

Perceived health in young adults with spina bifida

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The aims of this study were to compare the perceived health of young adults with spina bifida with a population without disability, and to determine the effect of the disease characteristics and resulting impairments on perceived health. This cross-sectional study is part of the Adolescents with Spina Bifida in the Netherlands study. Data were collected by physical examination and a questionnaire. In total, 179 patients (age range 16–25y) participated in the study and perceived health data were completed for 164 participants (92 females, 72 males; mean age 20y 7mo [SD 2y 9mo]). Twenty-six participants had spina bifida occulta and 138 had spina bifida aperta, of whom 115 also had hydrocephalus. Perceived health was measured with the Medical Outcome Study 36-item Short-form Health Survey (SF-36), a generic health status measure. SF-36 scores of young adults with spina bifida were below those of an age-matched population group for six of the eight domains. This difference was largest for the physical functioning domain. Although these differences were statistically significant they were small. Findings for the emotional health domains (vitality, mental health, role problems due to emotional problems) did not differ at all from the population group.

The life expectancy of patients with spina bifida has increased and many now live well into adulthood, which leads to new challenges. Most young adults with spina bifida face various impairments and activity limitations due to their condition,¹ which may affect their quality of life. Quality of life is a broad concept, covering aspects of health, participation, satisfaction with functioning, and general well-being.² Perceived health, also called self-reported health status or health-related quality of life (HRQL), covers symptoms of a disease or condition and the physical, cognitive, and social problems resulting from the symptoms. The perceived health of young adults with spina bifida is an important concept because it can help improve clinical care by describing the consequences of a disease or condition, and by improving the understanding of the impact of treatment options.³ However, perceived health has only rarely been studied in this group; most studies have focused on physical independence and living conditions,^{4,5} social relationships or vocation,^{4–9} feelings of competence,^{5,6,10} behavioural problems,^{8,11,12} or well-being.⁴

Parkin et al.¹³ developed a specific HRQL scale for children and adolescents with spina bifida. This scale consists of 47 items in 10 domains, including physical independence, recreation, emotions, medical care, and finances. Several researchers have used the questionnaire, reporting good quality of life among young adults with spina bifida.^{3,14} Unfortunately, the HRQL has only one total score for all 47 items, so that it cannot differentiate between, for example, physical independence and financial status.

Health questionnaires that result in a useful profile of different aspects of health are available, but have rarely been used for this patient group. Padua et al.^{15,16} used the best-known health questionnaire, the Medical Outcome Study 36-item Short-form Health Survey (SF-36),¹⁷ and found that greater disability and severe muscle deficit of the lower limbs were associated with poorer scores for the physical aspects of HRQL. By contrast, greater disability was associated with better scores on the cognitive aspects of HRQL. The authors hypothesized that sphincter problems play a key role in the HRQL pattern, but were unable to confirm this, perhaps because of their very small sample size ($n=12$).^{15,16}

Our literature search found that, to date, little is known about the perceived health of young adults with spina bifida. To identify the health care needs of this group, we administered the SF-36 as part of the Adolescents with Spina Bifida in the Netherlands (ASPINE) study, a cross-sectional study on physical and cognitive disabilities, health care, participation in society, and life satisfaction of the study participants.

The aims of this part of the ASPINE study were: (1) to describe the perceived health of young adults with spina bifida and compare this with that of a population without disability; and (2) to determine the association between disease characteristics and the resulting impairments and perceived health of these young adults.

Method

PARTICIPANTS

Young adults with different types of spina bifida (aperta and occulta; International Classification of Diseases [ICD] codes 741 and 756.17),¹⁸ aged between 16 and 25 years, and living in the Netherlands were included in the study. Non-Dutch speaking participants or participants with comorbidity that independently caused more physical and/or cognitive disabilities than

See end of paper for list of abbreviations.

the spina bifida itself were excluded from the study. Participants with a diagnosis of spina bifida occulta but without any neurological loss were also excluded.

