

RESEARCH PAPER

Restrictions in social participation of young adults with spina bifida

H. A. BARF^{1,2}, M. W. M. POST^{1,2}, M. VERHOEF^{1,2}, A. JENNEKENS-SCHINKEL^{2,3},
R. H. J. M. GOOSKENS^{2,4} & A. J. H. PREVO^{1,2}

¹Rehabilitation Centre De Hoogstraat, Utrecht, The Netherlands, ²Rudolf Magnus Institute of Neuroscience, University Medical Centre Utrecht, The Netherlands, ³Sector of Neuropsychology, Department of Medical Psychology, University Medical Centre Utrecht, Wilhelmina Children's Hospital, The Netherlands, and ⁴Department of Child Neurology, University Medical Centre Utrecht, Wilhelmina Children's Hospital, The Netherlands

Accepted July 2008

Abstract

Purpose. To determine participation restrictions of young adults with spina bifida (SB) in relation to health condition and activity limitations.

Method. A total of 179 persons aged 16–25 years and born with SB participated in a cross-sectional study. The main outcome on four domains of participation (independent living, employment, education and partner relationships) was assessed using a structured questionnaire.

Results. At the mean age of 21 years only 16% were living independently, more than one-third of the participants went to special secondary education, 53% of those who finished education did not have a regular job and 71% did not have a partner. Health condition variables (type of SB, hydrocephalus and level of lesion) and to a lesser extent activity limitations (wheelchair dependence and incontinence) were significant determinants for having participation restrictions. Perceived hindrances in participation included long-distance transportation (19–36%), accessibility (10–42%), physical impairments (22–40%), emotional barriers (20–32%) and financial limits (3–17%). More severe SB, defined as hydrocephalus, high level of lesion and wheelchair dependence, was related with more experienced hindrances due to long-distance transportation accessibility of buildings.

Conclusions. Many young adults with spina bifida experience participation restrictions. Severity of SB was negatively related to participation. Social integration should be a major focus in the professional guidance of youngsters with physical disabilities.

Keywords: *Spina bifida, hydrocephalus, young adults, participation, activities*

Introduction

Spina bifida (SB) is a health condition that is caused by a congenital neural tube disorder. Some 50 years ago the life expectancy of babies born with SB was poor [1,2]. The use of shunting for hydrocephalus and intermittent catheterisation have increased chances of survival and life expectancy for babies born with SB is 'near' normal [2–6].

A wide variety of physical impairments and delay of cognitive development are, however, common in persons born with SB [3,4,7–13]. The first few years of a child with SB are the most endangered. At

school age a somewhat quieter period starts, the focus mainly lying on education. New problems arise in the period of transition from childhood to adolescence and adulthood. The challenges in this period include (in)dependent living, work, social relationships, partnership and child wish.

One earlier Dutch study of functioning of young adults with SB aperta concluded that chances for living independently, attending regular education and having a regular job was small and was dependent on the level of neurological deficit [14]. International studies also show that unrestricted participation is difficult to reach for persons with

SB. Level of education is lower than in the general population [3,15,16], unemployment rate is high [3,4,10,15–20], only few can live independently without extra help [3,15,18] and persons with SB seem to have more difficulty finding a partner [3,15]. To improve the transition of youngsters with SB into adulthood and to adjust counselling by health care professionals to the needs of these youngsters, it is important to study determinants of social participation and hindrances for participation as perceived by young adults with SB.

The aim of this study is to examine participation restrictions of a large group of young adults born with SB in relation to disease characteristics, activity limitations and perceived hindrances for participation.

Methods

Participants

The present study was part of the Aspine project [7,11]. Participants were persons with SB *aperta* or *occulta* (International Classification of Diseases, 9th revision codes 741 and 756.17 respectively) [21] who were between 16 and 25 years of age and who had sufficient command of the Dutch language. Participants with co-morbidity that could independently induce serious physical and/or mental impairments were excluded. Participants were recruited through 11 of the 12 Dutch Spina Bifida Teams in coordination with the Dutch Spina Bifida Patients Association, organisations for sheltered homes, and rehabilitation centres. The ethics and research committees of the respective institutions approved the study. Written informed consent was obtained from the participants or their parents if applicable. Invitations were sent to 350 persons, of whom 179 participated in this study. Patient characteristics (age, gender, type of SB, level of lesion, presence of hydrocephalus), collected from medical records, were not significantly different ($\alpha = 0.05$) between the participating and the non-participating group.

