Physical Activity and Fitness in Adolescents and Young Adults with Myelomeningocele

Laurien Buffart

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Physical Activity and Fitness in Adolescents and Young Adults with Myelomeningocele

Lichamelijke Activiteit en Fitheid bij Jongeren en Jongvolwassenen met Meningomyelocèle

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Contents

Chapter 1	General introduction	7
Chapter 2	Triad of physical activity, aerobic fitness and obesity in adolescents and young adults with myelomeningocele	15
Chapter 3	Health-related physical fitness of adolescents and young adults with myelomeningocele	31
Chapter 4	Energy cost and physical strain of daily activities in adolescents and young adults with myelomeningocele	49
Chapter 5	Sports participation in adolescents and young adults with myelomeningocele and its role in total physical activity behaviour and fitness	63
Chapter 6	Lifestyle, participation and health-related quality of life in adolescents and young adults with myelomeningocele	81
Chapter 7	Cardiovascular disease risk factors and the relations with physical activity, aerobic fitness, and body fat in adolescents and young adults with myelomeningocele	97
Chapter 8	Barriers and facilitators of physical activity in adolescents and young adults with physical disabilities	117
Chapter 9	Intervention 'Active Lifestyle and Sports Participation'. Promoting physical activity in adolescents and young adults with childhood-onset physical disabilities	141
Chapter 10	General discussion	159
	Summary	173
	Samenvatting	179
	Dankwoord	185
	Curriculum vitae	189
	List of publications	190

Chapter 1

General Introduction

Spina bifida

Spina bifida means a 'split' or 'divided spine'.[1] It is a birth defect resulting from a failure in the closure of the neural tube. The development and closure of the neural tube are normally completed within 28 days after conception.[2,3] Spina bifida may be open or closed.[3,4] Closed spina bifida, or 'spina bifida occulta' refers to those cases in which the distal embryonic neural tube is not normal elongated. It is often present without neurological symptoms. However, there may be a defect in the vertebrae, or the skin overlying the defect may be dimpled, thickened, pigmented or hairy. In the open form of spina bifida, 'spina bifida aperta', the neural tube defect is not covered with skin. Two forms are distinguished: meningocele and myelomeningocele (MMC), where 'myelo' refers to neural tissue and 'cele' means 'swelling'.[1] Meningo refers to the meninges which are the surrounding membranes of the central nervous system. MMC is the most severe form of spina bifida, in which, unlike in meningocele, the cele of meningeal elements also contains neural tissue. MMC is often associated with brain anomalies including hydrocephalus and Arnold-Chiari type II malformations.[3]

Spina bifida has a world-wide prevalence of 4.7 per 10000 live births,[3] of which MMC is most common. In 1998, 112 children were born with spina bifida in the Netherlands.[5] Before 1960, as few as 10% of infants born with spina bifida survived, with most dying from infection or hydrocephalus.[3] After shunt improvements and more advanced treatment survival rates increased, and it is estimated that at least 50 to 75% of children born with spina bifida will nowadays survive into adulthood.[6,7]

In the Netherlands, there are 12 multidisciplinary teams, mostly focusing on treatment of children with spina bifida. Little is known about the functioning and health of young adults with spina bifida. Therefore, the ASPINE study (Adolescents with SPina bifida in the NEtherlands) was performed to explore physical functioning, cognitive functioning, independence in daily living, functioning in the community, perceived health and life satisfaction, as well as the present care and need for care.[8] This study showed that many adolescents and young adults with spina bifida have medical problems including urinary and faecal incontinence, obstipation, foot deformities and scoliosis.[9] Many are restricted in mobility and need walking aids (e.g. orthotics, crutches or canes) or wheelchairs,[9] which puts them at increased risk to develop an inactive lifestyle, and thus an unhealthy lifestyle. The latter topic was not incorporated in the ASPINE study and therefore we launched the current study to be able to further optimize health care for adolescents and young adults with MMC. In contrast to the ASPINE study, which included people with spina bifida occulta and aperta, we only focus on people with MMC.

Lifestyle in adolescents and young adults with MMC

Due to increased life expectancy of persons with MMC, lifestyle related diseases are of increasing concern. Similarly to persons with other chronic conditions, there is a shift from disability prevention towards prevention of secondary conditions.[10] The Healthy People 2010 report aims at promoting health and preventing illness, disability and premature death.[11] It emphasises that regular physical activity throughout life is important for maintaining a healthy body.[11] Health benefits include a lower risk of hypertension, diabetes, cardiovascular disease, some forms of cancer as well as improvement in weight loss or maintenance, physical fitness and quality of life.[12,13] In the Netherlands, only 52% of the general adult population meets the Dutch physical activity norm[14] of 30 minutes of moderate physical activity for at least 5 days per week.[15] In people with a disability, a physical inactive lifestyle is even more common than in the general population.[11,16,17] Therefore, they are at risk to experience a vicious cycle: inactivity leading to a reduction in aerobic fitness and an increase in body fat. This may result in higher physical strain during the performance of activities, leading to further inactivity.[18]

In adolescents and young adults with MMC, objective data on physical activity are scarce. Previously, a study in 14 adolescents and young adults with MMC was performed at our department in which physical activity was measured objectively with an accelerometry-based activity monitor (AM).[19] The AM consists of four uniaxial piezo-resistive accelerometers of which 2 were attached to the sternum and 1 to each thigh. In individuals who used a wheelchair, an additional accelerometer was attached to each wrist.[20] The accelerometers were connected to a portable data recorder, which was worn in a padded bag around the waist. The combination of accelerometers makes it possible to detect which postures (lying, sitting, standing), movements (walking, including walking stairs and running, cycling, wheelchair-driving, general non-cyclic movement) and transitions between postures are performed, how often and for how long.[19,20] AM measurements showed that adolescents and young adults with MMC spent on average 91 minutes on dynamic activities (composite measure of the separately detected activities walking, cycling, wheelchair-driving and general non-cyclic movement) per day whereas 178 minutes were registered in their able-bodied peers.[21] Furthermore, Bandini and co-workers measured energy expenditure in 16 adolescents with MMC and showed that the amount of energy expended on activities was similar between ambulatory adolescents with MMC and able-bodied controls, but lower in non-ambulatory adolescents with MMC.[22]

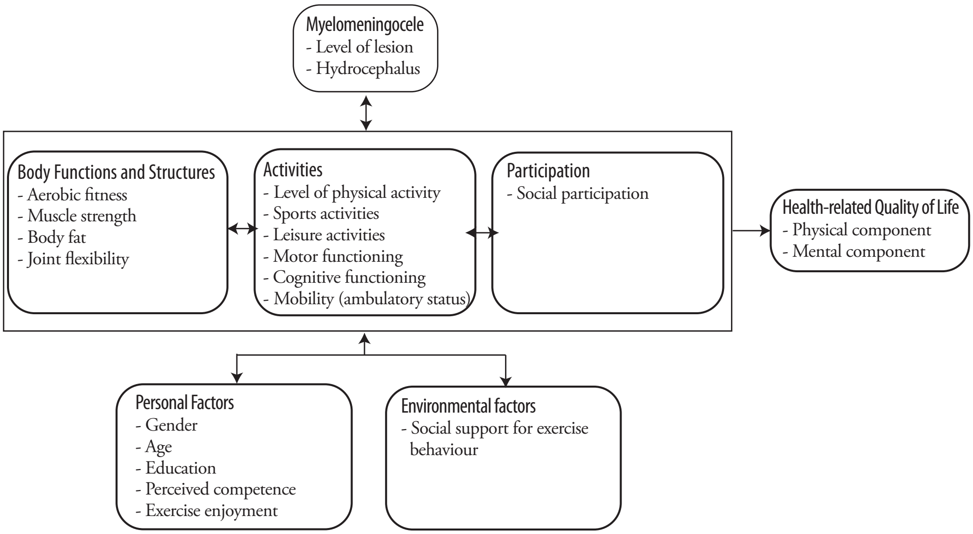
Besides physical activity, also fitness is important for health. In contrast to physical activity, which refers to a behaviour, related to the movements that people perform, fitness is a set of attributes that people have or achieve.[23] Peak oxygen uptake (peakVO_2), which is generally considered to be the gold standard for aerobic fitness, of the 14 adolescents and young adults with MMC of the previous study at our department was 20-30% lower than reference values.[24] Comparable deficits in aerobic fitness were found in children and adolescents with MMC aged 10 to 15 years.[25]

Improving levels of physical activity and fitness in adolescents and young adults with MMC is important to improve and maintain health during adulthood. During the transition from adolescence to adulthood, people start to develop their own lifestyle. This seems to be a critical period since studies in the general population showed declines in physical activity and increases in body fat.[26-28] Therefore it is important to encourage a healthy lifestyle at this age. To be able to give advices, and to develop and refine interventions to improve physical activity behaviour, levels of physical activity and fitness need to be studied, as well as associated factors and health benefits of physical activity and fitness. Sample sizes of previous studies on physical activity and fitness were too small to obtain insight in the extent of the problem of physical inactivity and low fitness, and limited the insight in associated factors. This emphasized the necessity to perform a larger study on this topic.