Participants were recruited from 11 of the 12 multidisciplinary spina bifida teams in the Netherlands. In addition, rehabilitation centres, special housing facilities, and special schools were approached to find potential participants. The Dutch Association of Patients with Spina Bifida also invited members to participate, and advertisements were placed in two national magazines and on the internet.

INSTRUMENTS

Data were collected by means of an interview and a physical examination (performed by a physician).

Hydrocephalus was defined as having either a shunt at the time of the physical examination or having had one previously.

In accordance with the International Standards for Neurological and Functional Classification of Spinal Cord Injury,¹⁹ level of lesion was defined as the lowest completely unimpaired dermatome level on both sides, measured with sensitivity to pin prick and light touch. Participants were divided into three subgroups based on level of lesion: (1) high level (HLL; L2 and above); (2) middle level (MLL; L3–L5); and (3) low level (LLL; S1 and below).¹

The impairments assessed in this study were visual acuity, increase of pain in head, neck, or back during the previous 24 months, spasticity, epilepsy, low IQ, scoliosis, lack of sitting balance, hip contractures, knee contractures, foot deformities, lack of mobility, urinary and/or faecal incontinence, obstipation, diaper use, pressure sores, and obesity. Obesity was measured as the sum of bicipital, tricipital, subscapular, and suprailiacal skinfold thickness. The cut-off point for obesity was a body-fat percentage of 25% for males and 35% for females.²⁰ Detailed information about the assessment of the other impairments and the cut-off points used has been provided elsewhere.¹

The SF-36 is a generic measuring instrument for health status. It measures the perceived health or HRQL and can be used to compare participants with various conditions with a population without disability. The SF-36 assesses eight health concepts: physical functioning, role limitations caused by physical health problems, role limitations caused by emotional problems, social functioning, emotional well-being, energy and fatigue, bodily pain, and general health perceptions. Scale

scores range from (0) worst possible health status to (100) best possible health status.

The scores for the age-matched Dutch population without disability were obtained from the validation study of the Dutch language version of the SF-36,²¹ from which scores for the 16- to 25-year age group ($n=201$) were computed to facilitate direct comparison with our group of young adults with spina bifida.

STATISTICS

Data were analyzed using SPSS (version 12.0.1). The reliability of the eight SF-36 domains was tested by determining Cronbach's alpha, which ranged from 0.76 for vitality to 0.93 for physical functioning. This means that all scales can be considered reliable. Mean scores and confidence intervals (CI) of the eight health concepts were calculated for the subgroups (occulta, aperta with hydrocephalus, aperta without hydrocephalus), the total study group, and the reference group. We used the independent samples *t*-test to test univariately the effect of demographic variables (age, sex), disease characteristics (hydrocephalus, level of lesion, type of spina bifida) and secondary impairments (visual acuity, pain in head, neck or back, plasticity, epilepsy, scoliosis, lack of sitting balance, hip contractures, knee contractures, foot deformities, lack of mobility, urinary and/or faecal incontinence, obstipation, diaper use, pressure sores, and obesity) on the outcome of the eight domains of the SF-36.

ETHICAL APPROVAL

The medical ethics committee of the University Medical Centre Utrecht approved the ASPINE study. Informed consent was obtained from all participants.

Results

PARTICIPANTS

In total, 350 patients were invited by mail to participate in this study, of whom 181 agreed to participate. Participants and non-participants were similar in age (mean 20y 5mo [SD 3y] versus 20y 4mo [SD 3y 1mo]), sex (41% female vs 49% male), type of spina bifida (79% occulta vs 86% aperta), level of lesion (19% vs 23% L2 and above; 66% vs 64% L3 to L5; and 15% vs 13% S1 and below), and being shunted for hydrocephalus (67% vs 64% being shunted). Two patients who were initially invited to participate in the study were later

Table I: Mean (range) SF-36 scores and confidence intervals in relation to type of spina bifida (SB; occulta, aperta without hydrocephalus [HC-], and aperta with hydrocephalus [HC+]) compared with a population without disability