Instruments

Medical records were examined according to a fixed protocol and participants underwent a physical and neuropsychological examination. The participants were interviewed between August 1999 and August 2001. Seven participants had the questionnaires filled in on their behalf by their parents.

Demographic variables were age and gender. *Health condition characteristics* included type of SB (*occulta* or *aperta*) and the presence of hydrocephalus, which was defined as having been shunted early in life to reduce intracranial pressure. Both were retrieved

from medical files. Further, level of lesion was defined as the lowest completely unimpaired dermatome levels on both sides with sensitivity to pin prick and light touch and was assessed by physical examination.

Activity limitations were assessed during the physical examination. Ambulatory status was categorised using an adaptation of the Hoffer scale for ambulation [22]. Persons without walking problems and community ambulators were considered ‘walkers’ and persons using a wheelchair for shorter or longer distances were considered ‘wheelchair dependent’. Incontinence was defined as having fecal and/or urinary accidents at least once a month and was dichotomised as continent or incontinent. As part of the neuropsychological examination intelligence was assessed using the Raven Standard Progressive Matrices [23], the number of correct answers was converted into an intelligence quotient (IQ) score with mean = 100 and SD = 15 in the population. An IQ below 85 was considered ‘below average’ and an IQ equal to or higher than 85 was considered ‘at least average’.

Participants were interviewed about their *social participation*, in terms of their educational career, employment status, marital status and independent living using structured questions.

Educational outcome was categorised as follows: primary or special education only; lower secondary education, higher secondary education (at least high school). Employment status was categorised in having a regular job for at least 20 h a week versus no employment, working in a sheltered workplace or having a job for less than 20 h a week. Marital status was categorised as currently having a long lasting relationship with a partner versus not currently having a long lasting relationship. Finally, participants were categorised as living independently versus living in a sheltered home or partially living independently with guidance versus living at home with the parents.

Perceived problems in relation to social participation were collected as part of a self-report questionnaire. The questionnaire was developed in an earlier, unpublished, study. Participants were asked to quantify the occurrence of five types of hindrances with respect to school or work, visiting family or friends and spending leisure time: long distance transportation, accessibility of buildings, physical impairments, emotional distress and costs. Participants quantified the occurrence of problems as ‘never’, ‘sometimes’, ‘regularly’ or ‘very often’. For the purpose of this study the last three quantifications were combined and the scores were dichotomised in two categories: ‘never experience problems’ and ‘experiences problems sometimes to very often’. Cronbach’s α of the questionnaire was 0.79 in this study.

Statistics

Descriptive statistics were used and differences in the proportions between subgroups were tested using the Chi-square test or the Fisher's exact test in case of two by two tables. All results were taken to be significant at a p -level below 0.05 two-sided. In case of multiple testing a Bonferroni correction was applied.

Results

The characteristics of the study group are shown in Table I. Most participants were born with SB aperta and 84% of these participants were shunted because of hydrocephalus early in life. Mean age was 20.7 years (SD 2.9); half of the participants were 21 years of age or older. Moderate to strong correlations existed between type of SB, having hydrocephalus and level of lesion (Kendall's τ 0.34–0.72; $p < 0.001$). These health conditions were also significantly related to wheelchair dependence, continence and IQ (Kendall's τ 0.25–0.61; $p < 0.003$). The intercorrelations between wheelchair dependence, continence and IQ were weak (Kendall's τ 0.17–0.27; $p < 0.03$).

Mobility

As shown in Table I, 39% of the participants were wheelchair user for household ambulation. An even larger proportion made use of an electric or self-propelled wheelchair (47%) or other aids like a tricycle or hand-bike (17%) for longer-distance transportation. Of all participants aged 18 years or older, 26% had a driver's license. Participants who were wheelchair dependent had less often a driver's license (12%) than participants who were not wheelchair dependent (36%; $p = 0.002$) and depended more often on special transportation (89% against 32%; $p < 0.001$).