International Classification of Functioning, Disability and Health

In Rehabilitation, the International Classification of Functioning, Disability and Health (ICF) is a useful tool to describe an individual's functioning. The ICF is developed by the World Health Organization (WHO) to provide a unified language and framework for the description of health and health status.[29] For a specific health condition, in this case MMC, it describes functioning at three levels: body function and structure, activity and participation. Body functions are the physiological functions of the body system, which include both physical and psychological functions. Because aerobic fitness is a measure of cardiorespiratory function, it can be assigned to this level. Activity refers to an individual's execution of a task or action, and therefore this level includes daily physical activity behaviour. Participation refers to the person-in-society and includes access to and use of community resources, which can facilitate physical activity,[30] or vice versa: people with higher levels of aerobic fitness or who are more physically active may participate better in society. Environmental and personal factors interact with the above-mentioned levels of functioning. Furthermore, MMC and its associated impairments, activity limitations and participation restrictions may have consequences for health-related quality of life. An overview of the ICF model including the parameters measured in the study is presented in Figure 1.

Figure 1 ICF model including the parameters assessed in this thesis.



Aims and contents of the thesis

This study aims to obtain objective insight in the levels of daily physical activity and fitness of adolescents and young adults with MMC and to study the associations with personal and environmental factors. Furthermore we studied whether levels of daily physical activity and fitness were associated with participation and health-related quality of life. By studying the associations with cardiovascular risk factors, we aimed to obtain insight in whether interventions aiming to increase physical activity and fitness may have the potential to prevent secondary diseases, such as cardiovascular diseases later in life.

Chapter 2 describes the levels of daily physical activity, aerobic fitness and body fat of adolescents and young adults with MMC, and studies associations with gender and ambulatory status. By studying inter-relationships, this chapter aims to obtain insight in whether the negative spiral of inactivity, leading to reduced fitness and increased body fat resulting in further inactivity, could be present in people with MMC.

Chapter 3 provides a more detailed description of health-related physical fitness addressing the components of health-related physical fitness as defined by Caspersen and co-workers,[23] i.e. aerobic fitness, muscle strength, joint flexibility and body composition. By assessing the relations between aerobic capacity and the other components of health-related physical fitness, we investigated whether focusing on peripheral factors such as muscle strength would have additional value for improving aerobic capacity.

Chapter 4 addresses the question whether energy cost and physical strain of daily activities is higher in adolescents and young adults with MMC than in able-bodied persons of the same age. Higher energy cost and strain of daily activities leads to increased fatigue and discomfort and consequently may lead to an inactive lifestyle. We present results of energy cost and physical strain of daily activities measured during a standardized activity protocol consisting of locomotion and household-related activities.

Chapter 5 focuses on sports participation in adolescents and young adults with MMC. We investigated how many persons participated in sports and we studied the association between sports participation and condition-related, personal and environmental factors. Furthermore we studied the role of sports participation in total physical activity behaviour and its association with fitness.

Chapter 6 describes participation restrictions and health-related quality of life of adolescents and young adults with MMC. In addition, by assessing associations with physical activity, aerobic fitness and body fat, we explored whether lifestyle changes may be beneficial for participation and health-related quality of life.

Chapter 7 explores cardiovascular risk factors. We studied whether adolescents and young adults with MMC were at increased risk of developing cardiovascular diseases later in life, and the associations with physical activity, aerobic fitness and body fat.

Chapter 8 describes correlates of physical activity behaviour in people with physical disabilities, based on a literature review. Furthermore it presents results of focus groups on perceived barriers and facilitators of physical activity of adolescents and young adults with childhood-onset physical disabilities.

Chapter 9 presents the design of the intervention 'Active Lifestyle and Sports Participation'. This intervention is developed to increase levels of physical activity and fitness of adolescents and young adults with childhood-onset physical disabilities, such as MMC. At present, this intervention is offered at the outpatient Rehabilitation department for young adults at Erasmus MC University Medical Centre and Rijndam rehabilitation centre in Rotterdam.

Chapter 10 describes the main findings of the thesis; it discusses the strengths and limitations of this study, as well as clinical implications and directions for future research.

References

1. Sandler A. Living with spina bifida: a guide for families and professionals. Chapel Hill: The University of North Carolina Press; 1997.
2. Sadler TW. Mechanisms of neural tube closure and defects. Ment Retard Dev Disabil Res Rev 1998; 4: 247-253.
3. Botto LD, Moore CA, Khoury MJ, Erickson JD. Neural-tube defects. N Engl J Med 1999; 341:1509-1519.
4. Lemire RJ. Neural tube defects. Jama 1988; 259: 558-562.
5. Anthony S, Dorrepaal CA, Zijlstra AG, de Walle HEK, Verheij JBGM, den Ouden AL. Aangeboren afwijkingen in Nederland: Gebaseerd op de landelijke verloskunde en neonatologie registratie Leiden: TNO-PG/JGD, 2001a; 2001.063; 2001.
6. Bowman RM, McLone DG, Grant JA, Tomita T, Ito JA. Spina bifida outcome: a 25-year prospective. Pediatr Neurosurg 2001; 34:114-120.
7. Oakeshott P, Hunt GM. Long-term outcome in open spina bifida. Br J Gen Pract 2003; 53: 632-636.
8. Verhoef M. Spina Bifida: implications for functioning and health in young adults. Utrecht: University Medical Centre Utrecht; 2005. PhD thesis; ISBN 9039340455.
9. 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: 420-427.
10. Rimmer JH. Health promotion for people with disabilities: the emerging paradigm shift from disability prevention to prevention of secondary conditions. Phys Ther 1999; 79: 495-502.
11. United States Departement of Health and Human Services. Healthy People 2010: Understanding and improving health. 2nd ed. Washington DC: US Government Printing Office, Nov 2000. Available from: http://www.health.gov/healthypeople.
12. US Department of Health and Human Services, 1996. Physical activity and health: a report of the Surgeon General. US Department of Health and Human Services, Centers of Disease Control and Prevention, National Center for Chronic Disease Prevention and Health Promotion. Atlanta, GA.

13. Haskell WL, Lee IM, Pate RR, Powell KE, Blair SN, Franklin BA, et al. Physical Activity and Public Health: Updated Recommendation for Adults from the American College of Sports Medicine and the American Heart Association. Med Sci Sports Exerc 2007; 39:1423-1434.
14. Kostalova B, Frenken F, Storm I. Norm gezond bewegen 2001-2004. In: Volksgezondheid Toekomst Verkenning, Nationale Atlas Volksgezondheid. Bilthoven: RIVM, <http://www.zorgatlas.nl> Beïnvloedende factoren\ Leefstijl\ Bewegen en sporten, 5 oktober 2005.
15. Kemper HGC, Ooijendijk WTM, Stiggelbout M. Consensus over de Nederlandse Norm voor Gezond Bewegen. Tijdschr Soc Gezondheidsz 2000; 78: 180-183.
16. Heath GW, Fentem PH. Physical activity among persons with disabilities--a public health perspective. Exerc Sport Sci Rev 1997; 25: 195-234.
17. Durstine JL, Painter P, Franklin BA, Morgan D, Pitetti KH, Roberts SO. Physical activity for the chronically ill and disabled. Sports Med 2000; 30:207-219.
18. Hoffman MD. Cardiorespiratory fitness and training in quadriplegics and paraplegics. Sports Med 1986; 3: 312-330.
19. 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: 349-356.
20. 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: 550-557.
21. Van den Berg-Emons HJ, Bussmann JB, Brobbel AS, Roebroeck ME, van Meeteren J, Stam HJ. Everyday physical activity in adolescents and young adults with meningomyelocele as measured with a novel activity monitor. J Pediatr 2001; 139: 880-886.
22. Bandini LG, Schoeller DA, Fukagawa NK, Wykes LJ, Dietz WH. Body composition and energy expenditure in adolescents with cerebral palsy or myelodysplasia. Pediatr Res 1991; 29: 70-77.
23. Caspersen CJ, Powell KE, Christenson GM. Physical activity, exercise, and physical fitness: definitions and distinctions for health-related research. Public Health Rep 1985; 100: 126-131.
24. Van den Berg-Emons HJ, Bussmann JB, Meyerink HJ, Roebroeck ME, Stam HJ. Body fat, fitness and level of everyday physical activity in adolescents and young adults with meningomyelocele. J Rehabil Med 2003; 35): 271-275.
25. Agre JC, Findley TW, McNally MC, Habeck R, Leon AS, Stradel L, et al. Physical activity capacity in children with myelomeningocele. Arch Phys Med Rehabil 1987; 68: 372-377.
26. Van Mechelen W, Twisk JW, Post GB, Snel J, Kemper HC. Physical activity of young people: the Amsterdam Longitudinal Growth and Health Study. Med Sci Sports Exerc 2000; 32: 1610-1616.
27. Telama R, Yang X. Decline of physical activity from youth to young adulthood in Finland. Med Sci Sports Exerc 2000; 32: 1617-1622.
28. Parsons TJ, Manor O, Power C. Physical activity and change in body mass index from adolescence to mid-adulthood in the 1958 British cohort. Int J Epidemiol 2006; 35: 197-204.
29. WHO. International Classification of Functioning, Disability and Health (ICF). Geneva: World Health Organization; 2001.
30. Rimmer JH. Use of the ICF in identifying factors that impact participation in physical activity rehabilitation among people with disabilities. Disabil Rehabil 2006; 28: 1087-1095.

