Lifestyle, participation, and health-related quality of life in adolescents and young adults with myelomeningocele

LAURIEN M BUFFART PHD | RITA J G VAN DEN BERG-EMONS PHD | JETTY VAN MEETEREN MD PHD | HENK J STAM MD PHD FRCP | MARIJ E ROEBROECK PHD

Department of Rehabilitation Medicine, Erasmus MC, University Medical Center, Rotterdam, The Netherlands.

Correspondence to Dr Laurien M Buffart at Erasmus MC, Department of Rehabilitation Medicine, PO Box 2040, 3000 CA Rotterdam, The Netherlands.
E-mail: l.buffart@erasmusmc.nl

PUBLICATION DATA
Accepted for publication 19th December 2008.
Published online 00th Month 2009.

LIST OF ABBREVIATIONS

HRQoL	Health-related quality of life
LIFE-H	Life Habits Questionnaire
MCS	Mental component summary
PCS	Physical component summary
SF-36	36-item Short-form Health Survey

ACKNOWLEDGEMENTS
This study was supported by the Johanna Children's Fund (Arnhem, The Netherlands; grant numbers 20000005/20000158). The authors thank all the participating adolescents and young adults with myelomeningocele. Sten Willemse is acknowledged for his statistical support.

This study aimed to describe participation and health-related quality of life (HRQoL) in adolescents and young adults with myelomeningocele and to explore their relationships with lifestyle-related factors. Fifty-one individuals with a mean age of 21 years 1 month (SD 4y 6mo) years participated (26 males, 25 females; 82% hydrocephalus, 55% wheelchair-dependent). Participation was assessed using the Life Habits Questionnaire, and HRQoL was assessed using the Medical Outcomes Study 36-item Short-form Health Survey. Physical activity was measured using an accelerometry-based activity monitor, fitness (peak oxygen uptake) was measured during a maximal exercise test, and the sum of four skin-folds was assessed to indicate body fat. Relationships were studied using logistic regression analyses. Of the participants, 63% had difficulties in daily activities and 59% in social roles. Participants perceived lower physical HRQoL than a Dutch reference population. Participants with higher levels of physical activity and fitness had fewer difficulties in participating in daily activities (odds ratio [OR]=8.8, p=0.02 and OR=29.7, p=0.02 respectively) and a higher physical HRQoL (OR=4.8, p=0.02 and OR=30.2, p=0.006 respectively), but not mental HRQoL. Body fat was not related to participation or HRQoL. In conclusion, a large proportion of individuals with myelomeningocele had difficulties in participation and perceived low physical HRQoL. Higher levels of physical activity and fitness were related to fewer difficulties in participation and higher physical HRQoL.

The increased life expectancy of people who are born with a physical disability such as myelomeningocele raises new challenges in providing optimal health care and preventing secondary conditions. In addition to medical problems such as urinary and faecal incontinence and pressure sores, and orthopaedic problems such as scoliosis and contractures,[1] many adolescents and young adults with myelomeningocele have a physically inactive lifestyle, and have low aerobic fitness and high levels of body fat.[2] Promoting healthy lifestyles by improving levels of physical activity and aerobic fitness, and by reducing excessive body fat, may benefit participation and health-related quality of life (HRQoL) in people with physical disabilities such as myelomeningocele.

Participation is an objective outcome of involvement in life situations and can be evaluated by assessing life habits, i.e. activities of daily living and social roles, and by addressing difficulty of performance and the assistance required.[3] Many adolescents and young adults with myelomeningocele are independent in most functional activities but report dependency for sphincter control, locomotion, and self-care,[4,5] particularly those with hydrocephalus and higher lesion levels.[5] Furthermore, many adolescents with myelomeningocele show difficulties with responsibilities in household tasks, decision making, money management, and engagement in activities with peers.[4]

HRQoL is a self-perceived outcome, based on subjective reports on various areas of health.[6] In the literature, HRQoL has also been referred to as self-reported health status[7] or perceived health.[8] Results of studies on HRQoL of adolescents and young adults with myelomeningocele are equivocal. Some studies showed that they perceived

lower levels of HRQoL compared with the able-bodied population, particularly regarding aspects of physical functioning, including self-care, continence, and mobility,[8,9] but also in emotional, social, and school domains.[9] Other studies showed overall good HRQoL.[10,11]