Social participation

In Table II participation outcomes are summarised and related to demographic characteristics, health condition and activity limitations. Half of the participants were still living with their parents. Thirty-one percent were living in an institution, independent living centre or sheltered home and 16% were living completely independent. More than one-thirds of all participants went to special secondary education or had no education beyond primary school and only 16% had a higher level of education. At the time of the study, half of the participants were still in full-time education. Of the 92 participants who finished their education, 47% had a regular job,

Table I. Characteristics of participants.

	Spina bifida total $N = 179$	Spina bifida without HC $N = 60$	Spina bifida with HC $N = 119$
Gender			
Male (%)	41	43	40
Female (%)	59	57	60
Age			
21 or older (%)	49	48	49
Younger than 21 (%)	51	52	51
Spina bifida			
Aperta (%)	79	38	100*
Occulta (%)	21	62	0
Level of lesion			
L2 or higher (%)	41	12	55*
L3–L5 (%)	38	35	39
S1 or lower (%)	21	53	5
Ambulation			
Wheelchair dependent (%)	39	7	55%*
Independent (%)	61	93	45
Continence			
Incontinent (%)	69	45	81*
Continent (%)	31	55	19
Intelligence			
IQ below 85 (%)	39	19	49*
IQ 85 or higher (%)	61	81	51

*Significant difference between participants with and without hydrocephalus; $p < 0.001$.

15% were working at a sheltered workplace and 38% were unemployed. Most participants (71%) did not have a partner at the time of the study. One-fifth had a steady relation with a boy or girl and 7% was married or living together.

Demographic characteristics in relation to social participation

There were no differences in social participation between men and women and between younger and older participants, except for place of residence. Participants aged 21 or older were more often living independently (28%) than participants aged younger than 21 (5%; $p < 0.001$).

Health condition in relation to social participation

Having SB aperta, having hydrocephalus and having a high level of lesion were all associated with more participation restrictions: higher frequency of special care living arrangements, special secondary education, not having a regular job and not having a partner (Table II). Level of lesion seemed to be

Table II. Social participation of young adults with SB (N = 179).

	Independent living		Education (secondary)		Employment†		Relationships			
	Parents N = 94	Special care N = 56	Independent N = 29	None or special N = 65	Regular low and medium level N = 84	Regular high level N = 29	Unemployed or sheltered workplace N = 49	Regular employment N = 43	No partner N = 127	Partner N = 52
Demographic										
Age										
≥21 (%)	53	31	16	37	47	16	53	47	71	29
<21 (%)										
	37	36	28**	35	48	17	51	49	68	32
	67	27	5	38	47	15	60	40	74	26
Gender										
Male (%)	58	30	12	40	44	16	44	56	74	26
Female (%)	49	32	19	34	50	16	59	41	69	31
Health condition										
Spina bifida										
Aperta (%)	51	39	10**	45	45	10**	62	38*	76	24*
Occulta (%)	57	3	41	5	54	41	13	88	51	49
Hydrocephalus										
With (%)	50	45	5**	51	42	8**	69	31**	80	20**
Without (%)	57	5	38	8	58	33	18	82	53	47
Lesion level										
L2 or above (%)	48	45	7***‡	55	41	4**	69	31	77	23
L3-L5 (%)	50	31	19	30	49	21	47	53	75	25
S1 or lower (%)	66	5	29	13	55	32	29	71	53	47
Activity limitations										
Wheelchair										
Dependent (%)	43	51	6**	58	41	1**	68	32	80	20
Independent (%)	59	18	23	23	51	26	43	57	65	35
Continence										
Incontinent (%)	50	37	12**‡	43	47	11*	64	36*	77	23*
Continent (%)	57	18	25	23	48	29	27	73	57	43
Intelligence										
Low IQ (%)	51	35	14	65	35	0**	73	27*	74	26
Average IQ (%)	43	41	15	17	57	25	35	65	70	30

*Significant difference at $p < 0.01$.
 **Significant difference at $p < 0.001$.
 †Only participants who finished education (N = 92).
 ‡Only significant different for age 21+ at $p < 0.01$.

related to higher participation in employment and relationships, but these differences were not significant after correction for multiple testing. As correlates of participation might be age-specific, all analyses were also performed for both age groups (>21 and <21) separately. Only one difference occurred: the association between level of lesion and residence was not significant in the group of participants younger than 21 years. In general, relationships between health condition and participation variables were somewhat stronger in the group of participants aged 21 years older than in the group younger than 21 years.