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Chapter 2

Triad of Physical Activity, Aerobic Fitness and Obesity in Adolescents and Young Adults with Myelomeningocele

J Rehabil Med 2008; 40: 70-75

Abstract

Objective: Comprehensively and objectively assess physical activity, aerobic fitness and body fat in adolescents and young adults with myelomeningocele and to investigate their relationships.
Design: Cross-sectional study.
Subjects: Fifty-one persons (26 males) with myelomeningocele aged 21.1 (standard deviation) 4.5) years.
Methods: Physical activity was measured with an accelerometry-based activity monitor. Aerobic fitness was defined as the maximum oxygen uptake during the last minute of a maximal exercise test. Body fat was assessed using sum of 4 skin-folds and body mass index. Correlations were studied using multiple regression analyses.
Results: Thirty-nine percent of the participants were inactive and another 37% were extremely inactive. Aerobic fitness was 42% lower than normative values and 35% were obese. Ambulatory status was related to daily physical activity (β= 0.541), aerobic fitness (β= 0.397) and body fat (β= –0.243). Gender was related to aerobic fitness (β= 0.529) and body fat (β= 0.610). Physical activity was related to aerobic fitness in non-ambulatory persons with myelomeningocele (β= 0.398), but not in ambulatory persons.
Conclusion: Adolescents and young adults with myelomeningocele were physically inactive, had poor aerobic fitness and high body fat. Differences exist between subgroups regarding gender and ambulatory status.

Introduction

In children and adolescents in Western countries the prevalence of inactive lifestyles and obesity has increased dramatically in the past decade.[1,2] Physical inactivity and obesity are considered to be independent risk factors of developing lifestyle-related diseases later in life, such as diabetes mellitus and cardiovascular disease.[3,4] In addition, poor aerobic fitness is known to be an independent risk factor for lifestyle-related diseases,[3] and evidence exists that it is the most powerful predictor of death due to cardiovascular disease.[5] Furthermore, adolescence and young adulthood seems to be a critical period, since daily physical activity tends to decline and body fat tends to increase.[6–8]

Compared with healthy peers, young people with chronic disabilities are restricted in the performance of daily activities.[9] Therefore, they are at increased risk of developing an inactive lifestyle. A negative spiral may then gradually develop: inactivity leading to a reduction in aerobic fitness and an increase in body fat, leading to further inactivity. In addition, inactivity, poor aerobic fitness and obesity may have serious consequences for quality of life.[10,11]

Myelomeningocele (MMC) is a common birth defect, with an incidence of 1:1000 live births.[12] Persons with MMC are often restricted in mobility,[13] which may lead to an inactive lifestyle. Due to increased life-expectancy of persons with MMC,[14] the importance of a healthy lifestyle has increased, in order to prevent secondary lifestyle-related diseases later in life.

Literature on daily physical activity, aerobic fitness and obesity in adolescents and young adults with MMC is scarce. Two studies with small sample sizes showed that daily physical activity was lower in adolescents and young adults with MMC than in healthy comparison persons.[15,16] Moreover, aerobic fitness of adolescents and young adults with MMC appeared to be 20–35% lower than in healthy comparison persons.[17,18] Some studies have reported high prevalence of obesity in persons with MMC,[19] up to 58%.[20] Due to inconsistent results, it is unclear whether health-related conditions are worse in non-ambulatory persons than in ambulatory persons.

In order to improve health throughout their lives, objective insight in the level of physical activity, aerobic fitness and obesity in adolescents and young adults with MMC is warranted. The present study aimed to comprehensively assess these health-related conditions in adolescents and young adults with MMC. In addition, we studied correlations between physical activity, aerobic fitness and obesity and their correlations with gender and ambulatory status.

Methods

Subjects

Recruitment

Adolescents and young adults with MMC, aged between 16 and 30 years, were recruited from 4 university hospitals in the Western part of the Netherlands (Rotterdam, Leiden, Utrecht and Amsterdam) and 5 rehabilitation centres in the adjacent region. Exclusion criteria were: inability to understand the measurements performed in this study (judged by the treating physician or a family member); complete dependence on a powered wheelchair; presence of disorders other than MMC that affect daily physical activity (e.g. rheumatoid arthritis); and contra-indication for a maximal exercise test. All participants and parents of adolescents aged less than 18 years gave written informed consent before participating in the study. The medical ethics committee of the Erasmus MC Rotterdam, and all other participating institutes approved the study.

A total of 171 people were invited by post to participate in this study, of whom 51 (30%) agreed. We received no response from 20% of those invited, possibly due to outdated addresses obtained from paediatric departments. The reasons for non-participation were known in 80% of the non-participants. The main reasons were no interest in the study, lack of time, duration and extensiveness of the measurements, or feeling uncomfortable with the 48-h measurement with the activity monitor (AM). For 90% of the non-participants information regarding age, gender, level of lesion and the presence of hydrocephalus was available (Table 1). Neurological level of lesion and the presence of hydrocephalus were collected from the medical records. We considered hydrocephalus to be present if a shunt was placed. Personal and disease-related characteristics did not differ between participants and non-participants, as measured with an independent *t*-test or χ^2 test. The study sample was therefore assumed to be representative.

Characteristics

In total, 26 males and 25 females (mean age 21.1 years; standard deviation (SD) 4.5) participated in the study. Ambulatory status was determined according to the classification of Hoffer et al.[21] Fifteen persons were community ambulators walking indoors and outdoors, 8 were household ambulators walking only indoors, and 28 were non (functional) ambulators. Non-functional ambulators walk only during therapy sessions, and non-ambulators are wheelchair-bound. Since we expected no difference in daily physical activity between non-functional ambulators and non-ambulators we considered them as a single group.

Table 1 Characteristics of participants and non-participants.

	Participants (n= 51)	Non-participants (n= 108)	p-value
Age (mean (SD) years)	21.1 (4.5)	21.6 (4.7)	0.66
Gender (% male)	51	45	0.62
Level of lesion (%)			0.14
Thoracic	2	9	
Thoracolumbar	14	7	
Lumbar	29	35	
Lumbosacral	41	42	
Sacral	14	7	
Hydrocephalus (%)	82	79	0.90

SD: standard deviation.

Measurements

Daily physical activity

Daily physical activity was determined using an AM (Temec Instruments, Kerkrade, The Netherlands). The AM is based on long-term ambulatory monitoring of signals from body-fixed accelerometers during daily life, aiming at the assessment of mobility-related activities.[22] Information can be obtained on which postures (lying, sitting, standing), movements (walking, cycling, wheelchair-driving, general non-cyclic movement) and transitions between postures are performed, when, how often and for how long. Validity studies in the able-bodied population and several patient groups, including patients who are wheelchair dependent, have shown that the AM is a valid and reliable instrument to quantify mobility-related activities.[22,23] Furthermore, the AM can detect differences in the level of daily physical activity between groups, which supports its validity and applicability in clinical research.[16,22]

Participants were fitted with the AM at home during 2 randomly selected consecutive weekdays. They were instructed to perform their usual daily activities, but were not allowed to swim or take a shower or bath during activity monitoring. To avoid measurement bias, the principles of the AM were explained to the participants after the measurements. All participants agreed with this procedure.