Little information is available on whether improving lifestyle may benefit participation and HRQoL in adolescents and young adults with myelomeningocele. Studies in individuals with other chronic conditions, such as spinal cord injury and cerebral palsy (CP), showed that physical activity and aerobic fitness were positively associated with participation and HRQoL.[12,13] In able-bodied adolescents and young adults, obesity was associated with lower HRQoL, but no difference was found in HRQoL between obese and non-obese individuals with myelomeningocele.[9]

Because of scarce and equivocal information, the first purpose of the study was to describe participation and HRQoL in a sample of adolescents and young adults with myelomeningocele. Second, in order to examine whether lifestyle interventions have the potential to improve participation and HRQoL in individuals with myelomeningocele, this study explored whether physical activity, aerobic fitness, and body fat were associated with participation and HRQoL.

METHOD

Participants

Adolescents and young adults, aged between 16 and 30 years, were recruited from four university hospitals in the western part of the Netherlands (Rotterdam, Leiden, Utrecht, and Amsterdam) and all rehabilitation centres in the south-west of the country. Exclusion criteria were the following: inability to understand the measurements performed in this study (as judged by the treating physician or a family member); complete dependence on a powered wheelchair; presence of disorders other than myelomeningocele that affect physical activity (e.g. rheumatoid arthritis); and contraindication for a maximal exercise test. We invited 171 individuals, of whom 51 participated in the study (response 30%). The main reasons for non-participation were lack of interest, or time or duration of the measurements. No difference was found between participants and non-participants regarding age, sex, lesion level, and presence of hydrocephalus, as tested with an independent *t*-test or χ^2 test.[2] All participants and parents of adolescents aged less than 18 years gave written informed consent before participating in the study. The medical ethics committees of Erasmus MC and of all the participating institutes approved the study. Information about the neurological level of lesion and presence of hydrocephalus was obtained from medical records. Hydrocephalus was considered to be present when a shunt had been placed. In two participants, hydrocephalus was mentioned in the medical record at some time, but no shunt was placed. We assumed that this was only minor hydrocephalus and therefore we categorized these participants as not having hydrocephalus.[1]

According to the classification of Hoffer,[14] participants were categorized as ambulatory (including community ambulators, *n*=15, and household ambulators, *n*=8) or non-ambulatory (*n*=28, also including non-functional ambulators). Education level was categorized as low (pre-vocational practical education or lower level) or high (secondary theoretical education and higher education).

Participation

Participation was assessed using the Life Habits Questionnaire (LIFE-H).[15] The short version, the LIFE-H 3.0, consists of 69 life habits covering the following 12 categories: nutrition, fitness, personal care, communication, housing, mobility, responsibilities, interpersonal relationships, community life, education, employment, and recreation. The first six categories refer to daily activities whereas the others are associated with social roles. The scoring is based on two specific elements: (1) the degree of difficulty in performing life habits (no difficulty, with difficulty, with substitution, or not accomplished); and (2) the type of assistance required performing the habit (no help, technical assistance or adaptation, human assistance). Both elements are combined in a scale ranging from 0 to 9, with 0 indicating total handicap (or no participation; the activity or social role is not accomplished or achieved) and 9 indicating optimal participation (the activity is performed without difficulty and without help). The mean scores for the two subdomains (i.e. daily activities and social roles) were calculated. A mean score of 8 or higher indicates no difficulty in performance either with or without assistance or assistive devices, and a score below 8 indicates difficulty in performance.

Health-related quality of life

The Medical Outcomes Study 36-item Short-form Health survey (SF-36)[7,16] was used to assess HRQoL in several domains: physical functioning, social functioning, role limitations due to physical problems, role limitations due to emotional problems, pain, mental health, vitality, and general health perception. All raw scores were linearly converted to a 0 to 100 scale providing sum scores for each domain. In addition, scores were summarized in two main scores, which were: a physical component summary (PCS) and a mental component summary (MCS). Higher domain and summary scores indicate higher levels of functioning or well-being.[16]

Lifestyle-related factors

Physical activity

The duration of dynamic activities (composite measure of the separately detected activities walking, including walking on stairs and running, cycling, general movement, and wheelchair-driving) was objectively assessed during two consecutive weekdays using an accelerometry-based activity monitor (Vitaport 3, Temec Instruments, Kerkrade, the Netherlands). The activity monitor has been shown to be a valid and reliable instrument to quantify mobility-related activities, including wheelchair-driving.[17,18] Participants were fitted with the activity monitor at their homes and were instructed to perform their usual daily activities, but not to swim or take a shower or bath during activity monitoring. Detailed procedures are described elsewhere.[2] The level of physical activity was defined as the average duration of dynamic activities on the first and second measurement day, expressed in minutes/day.