Activity limitations in relation to social participation

Wheelchair dependence was related to living (independently and educational outcome, but not to employment or to having a partner. Being incontinent was related to restrictions in all participation domains. IQ was related to type of education and employment, but not to type of residence and having a partner. Corrected for age, the association between continence and residence was not significant for participants younger than 21 years. In general, associations between activity limitations and participation were less pronounced than associations between health condition and participation.

Perceived hindrances in participation

A considerable number of participants reported hindrances in participation (see Table III). Most hindrances were reported in the domain of leisure (range 17–42%), but problems were also reported for visiting family and friends (range 9–32%) and attending work or school (range 3–37%). Problems were

most frequently related to physical impairments, followed by accessibility of buildings, long-distance transportation and emotional distress. Costs were not often mentioned as hindrance in participation.

Problems with long-distance transportation and accessibility of buildings were more often reported by persons with hydrocephalus, with a high level of lesion and who were wheelchair dependent. Participants who were wheelchair dependent reported two to four times more often problems with long distance transportation and accessibility of buildings ($p < 0.005$) and three times more often financial problems hindering leisure activities ($p < 0.001$). Age, gender, type of SB, continence and low IQ were not consistently related to perceived hindrances in participation. Also, occurrence of problems due to own physical impairments, emotional distress or financial restraints were not consistently related to either health condition or activity limitations.

Discussion

This study addresses social participation and its determinants in young adults with SB. The results indicate considerable participation restrictions. Healthy young adults leave their parents' homes at a mean age of 21 years [24], whereas only 28% of the participants older than 21 years in this study were living independently. One-third of the young adults with SB needed special education, against 5% of the general population [25]. The unemployment rate of 53% in this study was also higher than that of disabled persons between 15 and 64 years of age (28%) [26] and far higher than the 8% in the general population aged 15–24 years [24]. The domain of relationships, however, seemed to be less affected by having SB. Participation was related to type of SB, having hydrocephalus, being wheelchair-dependent and being incontinent.

Participants reported most perceived hindrances in participation in the domain of leisure, but they also reported problems regarding going to school or work and visiting family or friends. Physical impairment was most often mentioned as hindrance in participation, but accessibility of buildings, long-distance transportation and own emotional barriers were also frequently mentioned to hinder participation.

The results are similar to those of previous studies on social participation with respect to independent living, education and employment [3,4,10,14–20]. There was a clear relation between wheelchair dependence and perceived hindrances in participation in society, especially accessibility of buildings and long-distance transportation. This finding is not specific for young adults with SB. A recent study in a large representative sample of persons with chronic illness or physical disabilities showed that one-thirds

Table III. Proportion of young adults with SB perceiving hindrances in participation ($N = 179$).

	Work or education (%)	Visiting friends and family (%)	Leisure (%)
Long distance transportation	19*	26* ^{†,‡}	36* ^{†,‡}
Accessibility of building	10 [‡]	32* ^{†,‡,§}	42* ^{†,‡}
physical impairments	37	22 [¶]	40
Emotional distress	20	20	32
Costs	3	9	17* [‡]

Significance level for differences set at a corrected α of $p < 0.003$.

*Significant difference for subgroups of wheelchair dependence.

[†]Significant difference for subgroups of HC.

[‡]Significant difference for subgroups of level of lesions.

[§]Significant difference for subgroups of type of SB.

[¶]Significant difference for subgroups of age.

of persons with disabilities reported problems with steep slopes while entering (or leaving) public buildings, one-fifth reported problems with entering a bus or train, one-third reported that high thresholds and curbstones were reasons for them not to leave the house and 16% reported to experience problems with tight doorways. Furthermore, only 44% of those using special transportation were satisfied with this facility [26].

