery* . 2011;27 (6) : 967 –974 . https://doi.org/10.1007/s00381-010-1368-7 .
165. *Vu Minh Arnell M , Seljee Svedberg K , Lindehall B , Jodal U , Abrahamsson K . Adults with myelomeningocele: an interview study about life situation and bladder and bowel management . *J Pediatr Urol* . 2013;9 (3) : 267 –271 , 2013 .
166. *Young NL , Sheridan K , Burke TA , Mukherjee S , McCormick A . Health outcomes among youths and adults with spina bifida . *J Pediatr* . 2013;162 (5) : 993 –998 , 2013 .
167. *Akre C, Light A, Sherman L, Polvinen J, Rich M. What young people with spina bifida want to know about sex and are not being told. *Child Care Health Dev* .. 2015 https://doi.org/10.1111/cch.12282.
168. *Liu JS , Dong C , Casey JT , Greiman A , Mukherjee S , Kielb SJ . Quality of life related to urinary continence in adult spina bifida patients . *Cent* . 2015;68 (1) : 61 –67 , 2015 .
169. Ware Jr JE , Sherbourne CD . The MOS 36-item short-form health survey (SF-36). I. Conceptual framework and item selection . *Med Care* . 1992;30 (6) : 473 –483 .
170. Aaronson NK , Muller M , Cohen PDA , Essink-Bot ML , Fekkes M , Sanderman R , Sprangers MAG , te Velde A , Verrips E . Translation, validation, and norming of the Dutch language version of the SF-36 Health Survey in community and chronic disease populations . doi.org/ *J Clin Epidemiol* . 1998; 51 : 1055 –1068 . https://doi.org/10.1016/S0895-4356(98)00097-3 .
171. Fugl-Meyer AR , Bränholm I-B , Fugl-Meyer KS . Happiness and domain-specific life satisfaction in adult northern Swedes . *Clinical Rehabilitation* . 1991;5 (1) : 25 –33 . https://doi.org/10.1177/026921559100500105 .
172. Sawin KJ , Brei TJ , Buran CF , Fastenau PS . Factors associated with quality of life in adolescents with spina bifida . *J Holist Nurs* . 2002;20 (3) : 279 –304 . https://doi.org/10.1177/089801010202000307 .
173. Fugl-Meyer AR , Melin R , Fugl-Meyer KS . Life satisfaction in 18- to 64-year-old Swedes: in relation to gender, age, partner and immigrant status . *J Rehabil Med* . 2002;34 (5) : 239 –246 . https://doi.org/10.1080/165019702760279242 .
174. de Boer AG , van Lanschot JJ , Stalmeier PF , van Sandick JW , Hulscher JB , de Haes JC , Sprangers MA . Is a single-item visual analogue scale as valid, reliable and responsive as multi-item scales in measuring quality of life? . *Qual Life Res* . 2004;13 : 311 –320 . https://doi.org/10.1023/B:QURE.0000018499.64574.1f .
175. Hawthorne G , Richardson J , Osborne R . The Assessment of Quality of Life (AQoL) instrument: a psychometric measure of health-related quality of life . *Qual Life Res* . 1999;8 (3) : 209 –224 . https://doi.org/10.1023/a:1008815005736 .
176. Tomich PL , Helgeson VS . Is finding something good in the bad always good? Benefit finding among women with breast cancer . *Health Psychol* . 2004;23 (1) : 16 –23 . https://doi.org/10.1037/0278-6133.23.1.16 .
177. Carver CS . You want to measure coping but your protocol's too long: consider the brief COPE . *Int J Behav Med* . 1997;4 : 92 –100 .
178. Feeny D , Torrance GW , Furlong W . Health Utilities Index . In: Spilker B , ed . *Quality of life and pharmacoeconomics in clinical trials* . 2nd ed. Philadelphia: Lippincott-Raven Publishers ; 1996 .
179. Patrick DL , Martin ML , Bushnell DM , Yalcin I , Wagner TH , Buesching DP . Quality of life of women with urinary incontinence: further development of the incontinence quality of life instrument (I–QOL) . *Urology*. 1999;53 : 71 –76 .