Health concepts	Occulta (n=26)	Aperta HC- (n=23)	Aperta HC+ (n=115)	Total SB (n=164)	Norm. population ^b (n=201)
Physical functioning	67.5 ^a (56.3–78.6)	80.4 ^a (71.2–89.7)	34.4 ^a (29.8–39.0)	46.1 ^a (41.4–50.9)	94.2 (92.9–95.5)
Role physical ^c	58.7 ^a (41.3–76.0)	82.6 ^a (69.0–96.2)	68.9 ^a (62.2–75.7)	69.2 ^a (63.5–75.0)	88.3 (84.8–91.8)
Bodily pain	67.7 ^a (58.7–76.7)	81.7 (73.5–89.9)	75.6 ^a (70.9–80.3)	75.2 ^a (71.5–79.0)	82.5 (79.9–85.1)
General health	58.0 ^a (47.1–68.9)	71.8 ^a (61.7–82.0)	64.9 ^a (60.7–69.1)	64.8 ^a (61.1–68.4)	77.9 (75.5–80.3)
Vitality	62.7 ^a (55.5–69.9)	71.5 (63.3–79.8)	64.3 ^a (61.1–67.5)	65.1 ^a (62.3–67.8)	70.6 (68.4–72.8)
Social functioning	76.4 ^a (65.5–87.3)	89.7 (82.6–96.7)	81.8 ^a (77.7–86.0)	82.1 ^a (78.6–85.6)	87.8 (85.3–90.3)
Role emotional ^d	71.8 ^a (54.5–89.1)	84.1 (70.4–97.7)	85.2 (79.7–90.8)	82.9 (77.9–88.0)	83.7 (79.5–87.9)
Mental health	73.5 ^a (66.7–80.4)	81.9 (75.9–88.0)	76.8 (74.0–79.7)	77.0 (74.6–79.4)	77.8 (75.8–79.8)

^aSignificant differences between participants with spina bifida and normative (norm.) population. ^bMedical Outcome Study 36-item Short-form Health Survey (SF-36) normative data for Dutch population (age range 16–25y; Aaronson et al.). ²¹ ^cRole limitations due to physical health problems. ^dRole limitations due to emotional health problems.

excluded because of comorbidity independently inducing serious physical and/or cognitive disorders: one had a serious heart disease and one had a chromosomal disorder. Eleven participants diagnosed with spina bifida occulta but without any neurological loss were also excluded.

Data for this study of perceived health were completed for 164 participants. Forty per cent of the participants completed the questionnaire alone and 55% had some assistance from a caregiver. A caregiver mainly completed the questionnaire for 5% of participants. Mean age of the population (92 females, 72 males) was 20y 8mo (SD of 2y 11mo, range 16–25y). Fifty-two per cent of the participants were between 16 and 20 years of age, and 48% were between 21 and 25 years of age. In total, 26 participants had spina bifida occulta and 138 had spina bifida aperta.

PERCEIVED HEALTH OF PARTICIPANTS WITH SPINA BIFIDA RELATED TO THE REFERENCE POPULATION

Young adults with spina bifida showed poorer perceived health than the reference group for six of the eight SF-36 domains (Table I). The scores on the mental health domains (role limitations due to emotional problems, mental health) did not deviate significantly from those of the reference group. Participants with spina bifida occulta had significantly lower scores than the reference group on all SF-36 domains and showed poorer perceived health than the patients with spina bifida aperta with or without hydrocephalus, for seven of the eight domains (except physical functioning).

ASSOCIATIONS BETWEEN IMPAIRMENTS AND PERCEIVED HEALTH

Tables II and III show the bivariate tests for possible predictors

of perceived health. Significant associations were found between physical functioning and type of spina bifida, hydrocephalus, level of lesion, and most of the impairments. Higher level of lesion, pain in the head, neck or back, and obstipation were related to a lower scores on five or more domains of the SF-36.

Discussion

This study showed that the perceived health of young adults with spina bifida was poorer than that of an age-matched population group for six of the eight SF-36 domains assessed. The difference was largest for the physical functioning domain, covering walking, self-care ability, and strenuous activities. Although these differences were statistically significant, they were smaller than expected. The domains of emotional health (vitality, mental health, role problems due to emotional problems) did not show any difference with the reference group.