Contrary to expectations, no relation was found between severity of SB and perceived hindrances due to physical impairments or emotional distress. Apparently, young adults with a less serious SB are, in their own perception, just as restricted in their participation due to pain, fatigue, incontinence and emotional stress (shame, sadness) as young adults with severe SB. This finding confirms the results of Minchom et al., who refuted the assumption that SB would have less psychological impact in mildly disabled young persons [27]. An explanation might be that participants with mild SB more often have a socially active lifestyle or have higher expectations with respect in participation and consequently more acutely perceive hindrances than participants with severe SB.

Some limitations of the present study should be mentioned. The non-response was nearly 40%. However, every effort was made to approach all persons with SB, including those with SB occulta, and to examine every patient whom we were able to trace. Non-response analysis showed that the participating group was comparable with the non-response group on important demographic and illness characteristics. Hence, the findings may be generalised with regard to the population of young adults with SB in The Netherlands. Unavoidably, the group of participants with SB occulta included only patients diagnosed at birth or later in life, mostly because the defect caused physical complaints. Most certain there are many persons with an undiscovered SB occulta. Yet, this study showed that even persons with a less serious SB experienced restrictions in participation.

Second, young adults with an IQ below 70 were not excluded, unlike most other studies. This might have compromised the quality of the self-reported participation data. However, help from parents was allowed, if necessary. We insisted on including these participants because mental retardation is a possible outcome of the neural tube defect and may lead to serious participation problems. Excluding mentally retarded participants would possibly lead to a too optimistic view of the outcome in terms of social participation.

When comparing several possible determinants with outcome measures, a multivariate approach is usually an appropriate way to analyse the data. However, strong relationships between the variables

type of SB, hydrocephalus and level of lesion would have complicated the interpretation of the contribution of the individual predictors in multivariate analysis. The emphasis of this study was on description of the outcome in relevant subgroups and so much on the summed effect of having SB on the outcome of participation.

Our study showed that social participation of persons born with SB is far from optimal. We found that participation restrictions were related to severity of SB and, although to a lesser extent, to activity limitations like wheelchair dependence and incontinence.

Implications

Much more focus should be directed to reduce participation restrictions, especially with respect to leisure activities and mobility for both wheelchair dependent and non-wheelchair dependent young adults with SB. Course of actions must be undertaken like diminution of impact of physical impairments and abolish emotional distress. Levelling of barriers to social participation of wheelchair using persons with SB (and, for that matter, other physically disabled persons) due to inaccessibility of buildings should become a priority of counsellors and governmental authorities.

Acknowledgements

We thank all the patients who participated in this study. We also thank Olga de Wit and Maaike Dekker for their contribution in collecting the data, as well as the BOSK patients' association, the organisations for sheltered homes, and the following centres for their participation in this study: AMC Amsterdam, Franciscusoord Mytylschool, 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. This study was supported by Johanna Children's Fund (Arnhem, the Netherlands) and Stinafo (The Hague, the Netherlands).

References

1. Laurence KM. Effect of early surgery for spina bifida cystica on survival and quality of life. *Lancet* 1974;1:301-304.
2. Ouden AL den, Hirasing RA, Buitendijk SE, Jong-van-den-Berg LT, de Walle HE, Cornel MC. Prevalentie, klinisch beeld en prognose van neuralebuisdefecten in Nederland. Prevalence, clinical aspects and prognosis of neural tube defects in The Netherlands. *Ned Tijdschr Geneesk* 1996; 140:2092-2095.