Four uniaxial piezo-resistive accelerometers were attached to the sternum and thighs. In participants who used a wheelchair, an additional accelerometer was attached to both wrists. The accelerometers were connected to the AM, which was worn in a padded bag around the waist. Accelerometer signals were stored digitally on a PCMCIA flash card with a sampling frequency of 32 Hz. After the measurements, data were downloaded onto a computer for analysis by the Signal Processing and Inferencing Language.[24] A detailed description of the configuration of the sensors and the activity detection can be found elsewhere.[22,23]

The level of daily physical activity was defined as the duration of dynamic activities (composite measure of walking including walking stairs and running, cycling, general movement and wheelchair-driving), expressed in min/day. Since no differences existed between the first and second measurement day, average results were used for further analysis.

Levels of daily physical activity of persons with MMC were compared with a reference sample consisting of 21 males and 21 females without known impairments of similar ages (plus or minus 2 years), as measured with the AM at our department in other studies using the same protocol (mean= 163 min/day). We classified 1 SD and 2 SD below the reference mean as inactive (51–107 min/day) and extremely inactive (< 51 min/day), respectively.

Aerobic fitness

Aerobic fitness was measured in a progressive maximal aerobic test, based on the McMaster All-Out Progressive Continuous Cycling and Arm test,[25] on an electronically braked arm or cycle ergometer (Jaeger ER800SH and ER800 respectively; Jaeger Toennies, Breda, The Netherlands). Studying patients with cerebral palsy who were partly wheelchair-dependent, Bhambani et al.[26] concluded that maximal exercise testing during the main mode of ambulation elicits the highest oxygen uptake. Therefore, corresponding to their main mode of ambulation, participants performed an arm crank test (n= 33) while sitting in their own immobilized wheelchair with cranks at shoulder height, or a leg cycle exercise test (n= 17). One household ambulator was unable to visit the hospital to perform the test.

The test was preceded by a 3-min warm-up (5 Watt for arm ergometry and 20 Watt for cycle ergometry), followed by a resting period of 5 min. During the test, resistance was increased every 2 min with a variable load, ensuring that total individual exercise duration ranged from 8 to 12 min. The pedal/crank rate was 60 rpm and strong verbal encouragement was given throughout the test. The test was terminated when the participant stopped due to exhaustion or when the participant was unable to maintain the initial pedal/crank rate. Gas exchange was determined continuously using a breath-by-breath portable measurement system ($K4b^2$, COSMED, Rome, Italy). Calibration with reference gas was performed before each test. Aerobic fitness was defined as the mean oxygen uptake during the last minute of exercise ($peakVO_2$). Values of aerobic fitness were compared with normative values of Dutch sedentary males and females of similar ages.[27]

Body composition and body fat

Height was measured with a flexible tape while participants were lying on a bed. In case of contractures, measurements were performed from joint to joint. Body mass of ambulatory persons was obtained while standing on a Seca scale and for non-ambulatory persons while sitting on an electronic scale (Cormier, France).

Thickness of 4 skin-folds (biceps, triceps, subscapular, supra-iliac) was measured twice on the right side of the body with a Harpenden calliper (Harpenden, Burgess Hill, UK) and mean values were used for analyses. Because the skin-fold thickness of 2 non-ambulators was beyond that of the calliper jaws (80 mm), we reported these data as missing.

In addition, body mass index (BMI) was calculated from height and body mass by dividing the body mass by the length (in metres) squared. BMI ≥ 30 was classified as obese.

Statistical analysis

Results for daily physical activity, aerobic fitness and body fat are presented as mean and SD for the total study sample and for subgroups regarding ambulatory status. Multiple regression analyses were performed to study relations between daily physical activity, aerobic fitness and body fat. Independent variables (duration of dynamic activities (min/day), peakVO_2 (l/min), sum of 4 skin-folds (mm)) were entered stepwise in the multiple regression analyses. In case of differences between subgroups we included gender (0= male; 1= female) and ambulatory status (1= non(functional) ambulator; 2= household ambulator; 3= community ambulator) in the regression analyses as independent factors. In addition, we fitted specific regression models for ambulatory (including community and household ambulators) and non-ambulatory persons with MMC. Probability to enter was set at $p \leq 0.05$, and the probability to remove was set at $p \geq 0.10$. The standardized regression coefficients (β) and explained variance (R^2) of the regression models are presented. Statistical analyses were performed using SPSS 11.0 for Windows.

Results

Mean (SD) values of daily physical activity, aerobic fitness and body fat for the total study sample and for subgroups regarding ambulatory status are presented in Table 2.

During 24 h, adolescents and young adults with MMC spent 81 (SD 62) min on physical activities, which was less than the reference sample from previous studies at our department (163 (SD 56) min, $p < 0.001$). Thirty-nine percent of the participants were classified as inactive and another 37% as extremely inactive. Average aerobic fitness of adolescents and young adults with MMC was 42% (range 13–77%) lower than the Dutch normative values (1.48 (SD 0.52) l/min vs 2.56 (SD 0.41) l/min, respectively). Obesity (BMI ≥ 30) was found in 35% of the participants; 19% of the males were obese and 52% of the females with MMC.

Correlations between daily physical activity, aerobic fitness and body fat

Regression models of the total group showed that 29–50% of the variance in daily physical activity, aerobic fitness and body fat is determined by gender and ambulatory status (Table 3). Persons with a higher level of ambulatory status (community ambulators vs household ambulators vs non(functional) ambulators) were more physically active during the day (β= 0.541; $p < 0.001$), had higher aerobic fitness (β= 0.397; $p < 0.001$) and less body fat (β= –0.243; $p = 0.03$) compared with less ambulatory persons. Males had higher values of aerobic fitness (β= 0.529; $p < 0.001$) and less body fat (β= 0.610; $p < 0.001$) compared with females. Gender was not related to daily physical activity.

In some aspects, regression models for aerobic fitness differed between ambulatory and non-ambulatory persons with MMC (Table 4). Aerobic fitness was related to physical activity in non-ambulatory persons (β= 0.398; $p = 0.04$), but not in ambulatory persons with MMC. Body fat was not related to physical activity, or to aerobic fitness in both ambulatory and non-ambulatory persons with MMC.

Table 2 Daily physical activity, aerobic fitness and body fat of the total group and subgroups regarding ambulatory status.

	Total group	**Community ambulator**	**Household ambulator**	**Non ambulator**
	(n= 51)	(n= 15)	(n= 8)	(n= 28)
Height (cm)	157.7 (38.5)	165 (7.3)	164.1 (12.5)	151.6 (11.4)
Weight (kg)	67.5 (15.3)	63.9 (10.8)	65.1 (14.3)	70.1 (17.5)
Daily physical activity (min/day)	81 (62)[b]	127 (80)	93 (52)[a]	52 (32)[a]
Walking (min/day)		75 (44)	30 (31)	-
Wheelchair-driving (min/day)		-	33 (30)	34 (21)
Cycling (min/day)		13 (17)	-	-
Aerobic fitness (l/min)	1.48 (0.52)	1.85 (0.57)	1.44 (0.45)	1.29 (0.40)
Aerobic fitness (ml/kg/min)	22.6 (8.2)[a]	29.0 (7.7)	22.3 (6.6)[a]	19.2 (6.8)
Sum of 4 skin-folds (mm)	74.4 (38.5)[b]	59.1 (29.2)	65.5 (32.3)	86.0 (42.0)[b]
BMI (kg/m^2)	27.5 (6.6)	23.3 (3.6)	24.2 (5.2)	30.4 (6.7)

[a]$n - 1$. [b]$n - 2$.

Table 3 Regression models for daily physical activity, aerobic fitness and body fat for total group (n= 50).

Dependent variable	**Independent variable**	β	***p*-value**	R^2
Daily physical activity (min/day)	Ambulatory status	-0.541	< 0.001	0.29
Aerobic fitness (peakVO_2 in l/min)	Gender	-0.529	< 0.001	0.50
	Ambulatory status	-0.397	< 0.001	
Body fat (sum of 4 skin-folds in mm)	Gender	0.610	< 0.001	0.47
	Ambulatory status	0.243	0.03	

β= regression coefficient. R^2=explained variance.

Table 4 Regression models for daily physical activity, aerobic fitness and body fat for ambulatory (15 community and 7 household ambulators) and non-ambulatory persons (*n*= 28).