Our results are comparable with those of Padua et al.,^{15,16} except for a somewhat lower physical functioning score in our group (46.1 vs 57.6) and much higher scores for social functioning and role problems due to emotional problems in our group (82.1 and 82.9 vs 67.7 and 57.5 respectively). These differences might be due to chance findings in their very small group ($n=12$).¹⁵ Our overall finding of good perceived health is in line with the results of Sawin et al.³ and Kirpalani et al.,¹⁴ who used a condition-specific HRQL questionnaire. Our finding that the self-reported mental health of the participants was similar to that of healthy young adults is supported by the studies of Börjeson and Lagergren,⁴ and Rinck et al.⁷ Appleton et al.¹⁰ found lower self-perceptions among children (aged 7–18y) with spina bifida compared with

Table II: Mean (SD) scores of SF-36 domains for subgroups of participants with spina bifida.^a Mean scores are shown when significant differences were found with an independent samples *t*-test

	<i>n</i>	<i>Physical functioning</i>	<i>Role physical^b</i>	<i>Bodily pain</i>	<i>General health</i>	<i>Vitality</i>	<i>Social functioning</i>	<i>Role emotional^c</i>	<i>Mental health</i>
Sex									
Male	67	–	–	–	–	69.6 (17.8)	–	–	–
Female	97	–	–	–	–	61.9 (17.1)	–	–	–
Age, y									
0–20	84	–	–	79.0 (24.5)	68.9 (22.6)	–	–	–	–
21–25	79	–	–	71.5 (23.5)	60.5 (24.2)	–	–	–	–
Type of spina bifida									
Occulta	26	67.5 (27.6)	–	–	–	–	–	–	–
Aperta	136	42.1 (29.8)	–	–	–	–	–	–	–
Hydrocephalus									
Yes	115	34.4 (25.0)	–	–	–	–	–	–	–
No	49	73.6 (25.5)	–	–	–	–	–	–	–
Level of lesion									
L2 and above	70	22.9 (17.9) ^d	60.0 (40.5) ^d	–	60.1 (24.2) ^d	–	77.1 (25.5) ^d	–	74.2 (16.7) ^d
L3–L5	67	54.8 (24.7) ^d	75.4 (32.1)	–	68.6 (22.4)	–	83.6 (21.8)	–	77.9 (15.3)
S1 and below	27	85.0 (18.2) ^d	77.8 (36.9)	–	67.6 (24.0)	–	91.2 (12.9) ^d	–	82.4 (12.1)
IQ									
>70	133	48.7 (30.3)	–	–	–	–	–	–	–
≤70	23	28.9 (25.1)	–	–	–	–	–	–	–
Mobility									
Ambulant	98	65.4 (23.6)	74.7 (35.8)	–	–	–	84.8 (21.6)	–	–
Wheelchair	66	17.5 (12.6)	61.0 (38.3)	–	–	–	78.0 (24.0)	–	–

^aScore range of all Medical Outcome Study 36-item Short-form Health Survey (SF-36) domain scores is from 0 (worst possible scoring) to 100 (best possible scoring). ^bRole limitations due to physical health problems. ^cRole limitations due to emotional health problems. ^dSignificant difference for this specific level of lesion, compared with participants with other levels of lesion, measured with independent samples *t*-test ($p<0.05$).

an age-, sex-, and social class-matched sample in terms of intellectual skills, but not as regards global self-worth. Lindstrom and Eriksson⁵ described less favourable personal psychological conditions in children with spina bifida, but they included a broader age range (2–18y) and relied largely on parental reports.

We found significant differences in perceived health in the spina bifida group between the young adults with spina bifida occulta, those with spina bifida aperta without hydrocephalus, and those with spina bifida aperta with hydrocephalus. Patients with spina bifida occulta reported the poorest perceived health for all domains except physical functioning. This finding confirms earlier observations.^{15,16,22} The most likely

explanation is that patients with a different type of spina bifida have a different frame of reference. Young adults with spina bifida occulta usually attend mainstream schools and may use healthy peers as a reference group, whereas young adults with spina bifida aperta are more likely to attend special schools and may use peers with a disability as a reference group.^{10,23} It has also been hypothesized that patients with hydrocephalus may overrate their health due to cognitive impairments, but we found no support for this hypothesis in our study (data not shown).