Aerobic fitness

Aerobic fitness was assessed during a progressive maximal exercise test, based on the McMaster All-Out Progressive Continuous Cycling and Arm test[19] Depending on whether the main mode of ambulation was walking or wheelchair-driving, participants performed the test on an electronically braked cycle ergometer (Jaeger ER800; Jaeger Toennies, Breda, the Netherlands) or arm ergometer (Jaeger ER800SH) respectively. During the test, the resistance was increased every 2 minutes with a variable load (ensuring that the total exercise duration ranged from 8 to 12 min) until exhaustion. Detailed descriptions of the test can be found elsewhere.[20] Gas exchange was determined continuously using a breath-by-breath portable measurement system (K4b^2, COSMED, Rome, Italy). Aerobic fitness was defined as the mean oxygen uptake during the last 30 seconds of exercise (peakVO_2, in l/min).

Body fat

The thickness of four skin-folds (biceps, triceps, subscapular, suprailiac) was measured twice on the right side of the body with a calliper (Harpenden, Burgess Hill, UK) and the average sum (in mm) was used as an indicator of body fat. This technique was considered valid based on 2 to 3% differences with weighing under water.[21] Data of two non-ambulators were reported as missing because the skin-fold thickness was beyond that of the calliper jaws (80mm).

Statistical analyses

Results are presented as mean and SD or percentages of the group. In addition, group percentages of adolescents and young adults with difficulties in daily activities and social roles (score <8[22]) are presented. Sum scores of the eight domains of the SF-36 were compared with the mean scores of a Dutch reference population aged 16 to 40 years[7] using a one-sample *t*-test. The PCS and MCS were compared with a Dutch reference sample aged 20 to 29 years.[23]

Because data on participation and HRQoL were not normally distributed among our study sample, the outcome measures were dichotomized. The subdomains daily activities and social roles of the LIFE-H were dichotomized into difficulty (0) and no difficulty (1). Because clinically relevant cut-offs for good and poor PCS and MCS scores on the SF-36 are unknown, the median score (44.0 for PCS and 57.8 for MCS) was used to identify individuals with relatively low HRQoL (0) and high HRQoL (1) within the study sample.

Separate logistic regression analyses were conducted to model the outcome measures. For each outcome measure, the independent variables physical activity, aerobic fitness, and body fat were included one at a time to accommodate the small sample size. The following variables were checked for potential confounding: age, sex, ambulatory status, and level of education. Age and level of education were not confounders and, therefore, these two personal factors were studied as separate independent variables for each outcome measure. Sex (0=female; 1=male) and ambulatory status (0=non-ambulator; 1=ambulator) were confounders, adjusted for in the analyses. In the relationships with aerobic fitness, ambulatory status was used as a proxy for type of ergometer because of the high collinearity between the two variables.[20] Adjusted odds ratios (OR), including 95% confidence intervals, were presented. Nagelkerke R squares (R^2) were reported as an indicator of goodness of fit and $p \leq 0.05$ was considered significant.

RESULTS

Table I presents the personal characteristics of the participants and the results of the physical activity, aerobic fitness, and body fat assessments.

Participation

Overall, 63% of the participants had difficulties in daily activities, mainly in personal care, housing, and mobility, and 59% had difficulties in social roles, in particular with recreation and employment (Table II). Several personal factors were related to participation (Table III). Males were more likely to perceive no difficulty in daily activities than females (OR=7.1; p=0.004). Ambulators were more likely to perceive no difficulty in daily activities (OR=11.3; p<0.001) and social roles (OR=4.7; p<0.001) than non-ambulators. Adjusted for sex and ambulatory status, indi-

Table I: Descriptive results for personal and lifestyle-related factors of adolescents and young adults with myelomeningocele (*n*=51)

Personal factors (%)	
Sex, male	51
Level of lesion[a]	
Sacral	14
Lumbosacral	41
Lumbar	29
Thoracolumbar	14
Thoracic	2
Hydrocephalus[a]	82
Ambulators	45
Educational level, low[b]	33
Age, y mean (SD)	21.1 (4.5)
Lifestyle-related factors (mean [SD])	
Physical activity (min/day)[c]	81 (62)
Aerobic fitness, peakVO_2 (l/min)[d]	1.48 (0.52)
Body fat, sum of four skin-folds (mm)[c]	74.4 (38.5)

[a]As reported in medical records; [b]pre-vocational practical education or lower level; [c]*n*=49; [d]*n*=50.

viduals with a higher education level had fewer difficulties with social roles (OR=5.0; p=0.03).