	Ambulatory (*n*= 22)				Non-ambulatory (*n*= 28)			
Dependent variable	**Independent variable**	β	***p*-value**	R^2	**Independent variable**	β	***p*-value**	R^2
Daily physical activity (min/day)	-				PeakVO_2 (l/min)	0.398	0.04	0.16
Aerobic fitness (peakVO_2 in l/min)	Gender	-0.673	<0.001	0.45	Gender	-0.512	0.003	0.42
					Activity	0.365	0.03	
Body fat (sum of 4 skin-folds in mm)	Gender	0.513	0.02	0.26	Gender	0.725	< 0.001	0.53

β= regression coefficient. R^2= explained variance.

Discussion

The present study showed that daily physical activity and aerobic fitness of adolescents and young adults with MMC were 50% and 42% lower, respectively, compared with reference values and 35% of them were obese. Non-ambulatory persons spent less time on physical activity during the day than ambulatory persons and they had lower levels of aerobic fitness and more body fat. Therefore, compared with the general population, adolescents and young adults with MMC, and particularly non-ambulatory persons, might be at increased risk of developing lifestyle-related diseases later in life. Physical activity of non-ambulators with MMC was comparable to that of persons with spinal cord injury measured one year after discharge from the rehabilitation centre (62 min/day; unpublished results, Van den Berg-Emons). Aerobic fitness was low compared with other patient groups. Maximal oxygen uptake in young men (aged 19–23 years) with spastic diplegia assessed on a cycle ergometer was 22% higher (2.83 l/min)[28] than in the ambulatory men who participated in the present study (2.03 l/min). Average peakVO_2 in non-ambulatory males with MMC (1.57 l/min) during arm exercise was 21% lower than the average peakVO_2 of males with paraplegic spinal cord in¬juries with lesions below T10 (1.98 l/min).[29] The low level of aerobic fitness must, however, be interpreted with caution because peakVO_2 is influenced by the quantity of active muscle mass,[30–32] which may be reduced in ambulatory persons with MMC due to paresis of lower extremity muscles.

By studying these correlations, this study aimed at obtaining more insight in the negative spiral of inactivity leading to reduced aerobic fitness and increased body fat, resulting in further inactivity. However, no correlations were found between daily physical activity, aerobic fitness and body fat. Van den Berg-Emons et al.[18] found a correlation between daily physical activity and aerobic fitness in a small sample of adolescents and young adults with MMC. In the present study, daily physical activity was related only to ambulatory status. Therefore, it seemed worthwhile separately to investigate potential determinants of physical activity for ambulatory and non-ambulatory subgroups. Results of the present study could confirm the correlation between physical activity and aerobic fitness in non-ambulatory persons, but not in ambulatory. In ambulatory persons, physical fitness and body fat were not related to physical activity, which suggests that other factors play a role in the level of daily physical activity. Studies on personal and environmental barriers and facilitators for physical activity in adolescents and young adults with MMC are warranted.

In non-ambulatory persons, daily physical activity was related to aerobic fitness, which may suggest that a minimum level of aerobic fitness might be needed to be able to be physically active during the day. However, because of the cross-sectional study design, no causal relations can be established and the question remains whether adolescents and young adults with MMC are inactive because they are unfit or whether they are unfit because they are physically inactive. In the general population of this age, daily physical activity and aerobic fitness are poor to moderately related,[33,34] indicating that physical activity accounts for a small part of the variability in aerobic fitness. Although for most individuals increases in physical activity might produce increases in aerobic fitness, the amount of adaptations in fitness to a standard exercise dose may vary widely.[35] Insight into physical strain during daily activities might provide further insight into the correlation between physical activity and fitness.

In both ambulatory and non-ambulatory persons with MMC, body fat was not related to daily physical activity or aerobic fitness. This may suggest that other strategies, such as reducing energy intake, may be more important in preventing obesity than increasing daily physical activity or fitness. In addition, it has been suggested that excessive body fat, particularly in females, may originate directly from the disorder itself[20] and future studies including hormonal analysis of metabolism and medication are warranted in order to identify the mechanism for their obesity.

The strength of the present study is that daily physical activity, aerobic fitness and body fat were measured objectively in one sample, allowing us to study their correlations. Assessing daily physical activity using an objective AM is preferable to subjective questionnaires because the latter is prone to overestimating daily physical activity. However, because we used the AM, we were restricted to 2 days of monitoring. It has been suggested that at least 7 days of activity monitoring might be needed to characterize an individual's habitual activity pattern.[36] Because of the 2-day measurements of physical activity we might have underestimated the correlation between daily physical activity and aerobic fitness, but this did not hamper the comparison with reference values, which were based on the same method.[16]

Indicating body fat by thickness of 4 skin-folds or BMI might have some disadvantages. With skin-fold measurements, difficulties may arise when thickness of skin-folds is beyond that of the calliper jaws.[19] Nevertheless, the skin-fold technique is considered adequate for estimating body fat in patients with MMC based on 2–3% differences between this technique and weighing under water.[20] Because we determined the relationships between the health-related components using sum of 4 skin-folds, we consider the results valid. It has been suggested that BMI may overestimate body fat in persons with MMC because height is underestimated due to paralytic deformities.[19] Although we tried to minimize this problem by measuring height from joint to joint in case of contractures, large differences in BMI were found between subgroups regarding ambulatory status, which might relate to the height differences between the subgroups.

Aerobic fitness of non-ambulators was measured during arm ergometry, which may have led to an underestimation due to smaller active muscle mass compared with ambulators who were measured during cycle ergometry. Arm exercise in ambulatory persons typically induces a peakVO_2 of 70% of that achieved with lower extremity exercise.[37] If that 70% ratio is also applicable to non-ambulatory persons (who have considerable use of their arm and shoulder muscles) the non-ambulatory subjects may in fact have an average peakVO_2 of 1.84 l/min, which is comparable to that of ambulators. However, assuming that the main mode of ambulation elicits the highest oxygen uptake,[26] we selected the best available measure for each participant. Therefore, we consider the aerobic fitness results to be valid.

Although the response rate of the study was low, we assumed the study sample to be representative because participants and non-participants did not differ regarding personal and disease-related characteristics. Moreover, with a high prevalence of middle-level (lumbosacral) and high-level (lumbar or thoraco-lumbar) lesions, the present study sample was comparable to persons from a national study on adolescents with spina bifida in the Netherlands (ASPINE).[13] However, it is known that many persons with MMC lack initiative,[38] which may have influenced their willingness to participate in the present study, contributing to the low response rate. If this bias was present in this study, the current results would have overestimated the level of physical activity in the MMC population.

In conclusion, the present study demonstrated that adolescents and young adults with MMC have low levels of daily physical activity, poor aerobic fitness and excessive body fat. Since these factors are all independent risk factors for developing cardiovascular diseases later in life, the results emphasize the importance of improving these health-related conditions in order to maintain a healthy adult life. Because we did not find correlations between all health-related conditions, we suggest focusing on all 3 aspects simultaneously in future interventions. Future studies should focus on modifiable determinants of daily physical activity, aerobic fitness and body fat. To develop adequate intervention strategies to improve health in persons with MMC, differences between ambulatory and non-ambulatory persons should be taken into account.