We found close relationships between a wide range of impairments and the physical functioning domain, but only weak to moderate relationships between impairments and the

Table III: Mean (SD) scores of SF-36 domains for participants with spina bifida with or without secondary impairments.^a Mean (SD) scores are shown when significant differences were found with independent samples *t*-test

	<i>n</i>	<i>Physical functioning</i>	<i>Role physical^b</i>	<i>Bodily pain</i>	<i>General health</i>	<i>Vitality</i>	<i>Social functioning</i>	<i>Role emotional^c</i>	<i>Mental health</i>
Visual acuity									
No loss	146	–	–	–	–	–	–	–	–
Loss	11	–	–	–	–	–	–	–	–
Pain in head/neck/back									
No	119	–	73.1 (35.2)	80.6 (21.2)	69.8 (20.2)	68.2 (17.3)	84.2 (21.1)	–	–
Yes	45	–	58.9 (41.0)	61.0 (26.3)	51.4 (27.0)	56.7 (16.2)	76.4 (26.3)	–	–
Spasticity									
No	143	48.4 (29.6)	–	–	–	–	–	–	–
Yes	14	18.2 (22.9)	–	–	–	–	–	–	–
Epilepsy									
No	152	48.1 (30.3)	–	–	–	–	–	–	–
Yes	12	21.7 (27.5)	–	–	–	–	–	–	–
Scoliosis									
No	102	56.1 (28.6)	74.3 (34.9)	–	–	–	–	–	–
Yes	49	27.1 (24.5)	57.1 (40.2)	–	–	–	–	–	–
Sitting balance									
No help	136	50.0 (30.0)	–	–	–	–	–	–	–
Help	21	18.3 (13.7)	–	–	–	–	–	–	–
Hip contracture									
No	132	50.9 (30.1)	–	–	–	–	–	–	–
Yes	19	18.7 (9.4)	–	–	–	–	–	–	–
Knee contracture									
No	115	55.7 (28.3)	–	–	–	–	–	–	–
Yes	42	18.3 (14.6)	–	–	–	–	–	–	–
Foot deformity									
No	37	63.4 (32.2)	–	–	–	–	87.8 (12.7)	–	80.6 (11.4)
Yes	119	40.5 (27.6)	–	–	–	–	80.7 (24.6)	–	75.4 (16.7)
Urinary incontinence									
No	58	54.6 (31.5)	–	–	–	–	–	–	–
Yes	106	41.5 (29.6)	–	–	–	–	–	–	–
Faecal incontinence									
No	103	52.9 (31.3)	–	–	–	–	–	–	–
Yes	61	34.8 (26.6)	–	–	–	–	–	–	–
Obstipation									
No	96	52.6 (31.0)	74.5 (34.0)	80.8 (20.8)	70.1 (21.6)	67.7 (17.4)	86.5 (18.3)	–	79.2 (15.5)
Yes	68	37.0 (28.3)	61.8 (40.6)	67.3 (26.6)	57.2 (24.5)	61.4 (17.7)	75.9 (26.9)	–	74.0 (15.4)
Pressure sores									
No	132	–	–	–	–	–	–	–	–
Yes	25	–	–	–	–	–	–	–	–
Obesity									
No	103	56.0 (28.0)	–	–	–	–	–	–	–
Yes	29	30.3 (27.6)	–	–	–	–	–	–	–

^aScore range of all Medical Outcome Study 36-item Short-form Health Survey (SF-36) domain scores is from 0 (worst possible scoring) to 100 (best possible scoring). ^bRole limitations due to physical health problems. ^cRole limitations due to emotional health problems.