Health-related quality of life

The mean PCS score was 43.1 (range 24.0–58.9) and the mean MCS score was 55.0 (range 24.8–68.8; Table II). Adolescents and young adults with myelomeningocele perceived lower physical HRQoL than a Dutch reference sample (mean PCS score = 53.2; p<0.001), particularly in the domains physical functioning and general health. In general, they had higher scores on mental HRQoL than a Dutch reference sample (mean MCS score = 47.8; p<0.001), except for the domains vitality and social functioning.

Several personal factors were associated with HRQoL (Table IV). Females were 3.4 times more likely to perceive lower physical HRQoL than males (p=0.04). Nine males and 16 females had low physical HRQoL. Furthermore, low physical HRQoL was found in seven ambulators and 18 non-ambulators. Non-ambulators were 4.1 times more likely to perceive lower physical HRQoL than ambulators (p=0.02). Adjusted for sex and ambulatory status, a higher age was associated with a lower mental HRQoL (OR=0.86; p=0.04).

Lifestyle, participation, and health-related quality of life

Tables III and IV present the relationships between lifestyle and participation and between lifestyle and HRQoL respectively. Adjusted for sex and ambulatory status, individuals with higher levels of physical activity had fewer difficulties in daily activities (OR=8.8; p=0.02) and perceived a higher physical HRQoL (OR=4.8; p=0.02). Individuals with higher peakVO_2 had fewer difficulties in daily activities (OR=29.7; p=0.02) and perceived a higher physical HRQoL (OR=30.2; p<0.006). Lifestyle-related factors were not associated with mental HRQoL.

Table II: Participation and health-related quality of life of adolescents and young adults with myelomeningocele (MMC; *n*=51)

Participation (Life-H)	Mean (SD)	Difficulties (score <8), %
Daily activities		
Nutrition	8.0 (1.4)	29
Fitness	7.8 (1.2)	36
Personal care	7.3 (1.7)	53
Communication	8.4 (0.8)	33
Housing	7.2 (1.2)	75
Mobility	6.5 (1.6)	74
Daily activities sub-domain	7.5 (1.0)	*63*
Social roles		
Responsibility	8.1 (1.8)	22
Relationships	8.7 (0.6)	10
Community	7.4 (2.0)	41
Recreation	6.3 (1.8)	82
Education	7.4 (2.3)	32
Employment	7.5 (1.2)	56
Social roles sub-domain	7.5 (1.2)	59

Health-related quality of life (SF-36)	MMC, mean (SD)	Reference values,[a] mean	*p*-value
Domain			
Physical functioning	46.8 (23.7)	93.1	<0.001
Role physical	77.8 (34.1)	86.4	0.08
Bodily pain	84.8 (20.1)	80.9	0.18
General health	69.2 (23.7)	78.2	0.01
Vitality	66.4 (15.9)	70.7	0.06
Social functioning	81.1 (24.8)	87.8	0.06
Role emotional	85.0 (29.3)	85.4	0.92
Mental health	77.3 (16.7)	78.7	0.54
Physical component summary	43.1 (9.0)	53.2	<0.001
Mental component summary	55.0 (9.7)	47.8	<0.001

[a]Domain scores were compared with a Dutch reference population aged 16 to 40 years from Aaronson et al.[7] and the summary scores with a Dutch reference population aged 20 to 29 years from Blokstra et al.[23] Life-H, Life Habits Questionnaire; SF-36, 36-item short Form Health Survey.