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References

1. Ogden CL, Carroll MD, Curtin LR, McDowell MA, Tabak CJ, Flegal KM. Prevalence of overweight and obesity in the United States, 1999–2004. J Am Med Acad 2006; 295: 1549–1555.
2. Caspersen CJ, Pereira MA, Curran KM. Changes in physical activity patterns in the United States, by sex and cross-sectional age. Med Sci Sports Exerc 2000; 32: 1601– 1609.
3. Pate RR, Pratt M, Blair SN, Haskell WL, Macera CA, Bouchard C, et al. Physical activity and public health. A recommendation from the Centers for Disease Control and Prevention and the American College of Sports Medicine. J Am Med Acad 1995; 273: 402–407.
4. Powell KE, Thompson PD, Caspersen CJ, Kendrick JS. Physical activity and the incidence of coronary heart disease. Annu Rev Public Health 1987; 8: 253–287.
5. Myers J, Prakash M, Froelicher V, Do D, Partington S, Atwood JE. Exercise capacity and mortality among men referred for exercise testing. N Engl J Med 2002; 346: 793–801.
6. Van Mechelen W, Twisk JW, Post GB, Snel J, Kemper HC. Physical activity of young people: the Amsterdam Longitudinal Growth and Health Study. Med Sci Sports Exerc 2000; 32: 1610–1616.
7. Telama R, Yang X. Decline of physical activity from youth to young adulthood in Finland. Med Sci Sports Exerc 2000; 32: 1617–1622.
8. Parsons TJ, Manor O, Power C. Physical activity and change in body mass index from adolescence to mid-adulthood in the 1958 British cohort. Int J Epidemiol 2006; 35: 197–204.
9. Newacheck PW, Halfon N. Prevalence and impact of disabling chronic conditions in childhood. Am J Public Health 1998; 88: 610–617.
10. Stewart AL, Hays RD, Wells KB, Rogers WH, Spritzer KL, Greenfield S. Long-term functioning and well-being outcomes associated with physical activity and exercise in patients with chronic conditions in the Medical Outcomes Study. J Clin Epidemiol 1994; 47: 719–730.
11. Schwimmer JB, Burwinkle TM, Varni JW. Health-related quality of life of severely obese children and adolescents. J Am Med Acad 2003; 289: 1813–1819.
12. Botto LD, Moore CA, Khoury MJ, Erickson JD. Neural-tube defects. N Engl J Med 1999; 341: 1509–1519.
13. 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: 420–427.
14. Bowman RM, McLone DG, Grant JA, Tomita T, Ito JA. Spina bifida outcome: a 25-year prospective. Pediatr Neurosurg 2001; 34: 114–120.
15. Bandini LG, Schoeller DA, Fukagawa NK, Wykes LJ, Dietz WH. Body composition and energy expenditure in adolescents with cerebral palsy or myelodysplasia. Pediatr Res 1991; 29: 70–77.
16. Van den Berg-Emons HJ, Bussmann JB, Brobbel AS, Roebroeck ME, van Meeteren J, Stam HJ. Everyday physical activity in adolescents and young adults with meningomyelocele as measured with a novel activity monitor. J Pediatr 2001; 139: 880–886.
17. Agre JC, Findley TW, McNally MC, Habeck R, Leon AS, Stradel L, Birkebak R, Schmalz R. Physical activity capacity in children with myelomeningocele. Arch Phys Med Rehabil 1987; 68: 372–377.

18. Van den Berg-Emons HJ, Bussmann JB, Meyerink HJ, Roebroeck ME, Stam HJ. Body fat, fitness and level of everyday physical activity in adolescents and young adults with meningomyelocele. J Rehabil Med 2003; 35: 271–275.
19. Roberts D, Shepherd RW, Shepherd K. Anthropometry and obesity in myelomeningocele. J Paediatr Child Health 1991; 27: 83–90.
20. 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: 305–311.
21. 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.
22. 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: 349–356.
23. 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: 550–557.
24. Jain A, Martens WLJ, Mutz G, Weiss RK, Stephan E. Towards a comprehensive technology for recording and analysing of multiple physiological parameters within their behavioral and environmental context. In: Fahrenberg J, Myrtek M, editors. Ambulatory assessment: computer-assisted psychological and psychophysiological methods in monitoring and field studies. Seattle: Hogrefe & Huber Publishers; 1996, p. 215–236.
25. Bar-Or O, editor. Pediatric sports medicine for the practitioner. From physiologic principles to clinical applications. New York: Springer Verlag; 1983: p. 315–341.
26. Bhambhani YN, Holland LJ, Steadward RD. Maximal aerobic power in cerebral palsied wheelchair athletes: validity and reliability. Arch Phys Med Rehabil 1992; 73: 246–252.
27. Vos JA. Ergometrie en Trainingsbegeleiding Nederlands Paramedisch Instituut. [Ergometry and training supervision.] Amersfoort; 2001, p. 112–121 (in Dutch).
28. Lundberg A. Maximal aerobic capacity of young people with spastic cerebral palsy. Dev Med Child Neurol 1978; 20: 205–210.
29. Janssen TW, Dallmeijer AJ, Veeger DJ, van der Woude LH. Normative values and determinants of physical capacity in individuals with spinal cord injury. J Rehabil Res Dev 2002; 39: 29–39.
30. Davies CT, Sargeant AJ. Effects of training on the physiological responses to one- and two-leg work. J Appl Physiol 1975; 38: 375–377.
31. Davies CT, Sargeant AJ. Indirect determination of maximal aerobic power output during work with one or two limbs. Eur J Appl Physiol Occup Physiol 1974; 32: 207–215.
32. Lewis SF, Taylor WF, Graham RM, Pettinger WA, Schutte JE, Blomqvist CG. Cardiovascular responses to exercise as functions of absolute and relative work load. J Appl Physiol 1983; 54: 1314–1323.
33. Morrow JR, Freedson PS. Relationship between habitual physical activity and aerobic fitness in adolescents. Pediatr Exercise Sci 1994; 6: 315–329.
34. Malina RM. Physical activity and fitness: pathways from childhood to adulthood. Am J Hum Biol 2001; 13: 162–172.
35. Blair SN, Cheng Y, Holder JS. Is physical activity or physical fitness more important in defining health benefits? Med Sci Sports Exerc 2001; 33 Suppl 6: S379–S399; discussion S419–S420.

36. Trost SG, Pate RR, Freedson PS, Sallis JF, Taylor WC. Using objective physical activity measures with youth: how many days of monitoring are needed? Med Sci Sports Exerc 2000; 32: 426–431.
37. Astrand PO, Rodahl K. Evaluation of physical performance on the basis of tests. In: Astrand PO, Rodahl K, editors. Textbook of work physiology: physiological bases of exercise. Singapore: McGraw-Hill Book Co.; 1986, p. 354–390.
38. Dise JE, Lohr ME. Examination of deficits in conceptual reasoning abilities associated with spina bifida. Am J Phys Med Rehabil 1998; 77: 247–251.

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Chapter 3

Health-Related Physical Fitness of Adolescents and Young Adults with Myelomeningocele

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Abstract

To assess components of health-related physical fitness in adolescents and young adults with myelomeningocele (MMC), and to study relations between aerobic capacity and other health-related physical fitness components. This cross-sectional study included 50 adolescents and young adults with MMC, aged 16–30 years (25 males).

Aerobic capacity was quantified by measuring peak oxygen uptake (peakVO_2) during a maximal exercise test on a cycle or arm ergometer depending on the main mode of ambulation.

Muscle strength of upper and lower extremity muscles was assessed using a hand-held dynamometer. Regarding flexibility, we assessed mobility of hip, knee and ankle joints. Body composition was assessed by measuring thickness of four skin-folds. Relations were studied using linear regression analyses. Average peakVO_2 was 1.48 ± 0.52 l/min, 61% of the participants had subnormal muscle strength, 61% had mobility restrictions in at least one joint and average sum of four skin-folds was 74.8 ± 38.8 mm.

PeakVO_2 was significantly related to gender, ambulatory status and muscle strength, explaining 55% of its variance.

Adolescents and young adults with MMC have poor health-related physical fitness. Gender and ambulatory status are important determinants of peakVO_2. In addition, we found a small, but significant relationship between peakVO_2 and muscle strength.

Introduction

During the last decades, life expectancy of persons with myelomeningocele (MMC) has increased and many will nowadays survive into adulthood.[1] As a consequence, lifestyle-related diseases, such as cardiovascular disease and diabetes mellitus, will be of increasing concern in this patient group. Therefore, more attention towards a healthy lifestyle is warranted. Similar to the general population, persons with MMC develop their own lifestyle during the transition from adolescence to adulthood. At this age, special attention should be paid to optimize the lifestyle in order to improve health throughout life.

Physical fitness is recognized as an important component of health[2,3] and it may be important for the performance of functional activities and quality of life.[4,5] Low physical fitness may result in high physical strain during the performance of activities.[6] As a consequence, activity levels may decrease due to fatigue and discomfort, exacerbating low physical fitness. Caspersen and co-workers defined several health-related components of physical fitness, i.e. aerobic capacity, muscle strength and endurance, flexibility and body composition.[7]

Only a few studies are available on health-related physical fitness in persons with MMC. In a previous study in adolescents and young adults with MMC, we found that average peak oxygen uptake (peakVO_2) was 42% lower than normative values of healthy peers, with lower values in non-ambulatory than in ambulatory persons.[8] Several other studies have also reported low aerobic capacity.[9-11] Furthermore, children with MMC, and particularly non-ambulatory children, were found to have reduced strength of lower extremity muscles.[9,12,13] Hip and knee contractures have been reported in respectively 18% and 36% of adolescents and young adults with MMC and hydrocephalus.[14] Finally, high levels of body fat have been found in persons with MMC;[15-17] previously we reported that 35% of adolescents and young adults with MMC were obese.[8] Because these previous studies lack simultaneous assessment of several health-related physical fitness components, the relation between aerobic capacity and other components remains unclear. Insight in these interrelations may show for example, whether focusing on peripheral factors such as muscle strength would have additional value for improving aerobic capacity.