other domains of perceived health. A higher level of lesion, pain in the head, neck, or back, and obstipation were related to lower scores for most domains of the SF-36. Our results cannot be easily compared with those reported in the literature. The study by Padua et al.,^{15,16} the only other study that used the SF-36, found no relationships between level of lesion and the physical and cognitive component scores, but they did not include other impairments in their small-scale study.^{15,16} Kirpalani et al.¹⁴ reported that 31% of the variance of their HRQL score was predicted by functional characteristics (bladder, bowel, mobility, number of operations, and of shunt revisions). Sawin et al.³ found no relationship between a complication index (number of urinary tract infections, skin ulcers, and hospitalizations) and the same HRQL score. Shoenmakers²⁴ found that being independent in terms of mobility was the only factor that contributed to a better HRQL in children aged 1 to 18 years; however, her model included only functional abilities. Furthermore, previous studies did not analyze individual domains of health. Our study adds to the literature by including a large group of relatively homogeneous age (16–25y), addressing several disease characteristics and secondary impairments, and using a standard health status measure that allows a multifaceted description of perceived health.

Conclusions

LIMITATIONS

One limitation of our study is that only about half of those patients invited to participate did so, which may reduce the extent to which the findings can be generalized. This low response rate might be due to the nationwide design of the study in which participants were invited by mail. Furthermore, the population of adolescent patients who could be bored of hospital attendances might be of influence. Generalized differences between participants and non-participants in terms of age, sex, hydrocephalus, type of spina bifida (occulta or aperta), or level of lesion were tested, as far as possible, and found to be insignificant. Our study only included a specific subgroup of participants with spina bifida occulta, consisting of patients with neurological loss, bladder problems, or bowel problems. The reason for excluding patients with spina bifida occulta without any loss of function was to unify this specific patient group. Many patients with spina bifida occulta without any loss of function may not even be aware that they have spina bifida occulta.

A second limitation concerns the SF-36. Although it is now the most commonly used health status measure and has been validated in many different diagnostic groups,^{25,26} it is not a measure specifically developed for spina bifida and may lack items that are very relevant to this patient group. Moreover, patients with severe physical disability may make a distinction between the concepts of 'health' and 'disability'. They may not interpret secondary impairments like incontinence, scoliosis, or spasticity as impaired health, and may, therefore, not 'feel ill'.²⁷ This might explain our finding that only pain and obstipation were related to poorer general health.

IMPLICATIONS

This study showed that the perceived health of young adults with spina bifida was poorer than that of their age-matched peers, especially as regards physical functioning. Pain and

obstipation were related to poorer perceived health for several domains of the SF-36. Physicians should, therefore, pay attention to these impairments in the treatment of young adults, to improve their perceived health. Future research might include determinants like coping behaviours, personality, family, and peer group to find a better explanation of perceived general and mental health.

Accepted for publication 8th October 2006.

Acknowledgements

This study was supported by the Johanna Children's Fund (Arnhem) and Stinafo (The Hague). We would like to thank Olga de Wit and Maaik Dekker for their contribution in collecting the data. We would also like to thank the BOSK patients association, the organizations for sheltered homes, and the following centres for their participation in this study: AMC Amsterdam, Franciscusoord Mytyschool, Isala clinics Zwolle, LUMC Leiden, Rehabilitation Centre Het Roessingh, Rehabilitation Centre Revalidatie Friesland, Rehabilitation Centre Tolbrug, St Elisabeth Hospital Tilburg, UMC Nijmegen, UMC Utrecht, University Hospital Rotterdam, University Hospital Maastricht, VUMC Amsterdam, and Werkenrode.

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List of abbreviations

ASPINE	Adolescents with Spina Bifida in the Netherlands
HRQL	Health-related quality of life
SF-36	Medical Outcome Study 36-item Short-form Health Survey

Mac Keith Meetings

Forthcoming Meetings



Fetal Brain Damage and Placenta

7 March 2007

The impact of impaired placental function on neurodisability and interventions to reduce it.

Managing Mystery Illnesses – Specialty Treatment of Hard to Explain Symptoms

30 April 2007

Health Issues for Child Refugees and Asylum Seekers

31 May 2007

All meetings will be held at the Royal Society of Medicine, London, UK

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