180. Cohen SR , Mount BM , Bruera E , Provost M , Rowe J , Tong K . Validity of the McGill Quality of Life Questionnaire in the palliative care setting: a multi-centre Canadian study demonstrating the importance of the existential domain . *Palliat Med* . 1997;11 : 3 –20 .
181. Krogh K , Christensen P , Sabroe S , Laurberg S . Neurogenic bowel dysfunction score . *Spinal Cord* . 2006;44 (10) : 625 –631 . https://doi.org/10.1038/sj.sc.3101887 .
182. Lefante Jr JJ , Harmon GN , Ashby KM , Barnard D , Webber LS . Use of the SF-8 to assess health-related quality of life for a chronically ill, low-income population participating in the Central Louisiana Medication Access Program (CMAP) . *Qual Life Res* . 2005;14 (3) : 665 –673 . https://doi.org/10.1007/s11136-004-0784-0239-52 .
183. Simeoni MC , Sapin C , Antoniotti S , Auquier P . Health-related quality of life reported by French adolescents: a predictive approach of health status? . *J Adolesc Health* . 2001;28 (4) : 288 –294 . https://doi.org/10.1016/s1054-139x(00)00198-1 .
184. Austin JK , Huberty TJ . Development of the Child Attitude Toward Illness Scale . *Journal of Pediatric Psychology* . 1993;18 (4) : 467 –480 . https://doi.org/10.1093/jpepsy/18.4.467 .
185. Steinberg L . Impact of puberty on family relations: Effects of pubertal status and pubertal timing . *Developmental Psychology* . 1987;23 (3) : 451 –460 . https://doi.org/10.1037/0012-1649.23.3.451 .
186. Prinz RJ , Foster S , Kent RN , O'Leary KD . Multivariate assessment of conflict in distressed and nondistressed mother-adolescent dyads . *J Appl Behav Anal* . 1979;12 (4) : 691 –700 . https://doi.org/10.1901/jaba.1979.12-691 .
187. Slavin LA . Validation Studies of the PEPSS, a Measure of Perceived Emotional Support for Use with Adolescents . *Journal of Adolescent Research* . 1991;6 (3) : 316 –335 . https://doi.org/10.1177/074355489163004 .
188. Petersen AC , Schulenberg JE , Abramowitz RH , Offer D , Jarcho HD . A self-image questionnaire for young adolescents (SIQYA): Reliability and validity studies . *J Youth Adolesc* . 1984;13 (2) : 93 –111 . https://doi.org/10.1007/BF02089104 .
189. Bennion LD , Adams GR . A revision of the extended version of the objective measure of ego identity status: an identity instrument for use with late adolescents . *J Adolesc Res* . 1986;1 : 183 –197 .
190. Farrell AD , Danish SJ , Howard CW . Relationship between drug use and other problem behaviors in urban adolescents . *J Consult Clin Psychol* . 1992;60 (5) : 705 –712 . https://doi.org/10.1037//0022-006x.60.5.705 .
191. Connor KM , Davidson JRT . Development of a new resilience scale: the Connor-Davidson Resilience Scale (CD-RISC) . *Depress Anxiety* . 2003;18 : 76 –83 .
192. Joseph S , Williams R , Yule W . Crisis support, attributional style, coping style and post-traumatic symptoms . *Personality and Individual Differences* . 1992; 13 : 1249 –1251 . https://doi.org/10.1016/0191-8869(92)90262-N .
193. Schwarzer R , Jerusalem M . Generalised Self-Efficacy Scale . In: Weinman J , Wright S , Johnston M , eds . *Measures in health psychology: a user's portfolio. Causal and control beliefs* . Windsor, UK: Nfer-NELSON ; 1995 : 35 –37 .
194. *Ridosh MM , Adams W , MagaÃ±a F , Sawin KJ , Holmbeck GN . *Trajectories of self-management and independence in youth with spina bifida:*

Demographic predictors of growth [published online ahead of print, 2022 Oct 7] . *Child Care Health Dev* . 2022 . https://doi.org/10.1111/cch.13065 .