Due to the scarcity of studies in persons with MMC, the first aim of this study was to describe health-related physical fitness i.e. aerobic capacity, muscle strength, joint flexibility and body composition in a relatively large group of adolescents and young adults with MMC, allowing descriptions of subgroups regarding gender and ambulatory status. Secondly, we wanted to study the relation between aerobic capacity and other components of health-related physical fitness, controlled for relevant personal and disease-related characteristics. Studies in persons with other chronic conditions such as spinal cord injury have shown relations between aerobic capacity and muscle strength.[18-20] Therefore, also in persons with MMC, we expected aerobic capacity to be related to muscle strength.

Method

Participants

Recruitment

Adolescents and young adults with MMC, aged between 16 and 30 years, were recruited from the university hospitals in Rotterdam, Leiden, Utrecht and Amsterdam and all rehabilitation centers in the Southwest of the Netherlands. Exclusion criteria were complete dependence on an electric wheelchair, presence of disorders other than MMC that affect daily physical activity (e.g. rheumatoid arthritis), and presence of disorders that contra-indicate a maximal exercise test (e.g. exercise-induced ischemia or arrhythmias, uncontrolled hypertension and exercise limitation due to chronic obstructive pulmonary disease). We invited 171 persons of whom 50 participated in the study (29%). Main reasons for non-participation were no interest, lack of time or duration of the measurements. No differences were found between participants and non-participants regarding age, gender, level of lesion and presence of hydrocephalus, as measured with an independent *t*-test or Chi square test.[8] All participants and parents of adolescents aged less than 18 years gave written informed consent before participating in the study. The Medical Ethics Committee of the Erasmus MC Rotterdam and of all participating institutes approved the study.

Characteristics

In total, 25 males and 25 females (mean age 21.2 ± 4.5 years) participated in the study. Table 1 presents personal and disease-related characteristics of participants. Neurological level of lesion and the presence of hydrocephalus were obtained from the medical records. Five categories of neurological level were distinguished: thoracic, thoracolumbar, lumbar, lumbosacral and sacral. We considered hydrocephalus to be present when a shunt was placed. Ambulatory status was determined according to the classification of Hoffer and co-workers:[21] (1) community ambulator, walking indoors and outdoors, (2) household ambulator, walking only indoors, and (3) non(functional) ambulator. Non-functional ambulators walk only during therapy sessions and non-ambulators are completely wheelchair dependent. Since main mode of ambulation in daily life is similar between non-functional ambulators and non-ambulators, we combined these two groups. Educational level was categorized as low (prevocational practical education or lower level), medium (pre-vocational theoretical education and secondary education) or high (higher education and university).[22]

Table 1 Personal and disease-related characteristics of participants (*n*= 50).

Gender (% male)	50
Age (mean ± SD in years)	21.2 ± 4.5
Height (mean ± SD in m)	1.57 ± 0.12
Body mass (mean ± SD in kg)	67.6 ± 15.6
Level of lesion (%)	
Thoracic	2
Thoracolumbar	14
Lumbar	28
Lumbosacral	42
Sacral	14
Hydrocephalus (%)	82
Ambulatory status (%)[1]	
Community ambulator	30
Household ambulator	14
Non (functional) ambulator	56
Educational level (%)	
Low	37
Medium	39
High	24

[1]no gender differences as tested with a Chi square test.

Aerobic capacity

Aerobic capacity was measured in a progressive maximal exercise test, based on the McMaster All-Out Progressive Continuous Cycling and Arm test,[23] on an electronically braked arm or cycle ergometer (Jaeger ER800SH and ER800 respectively; Jaeger Toennies, Breda, The Netherlands). Studying patients with cerebral palsy who were partly wheelchair-dependent, Bhambani and coworkers concluded that maximal exercise testing during the main mode of ambulation elicits the highest oxygen uptake.[24] Therefore, depending on their main mode of ambulation, participants performed an arm crank test (*n*= 33) while sitting in their own immobilized wheelchair with cranks at shoulder height, or a leg cycle exercise test (*n*= 17). The test was preceded by a 3-minute warm-up (5 W for arm ergometry and 20 W for cycle ergometry), followed by a resting period of 5 min. During the test, resistance was increased every 2 min with a variable load, ensuring that total individual exercise duration ranged from 8 to 12 min.

The pedal/crank rate was 60 rpm and strong verbal encouragement was given throughout the test. The test was terminated when the subject stopped due to exhaustion. Gas exchange was determined continuously using a breath-by-breath portable measurement system (K4b^2, COSMED, Rome, Italy). Calibration was performed before each test with reference gases. Heart rate was measured continuously using a heart rate (HR) monitor which was attached to the system, and participants were fitted with a transmitter belt around the chest (Polar Electro, Finland). Aerobic capacity was defined as the mean oxygen uptake during the last 30 s of exercise (peakVO_2, in l/min). In addition, for those measured during cycle ergometry, values of aerobic capacity were expressed as percentage of reference values of Dutch able-bodied sedentary males and females of similar ages, as estimated from a submaximal exercise test on a cycle ergometer using the nomogram of Åstrand.[25] Peak work load (peakW) was defined as the highest work load maintained for at least 1 min.

In addition, the ventilatory anaerobic threshold (VAT) was estimated by the ventilatory equivalent method, when the ventilatory equivalent for O_2 (VE/VO_2) and the end tidal O_2 partial pressure ($PETO_2$) increased while ventilatory equivalent for CO_2 (VE/VCO_2) and end-tidal CO_2 partial pressure ($PETCO_2$) remained stable.[26,27] VAT was also expressed relative to the measured $peakVO_2$ (VAT%). HR and respiratory exchange ratio (RER) were used as objective criteria for maximal exercise. Subjective strain was measured at the end of the final stage using the modified Borg scale of rating perceived exertion (RPE), which is a vertical scale labelled from 0 (no effort at all) to 10 (maximal effort).[28,29]

Muscle strength

We measured strength of two large muscle groups of the lower and upper extremity with a hand-held dynamometer (MicroFet, Hoggan Health Industries Inc, West Jordan, UT, USA) using the "break" testing method. We measured strength of hip flexors and knee extensors in persons whose main mode of ambulation was walking. In persons whose main mode of ambulation was wheelchair-driving, we measured strength of shoulder abductors and elbow extensors. The positions and the performance of the measurements were according to Van der Ploeg and co-workers.[30] The applicator of the dynamometer was held against the distal part of the limb segment, and participants were asked to build up their force to a maximum against it. The examiner applied sufficient resistance just to overcome the force exerted by the participant, and the applicator was then immediately moved away from the limb segment and the measured force was recorded. Each trial lasted approximately 4 s, and three repetitions were performed with 1 min rest in between. We used the average value of three repetitions of the dominant side for further analyses because we found no differences between the dominant and non-dominant side (tested with a paired samples *t*-test). To assess whether muscle strength was subnormal and to be able to compare strength of upper and lower extremity muscles, values were normalized to Z-scores using reference values of healthy males and females. For hip flexors, shoulder abductors and elbow extensors we used reference values of Phillips and co-workers for males and females aged 20–29 years[31] and for knee extensors, we used reference values of Bohannon and co-workers.[32] In case the examiner could not resist the muscle strength, Z-score was set at 2. We used the lowest Z-score of the upper or lower extremity as indicator of muscle strength, and we considered muscle weakness to be present when Z-score ≤ -2.

Flexibility
As indicator of flexibility of lower extremity we assessed passive mobility of hip and knee joint while participants were lying supine and of the ankle joint while they were sitting. We considered mobility to be restricted when extension of hip and knee joint and ankle dorsal flexion did not reach neutral position. Since no differences were found in mobility restrictions between dominant and non-dominant side (tested with the Wilcoxon signed rank test) we used the results of the dominant side to calculate a sum score of joint mobility ranging from 0 (no mobility restrictions in any joint) to 3 (all three joints have mobility restrictions).