3. Bowman RM, McLone DG, Grant JA, Tomita T, Ito JA. Spina bifida outcome: a 25-year prospective. *Pediatr Neurosurg* 2001;34:114–120.
4. Hunt GM, Oakeshott P, Kerry S. Link between the CSF shunt and achievement in adults with spina bifida. *J Child Psychol Psychiatry* 1999;67:591–595.
5. Lorber J. Results of treatment of myelomeningocele: an analysis of 524 unselected cases, with special reference to possible selection for treatment. *Dev Med Child Neurol* 1971;13:279–303.
6. McCormick MC, Charney EB, Stemmler MM. Assessing the impact of a child with spina bifida on the family. *Dev Med Child Neurol* 1986;28:53–61.
7. Barf HA, Verhoef M, Jennekens-Schinkel A, Post MWM, Gooskens RHJM, Prevo AJH. Cognitive status of young adults with spina bifida. *Dev Med Child Neurol* 2003;45:813–820.
8. McDonnell GV, McCann JP. Issues of medical management in adults with spina bifida. *Childs Nerv Syst* 2000;16:222–227.
9. Morgan DJ, Blackburn M, Bax M. Adults with spina bifida and/or hydrocephalus. *Postgrad Med J* 1995;71:17–21.
10. Steinbok P, Irvine B, Cochrane DD, Irwin BJ. Long-term outcome and complications of children born with meningo-myelocoele. *Childs Nerv Syst* 1992;8:92–96.
11. Verhoef M, Barf HA, Post MWM, van Asbeck FW, Gooskens RHJM, Prevo AJH. Secondary impairments in young adults with spina bifida. *Dev Med Child Neurol* 2004;46:420–427.
12. Wills KE. Neuropsychological functioning in children with spina bifida and/or hydrocephalus. *J Clin Child Psychol* 1993;22:247–265.
13. Snow JH, Prince M, Souheaver G, Ashcraft E, Stefans V, Edmonds J. Neuropsychological patterns of adolescents and young adults with spina bifida. *Arch Clin Neuropsychol* 1994;9:277–287.
14. Staal-Schreinemachers AL, Vos-Niël JME, Begeer JH. Toekomstperspectieven voor kinderen met spina bifida aperta. Future prospects for children with spina bifida aperta. *Ned Tijdschr Geneesk* 1996;140:1268–1272.
15. Kokkonen J, Saukkonen AL, Timonen E, Serlo W, Kinnunen P. Social outcome of handicapped children as adults. *Dev Med Child Neurol* 1991;33:1095–1100.
16. Hurley AD, Bell S. Educational and vocational outcome of adults with spina bifida in relationship to neuropsychological testing. *Eur J Pediatr Surg* 1994;4(Suppl 1):17–18.
17. Tew B, Laurence KM, Jenkins V. Factors affecting employability among young adults with spina bifida and hydrocephalus. *Z Kinderchir* 1990;45(Suppl 1):34–36.
18. Oakeshott P, Hunt GM. Long-term outcome in open spina bifida. *Br J Gen Pract* 2003;53:632–636.
19. Laurence KM, Beresford A. Degree of physical handicap, education, and occupation of 51 adults with spina bifida. *Br J Prev Soc Med* 1976;30:197–202.
20. Schriener KF, Roessler RT, Johnson P. Identifying the employment concerns of people with spina bifida. *J Appl Rehabil Couns* 1993;24:32–37.
21. World Health Organization. *International Classification of Diseases, 9th revision Dutch edn*. Utrecht: Prismant; 2003.
22. Hoffer MM, Feiwell E, Perry R, Perry J, Bonnett C. Functional ambulation in patients with myelomeningocele. *J Bone Joint Surg Am* 1973;55:137–148.
23. Raven JC. *Standard Progressive Matrices*. Oxford: Oxford Psychologist Press; 1996.
24. Centraal Bureau voor Statistiek (Statistics Netherlands). *STATLINE 2007*. Available from: www.cbs.nl
25. OC&W (Dutch Ministry of Education, Culture and Science). *Onderwijs Cultuur en Wetenschappen in kerncijfers [Education Culture and Sciences: key figures] 2002* Available from: www.minocw.nl/
26. Brink-Muinen Avd, Spreeuwenberg P, Rijken PM. *Kerngegevens Maatschappelijke situatie 2006, Nationaal Panel Chronisch zieken en Gehandicapten Keyfigures Social participation 2006, national panel of chronically ill and disabled persons*. Utrecht: NIVEL; 2007.
27. Minchom PE, Ellis NC, Appleton PL, Lawson V, Boll V, Jones P, Elliott CE. Impact of functional severity on self concept in young people with spina bifida. *Arch Dis Child* 1995;73:48–52.