195. Rosenberg M . *Society and the Adolescent Self-Image* . Princeton, NJ: Princeton University Press ; 1965 .
196. Matthews CG , Kløve H . *Instruction manual for the Adult Neuropsychology Test Battery* . Madison, WI: University of Wisconsin Medical School ; 1964 .
197. Owen AM , Downes JJ , Sahakian BJ , Polkey CE , Robbins TW . Planning and spatial working memory following frontal lobe lesions in man . *Neuropsychologia* . 1990;28 (10) : 1021 –1034 . https://doi.org/10.1016/0028-3932(90)90137-d .
198. Owen AM , Sahakian BJ , Semple J , Polkey CE , Robbins TW . Visuo-spatial short-term recognition memory and learning after temporal lobe excisions, frontal lobe excisions or amygdalo-hippocampectomy in man . *Neuropsychologia* . 1995;33 (1) : 1 –24 . https://doi.org/10.1016/0028-3932(94)00098-a .
199. Owen AM , James M , Leigh PN , Summers BA , Marsden CD , Quinn NP , Lange KW , Robbins TW . Fronto-striatal cognitive deficits at different stages of Parkinson's disease . *Brain* . 1992;115 (Pt 6) : 1727 –1751 . https://doi.org/10.1093/brain/115.6.1727 .
200. Milner B . Interhemispheric differences in the localization of psychological processes in man . *Br Med Bull* . 1971;27 (3) : 272 –277 . https://doi.org/10.1093/oxfordjournals.bmb.a070866 .
201. Downes JJ , Roberts AC , Sahakian BJ , Evenden JL , Morris RG , Robbins TW . Impaired extra-dimensional shift performance in medicated and unmedicated Parkinson's disease: evidence for a specific attentional dysfunction . *Neuropsychologia* . 1989;27 (11-12) : 1329 –1343 . https://doi.org/10.1016/0028-3932(89)90128-0 .
202. Benedict R , Schretlen D , Groninger L , Brandt J . Hopkins Verbal Learning Test – Revised: Normative Data and Analysis of Inter-Form and Test-Retest Reliability . *The Clinical Neuropsychologist* . 1998;12 (1) : 43 –55 . https://doi.org/10.1076/clin.12.1.43.1726 .
203. Jennekens-Schinkel A , Lanser JB , van der Velde EA , Sanders EA . Performances of multiple sclerosis patients in tasks requiring language and visuoconstruction . *Assessment of outpatients in quiescent disease stages. J Neurol Sci* . 1990;95 (1) : 89 –103 . https://doi.org/10.1016/0022-510x(90)90119-8 .
204. Schmidt M . *Rey Auditory and Verbal Learning Test. A handbook. Los Angeles* . Western: Psychological Services ; 1996 .
205. Berg M , Butler A , Dahl M , Eklund P , Norrlin S , Rönnblom Hållén E , et al . *Independence and health in adolescents and young adults with spina bifida . Needs and measures* . Uppsala: Folke Bernadotte regionhabilitering ; 2008 (in Swedish) .
206. Rossetti HC , Lacritz LH , Cullum CM , Weiner MF . Normative data for the Montreal Cognitive Assessment (MoCA) in a population-based sample . *Neurology* . 2011;77 (13) : 1272 –1275 . https://doi.org/10.1212/WNL.0b013e318230208a .
207. Tallberg IM , Ivachova E , Jones Tingheg K , Ostberg P . Swedish norms for word fluency tests: FAS, animals, and verbs . *Scandinavian Journal of Psychology* . 2008;49 : 479 –485 . https://doi.org/10.1111/j.1467-9450.2008.00653.x .
208. Appelbaum, P.S., & Grisso, T. (2001). MacArthur competence assessment tool for clinical research. Sarasota, FL: Professional Resource Exchange.
209. Ridosh MM , Stiles-Shields C , Stern A , Winning AM , Anderson L , Sawin KJ , Holmbeck GN . The Adolescent/Young Adult Self-Management and Independence Scale (AMIS-II): Expanding evidence for validity and reliability . *Journal of Pediatric Rehabilitation Medicine* . 2021;14 (4) : 583 –596 . https://doi.org/10.3233/PRM-20067 .