DISCUSSION

The present study showed that many adolescents and young adults with myelomeningocele had difficulties in

Table III: Relationships between personal and lifestyle-related factors and participation in adolescents and young adults with myelomeningocele (*n*=51)

	Daily activities					Social roles				
			Logistic regression analyses					Logistic regression analyses		
	Difficulty (*n*=32)	No difficulty (*n*=19)	OR (95% CI)[c]	*p*	R^2	Difficulty (*n*=30)	No difficulty (*n*=21)	OR (95% CI)[c]	*p*	R^2
Univariate regression										
Sex, *n* (%) male	11 (37)	15 (79)	7.1 (1.9, 26.9)	0.04	0.24	14 (47)	12 (57)	1.5 (0.5, 4.7)	0.46	0.01
Ambulatory status, *n* (%) non-ambulatory	24 (75)	5 (21)	11.3 (2.9, 43.9)	<0.001	0.34	21 (70)	7 (33)	4.7 (1.4, 15.4)	0.01	0.17
Multivariate regression										
Personal factors										
Education, *n* (%) low	14 (44)	3 (16)	4.1 (0.8, 24.4)	0.09	0.55	14 (47)	3 (14)	5.0 (1.1, 22.3)	0.03	0.29
Age, mean (SD) y	21.1 (4.7)	21.1 (4.2)	1.07 (0.89, 1.28)	0.49	0.51	20.3 (4.5)	22.2 (4.4)	1.13 (0.97, 1.33)	0.12	0.23
Lifestyle-related factors										
Physical activity, mean (SD) min/day[a]	56.8 (34.4)	122.1 (77.4)	8.8 (1.4, 54.2)[d]	0.02	0.61	61.8 (35.5)	108.3 (81.2)	2.2 (0.8, 5.6)[d]	0.11	0.22
Aerobic fitness (peakVO_2), mean (SD) l/min[b]	1.21 (0.39)	1.93 (0.42)	29.7 (1.7, 506.7)	0.02	0.67	1.32 (0.43)	1.72 (0.59)	5.3 (0.8, 33.0)	0.08	0.28
Body fat (sum of four skin-folds), mean (SD) mm[a]	85.0 (37.6)	57.7 (34.4)	1.00 (0.97, 1.03)	0.99	0.48	78.3 (36.6)	68.8 (41.3)	1.00 (0.98, 1.03)	0.71	0.20

[a]*n*=49; [b]*n*=50; [c]odds ratios (OR) adjusted for sex and ambulatory status for multivariate regression analyses. [d]In the regression analyses, physical activity is expressed in hours per day. CI, confidence interval; R^2, Nagelkerke R square.

Table IV: Relationships between personal and lifestyle-related factors and health-related quality of life in adolescents and young adults with myelomeningocele (*n*=51)

	Physical component summary					Mental component summary				
			Logistic regression analyses					Logistic regression analyses		
	Low (*n*=25)	High (*n*=26)	OR (95% CI)[c]	*p*	R^2	Low (*n*=25)	High (*n*=26)	OR (95% CI)[c]	*p*	R^2
Univariate regression										
Sex, *n* (%) male	9 (36)	17 (65)	3.4 (1.1, 10.6)	0.04	0.11	12 (48)	14 (54)	1.1 (0.4, 3.3)	0.89	0.06
Ambulatory status, *n* (%) non-ambulatory	18 (72)	10 (39)	4.1 (1.3, 13.4)	0.02	0.15	11 (44)	17 (65)	0.5 (0.2, 1.5)	0.20	0.17
Multivariate regression										
Personal factors										
Education, *n* (%) low	8 (32)	9 (35)	0.7 (0.2, 2.7)	0.64	0.22	7 (28)	10 (39)	0.7 (0.2, 2.4)[c]	0.57	0.08
Age, mean (SD) y	22.4 (4.6)	19.8 (4.1)	0.87 (0.75, 1.00)	0.06	0.30	22.5 (4.6)	19.7 (4.1)	0.86 (0.74, 1.00)	0.04	0.18
Lifestyle-related factors										
Physical activity, mean (SD) min/day[a]	54.4 (33.4)	108.2 (73.4)	4.8 (1.3, 17.8)[d]	0.02	0.40	83.0 (74.9)	78.6 (48.6)	1.1 (0.6, 2.2)[d]	0.71	0.07
Aerobic fitness ($peakVO_2$), mean (SD) l/min[b]	1.17 (0.39)	1.78 (0.48)	30.2 (2.7, 342.1)	0.006	0.47	1.53 (0.59)	1.44 (0.48)	0.81 (0.2, 3.7)	0.78	0.09
Body fat (sum of four skin-folds), mean (SD) mm[a]	82.6 (35.0)	67.2 (40.6)	1.00 (0.98, 1.03)	0.73	0.19	69.7 (32.9)	78.6 (43.0)	1.00 (0.99, 1.03)	0.52	0.10

[a]*n*=49; [b]*n*=50, [c]odds ratios (OR) adjusted for sex and ambulatory status for multivariate regression analyses. [d]In the regression analyses, physical activity is expressed in hours per day. CI, confidence interval; R^2, Nagelkerke R square.

daily activities and social roles and that they perceived lower physical HRQoL than a Dutch reference sample. Individuals with higher levels of physical activity and aerobic fitness had fewer difficulties in daily activities and perceived a higher physical HRQoL.

Participation

The finding that a large proportion of individuals with myelomeningocele had difficulties in participation, particularly regarding self-care, housing, mobility, recreation, and employment, is in line with previous findings on dependency in various activities of daily living[4,5] and subnormal levels of decision making and participation.[4] Also, compared with adolescents and young adults with CP with normal intelligence, individuals with myelomeningocele had more difficulties in participation; 21% of individuals with CP had difficulties in daily activities and 24% in social roles[22] compared with 63% and 59% in individuals with myelomeningocele respectively.

Individuals who were wheelchair-dependent and individuals with a low education level had more difficulties in participation. Educational support was also found to be important for work participation of young adults with myelomeningocele.[24] In comparison, in adolescents and young adults with CP, participation restrictions were mainly attributable to restricted gross motor functioning and low education level.[22] In addition to these associated factors, to improve participation, it might be relevant to focus on modifiable lifestyle-related factors.

Health-related quality of life

Lower physical HRQoL than reference values confirms previous results.[8,9] Also, the results on the different domains of mental HRQoL were comparable to the results from young adults with spina bifida aperta and hydrocephalus from the national ASPINE study.[8] Non-ambulators were more likely to perceive lower physical HRQoL, which is in line with previous findings that individuals with myelomeningocele with lower mobility and self-care ability perceived lower physical HRQoL.[25] Mental HRQoL was not associated with ambulatory status. This is in contrast to the study of Padua et al. in a small sample of adolescents with spina bifida, who found that higher mobility and self-care ability was correlated with higher psychological distress and more severe role disability because of emotional problems.[25]

Individuals with myelomeningocele perceived higher physical HRQoL than adults with paraplegia, who had an average PCS score of 29.[26] In contrast to individuals with myelomeningocele, who are restricted in mobility from birth, mobility problems of people with paraplegia are acquired at later ages, which may result in lower perceived physical HRQoL. Furthermore, different results may be related to age. The participants in our study were younger. Older individuals with myelomeningocele perceived lower mental and physical HRQoL, which underlines the need to pay attention to improving HRQoL when patients are growing into adulthood.

Lifestyle and participation

The present findings that higher levels of physical activity and aerobic fitness were associated with fewer difficulties in participation are in line with previous results in adults with spinal cord injury showing higher levels of physical activity to be associated with lower participation and disability.[27] Our study does not support the suggestion of Manns et al. that physical activity may play a more important role in participation than aerobic fitness.[27] On the contrary, our results indicated that aerobic fitness not only was associated with participation in daily activities but also tended to have a relationship with social roles, whereas physical activity did not. It is possible that increasing aerobic fitness may reduce physical strain and associated fatigue, which may result in increased performance in daily activities and social roles. Because of the cross-sectional study design, causality cannot be established and future studies should confirm whether improving levels of physical activity and aerobic fitness has positive effects on the performance of daily activities and social roles.

Lifestyle and health-related quality of life

The finding that physical activity and aerobic fitness were positively associated with physical HRQoL illustrates the importance of addressing physical activity and fitness in the health care of adolescents and young adults with chronic conditions such as myelomeningocele. However, causality needs to be established in future studies, since it may also be that participants were more physically active because they perceived a higher physical HRQoL. The median was used as an unbiased cut-off between relatively lower and higher HRQoL within the study sample. Future studies with larger sample sizes are warranted to study clinically meaningful cut-off points between good and poor HRQoL in adolescents and young adults with myelomeningocele.

No relationship was found between HRQoL and body fat, which is in accordance with previous findings in individuals with myelomeningocele, but in contrast with findings in the able-bodied population.[9] Furthermore, unlike physical HRQoL, mental HRQoL was not associated with levels of physical activity and aerobic fitness, suggesting that mental HRQoL may be more influenced by factors other than lifestyle-related factors. Parental hope and other family factors, positive attitudes, communication efficacy, coping strategies, and social support were suggested to be important.[10,11]

Study limitations

A selection bias may have occurred in that the more active and fitter individuals with myelomeningocele had a higher interest in participating in this study. This may lead to problems in generalizability, since it may have underestimated participation difficulties and overestimated HRQoL in this population. This may also explain the finding that participants in the present study perceived higher levels of physical functioning and less pain compared with participants of the ASPINE study. The hypothesis that individuals with lower participation and HRQoL may benefit more from improved physical activity levels and aerobic fitness should be tested in future longitudinal studies.

Another limitation was the relatively small sample size and, consequently, low statistical power. This may have resulted in the large confidence intervals. The results will have to be interpreted with some caution because the sample size was rather small with regard to the number of independent variables (n=3) in the regression models.

The strength of monitoring physical activity with the activity monitor is that it provides detailed objective information on mobility-related activities in both ambulatory and non-ambulatory individuals. Its weakness lies in the 2-day monitoring, which hampered measuring an individual's actual activity pattern. However, analyses of variance showed that the variance between the two measurement days was small enough to be able to distinguish active and inactive people (intraclass correlation coefficient = 0.89). Therefore, we assumed that the relationships from the present study were reasonable.

Furthermore, the present study focused on self-perceived HRQoL, rather than on generic quality of life and, therefore, does not include concepts such as life satisfaction or well-being. Results of the national ASPINE study showed that adolescents and young adults with spina bifida had similar overall life satisfaction as a population reference group, but had lower satisfaction with their sex life, partnership relations, and self-care ability.[28]

In conclusion, a large proportion of adolescents and young adults with myelomeningocele, in particular non-ambulators and females, had participation difficulties and perceived lower physical HRQoL compared with reference values of the general population. People with higher levels

of physical activity and fitness had fewer participation difficulties and higher physical HRQoL. These findings illustrate that physical activity and fitness should be addressed in health care. Whether improving levels of physical activity and fitness improves participation and HRQoL needs to be confirmed in future studies.

The following members of the Transition Research Group South West Netherlands contributed to this study:

W G M Janssen MD, Department of Rehabilitation Medicine, University Medical Center Rotterdam; M P Bergen MD PhD, D Spijkerman MD, Rijndam Rehabilitation Center; M Rol MD, Sophia Rehabilitation, The Hague and Delft; H vd Heijden-Maessen MD, Rijnlands Rehabilitation Center, Leiden; H J R Buijs MD, Rehabilitation Center 'De Waarden', Dordrecht, Th Voogt MSc, Foundation of Rehabilitation Medicine Zeeland, Goes; and J H Arendzen MD PhD, M S van Wijlen-Hempel MD PhD, Department of Rehabilitation Medicine, Leiden University Medical Center. In addition, F W van Asbeck MD PhD and M W Post PhD the University Medical Center Utrecht, and C J McDonald-ten Thij, VU University Medical Center collaborated in the study.

REFERENCES

1. Verhoef M, Barf HA, Post MW, van Asbeck FW, Gooskens RH, Prevo AJ. Secondary impairments in young adults with spina bifida. *Dev Med Child Neurol* 2004; **46:** 420–27.
2. Buffart LM, Roebroeck ME, Rol M, Stam HJ, van den Berg-Emons RJ. Triad of physical activity, aerobic fitness and obesity in adolescents and young adults with myelomeningocele. *J Rehabil Med* 2008; **40:** 70–75.
3. Noreau L, Fougeyrollas P. Long-term consequences of spinal cord injury on social participation: the occurrence of handicap situations. *Disabil Rehabil* 2000; **22:** 170–80.
4. Buran CF, Sawin KJ, Brei TJ, Fastenau PS. Adolescents with myelomeningocele: activities, beliefs, expectations, and perceptions. *Dev Med Child Neurol* 2004; **46:** 244–52.
5. 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:** 114–19.
6. Livingston MH, Rosenbaum PL, Russell DJ, Palisano RJ. Quality of life among adolescents with cerebral palsy: what does the literature tell us? *Dev Med Child Neurol* 2007; **49:** 225–31.
7. Aaronson NK, Muller M, Cohen PD, et al. Translation, validation, and norming of the Dutch language version of the SF-36 Health Survey in community and chronic disease populations. *J Clin Epidemiol* 1998; **51:** 1055–68.
8. Verhoef M, Post MW, Barf HA, van Asbeck FW, Gooskens RH, Prevo AJ. Perceived health in young adults with spina bifida. *Dev Med Child Neurol* 2007; **49:** 192–97.
9. Abresch RT, McDonald DA, Widman LM, McGinnis K, Hickey KJ. Impact of spinal cord dysfunction and obesity on the health-related quality of life of children and adolescents. *J Spinal Cord Med* 2007; **30:**(Suppl. 1) S112–18.
10. Kirpalani HM, Parkin PC, Willan AR, Fehlings DL, Rosenbaum PL, King D, Van Nie AJ. Quality of life in spina bifida: importance of parental hope. *Arch Dis Child* 2000; **83:** 293–97.
11. Sawin KJ, Brei TJ, Buran CF, Fastenau PS. Factors associated with quality of life in adolescents with spina bifida. *J Holist Nurs* 2002; **20:** 279–304.
12. Devillard X, Rimaud D, Roche F, Calmels P. Effects of training programs for spinal cord injury. *Ann Readapt Med Phys* 2007; **50:** 490–498, 480–499.
13. Verschuren O, Ketelaar M, Gorter JW, Helders PJ, Uiterwaal CS, Takken T. Exercise training program in children and adolescents with cerebral palsy: a randomized controlled trial. *Arch Pediatr Adolesc Med* 2007; **161:** 1075–81.
14. Hoffer MM, Feiwell E, Perry R, Perry J, Bonnet C. Functional ambulators in patients with myelomeningocele. *J Bone Joint Surg Am* 1973; **55:** 137–48.
15. Fougeyrollas P, Noreau L, St-Michel G. Life Habits measure – shortened version (LIFE-H 3.0). Lac St-Charles, Quebec, Canada: COCIDIH, 2001.
16. Ware JE. SF-36 Physical and mental health summary scales: a user's manual. Boston, MA: The Health Institute, 1994.
17. Bussmann JB, Martens WL, Tulen JH, Schasfoort FC, van den Berg-Emons HJ, Stam HJ. Measuring daily behavior using ambulatory accelerometry: the Activity Monitor. *Behav Res Methods Instrum Comput* 2001; **33:** 349–56.
18. Postma K, van den Berg-Emons HJ, Bussmann JB, Sluis TA, Bergen MP, Stam HJ. Validity of the detection of wheelchair propulsion as measured with an activity monitor in patients with spinal cord injury. *Spinal Cord* 2005; **43:** 550–57.
19. Bar-Or O. Pediatric sports medicine for the practitioner. From physiologic principles to clinical applications. New York: Springer Verlag, 1983: 315–41.
20. Buffart LM, van den Berg-Emons RJ, van Wijlen-Hempel MS, Stam HJ, Roebroeck ME. Health-related physical fitness of adolescents and young adults with myelomeningocele. *Eur J Appl Physiol* 2008; **103:** 181–88.
21. Mita K, Akataki K, Itoh K, Ono Y, Ishida N, Oki T. Assessment of obesity of children with spina bifida. *Dev Med Child Neurol* 1993; **35:** 305–11.
22. Donkervoort M, Roebroeck M, Wiegerink D, van der Heijden-Maessen H, Stam H. The Transition Research Group South West N. Determinants of functioning of adolescents and young adults with cerebral palsy. *Disabil Rehabil* 2007; **29:** 453–63.
23. Blokstra A, Smit H, Bueno de Mesquita HB, Seidell JC, Verschuren WMM. Monitoring van risicofactoren en gezondheid in Nederland (Morgen-project), 1993-1997. Leefstijl- en risicofactoren: prevalenties en trends. Bilthoven: Rijksinstituut voor de volksgezondheid en milieu (RIVM); 2005. Report No.: 263200008/2005.

24. 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:** 772–77.
25. Padua L, Rendeli C, Rabini A, Girardi E, Tonali P, Salvaggio E. Health-related quality of life and disability in young patients with spina bifida. *Arch Phys Med Rehabil* 2002; **83:** 1384–88.
26. Fisher CG, Noonan VK, Smith DE, Wing PC, Dvorak MF, Kwon BK. Motor recovery, functional status, and health-related quality of life in patients with complete spinal cord injuries. *Spine* 2005; **30:** 2200–07.
27. Manns PJ, Chad KE. Determining the relation between quality of life, handicap, fitness, and physical activity for persons with spinal cord injury. *Arch Phys Med Rehabil* 1999; **80:** 1566–71.
28. 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:** 458–63.