Body composition
Height was measured with a flexible tape while lying on a bed. In case of joint contractures, measurements were performed from joint to joint. Body mass of ambulatory persons was obtained while standing on a Seca scale and of non-ambulatory persons while sitting on an electronic scale (Cormier, France). Thickness of four skin-folds (biceps, triceps, subscapular, and suprailiac) was measured twice on the right side of the body with a Harpenden caliper (Burgess Hill, UK) and mean values were used for further analyses. In addition, sum of four skin-folds were expressed as percentage of reference values of Dutch sedentary males and females of similar ages.[25]

Statistical analysis
Results of health-related fitness components are presented as mean ± standard deviation (SD) for the total group and for subgroups regarding gender and ambulatory status.
Simple linear regression analyses were used to study relations between personal and disease-related characteristics and peakVO_2 (in l/min), in order to detect relevant determinants to include in the multiple regression models. Multiple linear regression analyses for peakVO_2 were carried out in two steps. First, we built a model including relevant personal and disease-related characteristics. In the second step, we studied whether other health-related physical fitness components, i.e. muscle strength (Z-score), sum of four skin-folds (mm), and joint mobility (number of restricted joint), were significantly related to peakVO_2, controlling for the personal and disease-related characteristics from step 1. We presented the standardized regression coefficients (β) and explained variance (R^2) of the linear regression models. Statistical analyses were performed using SPSS 12.0 for Windows. $P\leq 0.05$ were considered significant.

In the analyses, we pooled the data of peakVO_2 measured during arm and cycle ergometry, and corrected for differences in exercise mode. Due to high collinearity between type of ergometer and ambulatory status (correlation coeffcient (r)= 0.92, $p<$ 0.001), we adjusted the analyses for ambulatory status as proxy for type of ergometer, because clinically, ambulatory status would be more meaningful.

Furthermore, due to overlap between lesion level and ambulatory status (r= 0.58; $p<$ 0.001), we chose to only correct for ambulatory status in the multiple regression analyses. Ambulatory status was determined during the study, whereas level of lesion was obtained from medical records. The national study on adolescents with spina bifida in the Netherlands (ASPINE) reported that the level of lesion as mentioned in medical records may be unreliable because lesions are determined at different ages, sometimes using the motor level and sometimes using the sensory level, and often lacking descriptions of methods.[14]

Results

Descriptive results of aerobic capacity, muscle strength, joint mobility and body composition for the total group and for subgroups regarding gender and ambulatory status are presented in Table 2. Average peakVO_2 was 1.48 ± 0.52 l/min. For persons measured during cycle ergometry (n= 17), peakVO_2 corresponded to 67 ± 15% of reference values. Sixty-one percent of the participants had subnormal muscle strength as indicated by Z-scores and 61% had mobility restrictions in one or more joints. Average sum of four skin-folds was 74.8 ± 38.8 mm, corresponding to 159 ± 77% of normative values (Table 2). According to objective and subjective criteria most participants reached their peak exercise performance. Average peakHR was 174 ± 19 beats per minute, which was 90.4 ± 9.6% of the age predicted maximum (220—age for cycle ergometry; 210—age for arm ergometry) and average peakRER was 1.17 ± 0.22. Average RPE was 6.2 ± 2.2, indicating that participants experienced the exercise as heavy to very heavy.

Several personal and disease-related characteristics were related to peakVO_2 (Table 3). PeakVO_2 was higher in males than in females (β= -0.61; p< 0.001), higher in community ambulatory persons than in household and nonambulatory persons (β= -0.48; p< 0.001) and higher in persons with a lower level of lesion (β= -0.43; p= 0.002). Age, presence of hydrocephalus and educational level were not related to peakVO_2.

Fifty percent of the variance in peakVO_2 was explained by gender and ambulatory status (Table 3). In addition, when controlling for both variables, we found that participants with higher muscle strength had higher values of peakVO_2 (β= 0.22; p= 0.04) explaining an additional 5% of the variance in peakVO_2. Furthermore, we found that participants with higher sum of four skin-folds tended to have higher values of peakVO_2 (β= 0.25; p= 0.08).

Table 2 Descriptive results of health-related physical fitness components for the total group and for subgroups regarding gender and ambulatory status.

		Gender		Ambulatory status		
	All	Male	Female	Community	Household	Non(functional)
	(*n*= 50)	(*n*= 25)	(*n*= 25)	(*n*= 15)	(*n*= 7)	(*n*= 28)
Aerobic capacity (mean ± SD)						
PeakVO$_2$ (l/min)	1.48 ± 0.52	1.78 ± 0.51	1.18 ± 0.30	1.85 ± 0.57	1.44 ± 0.45	1.29 ± 0.40
% of reference values*	67 ± 15	71 ± 13	61 ± 18	68 ± 16	54 ± 2	-
PeakVO$_2$ (ml/[kg.min])	22.6 ± 8.2	28.1 ± 7.0	17.0 ± 4.7	29.0 ± 7.7	22.3 ± 6.6	19.2 ± 6.8
Peak oxygen pulse (ml/bpm)	8.7 ± 3.0	10.1 ± 2.8	7.3 ± 2.4	10.7 ± 2.8	7.8 ± 2.4	7.8 ± 2.8
PeakRER	1.17 ± 0.22	1.17 ± 2.28	1.18 ± 0.20	1.15 ± 0.22	1.27 ± 0.16	1.16 ± 0.24
PeakW (watts)	91 ± 42	113 ± 43	69 ± 28	123 ± 42	97 ± 35	73 ± 34
PeakHR (bpm)	174 ± 19	179 ± 16	169 ± 20	173 ± 21	183 ± 14	172 ± 18
PeakHR % of predicted maximum	90 ± 10	92 ± 8	89 ± 10	87 ± 10	95 ± 8	91 ± 10
VAT (l/min)	1.20 ± 0.43	1.39 ± 0.44	1.01 ± 0.32	1.55 ± 0.45	1.07 ± 0.29	1.05 ± 0.34
VAT%	82 ± 15	80 ± 14	86 ± 16	84 ± 10	77 ± 22	83 ± 16
Muscle strength (mean ± SD)						
Z-score	-2.1 ± 1.8	-2.3 ± 2.1	-1.9 ± 1.5	-2.7 ± 2.2	-2.0 ± 1.6	-1.8 ± 1.7
Weak strength: Z-score ± -2 (%)	61	58	64	79	57	54
Joint mobility (median [range])						
Number of restricted joints	1 [0 - 3]	1 [0 - 3]	1 [0 - 3]	0 [0 - 2]	1 [0 - 2]	1.5 [0 - 3]
Impaired mobility in any joint (%)	61	54	67	29	57	82
Body composition (mean ± SD)						
Sum of 4 skin-folds (mm)	74.8 ± 38.8	51.2 ± 24.6	100.4 ± 35.1	59.1 ± 29.2	66.5 ± 34.7	86.0 ± 42.0
% of reference values	159 ± 77	146 ± 79	173 ± 73	121 ± 52	160 ± 101	181 ± 75

* Only for those measured on the cycle ergometer, *n*= 17 (10 males, 7 females).

Table 3 Regression models for aerobic capacity.

Independent variables	Aerobic capacity (peakVO_2, in l/min)		
	β	*p*-value	R^2
Simple regression analysis			
Personal characteristics			
Gender	**-0.61**	< 0.001	0.36
Age	-0.07	0.65	-
Lesion level	**-0.43**	0.002	0.17
Hydrocephalus	-0.18	0.22	-
Ambulatory status	**-0.48**	< 0.001	0.22
Educational level	0.13	0.39	-
Multiple regression analysis			
Step 1[a]			
Personal characteristics			0.50
Gender	**-0.55**	< 0.001	
Ambulatory status	**-0.40**	< 0.001	
Step 2[b]			
Inter-relations			
Muscle strength (Z-score)	**0.22**	0.04	0.55
Body composition (Sum of 4 skin-folds in mm)	0.25	0.08	0.53
Joint mobility (number of restricted joints)	0.00	0.98	-

Significant betas are presented in bold. [a]Due to large overlap between ambulatory status and lesion level, we only included ambulatory status. [b]Relations between aerobic capacity and other fitness components controlled for gender and ambulatory status. β= standardized regression coefficient. R^2= explained variance. Gender male (0), female (1). Lesion level sacral (1), lumbosacral (2), lumbar (3), thoracolumbar (4), thoracic (5). Hydrocephalus no (0), yes (1). Ambulatory status community ambulator (1), household ambulator (2), non(functional) ambulator (3). Educational level: low (0), medium (1), high (2).

Discussion

Components of health-related physical fitness

In the present study, several health-related components of physical fitness were studied simultaneously in a relatively large group of adolescents and young adults with MMC. In general, most participants had poor health-related physical fitness.

Compared to the general population and compared to other patient groups, persons with MMC had low aerobic capacity. Regarding community ambulatory persons with MMC, values of peakVO_2 were 32% lower than the reference values for able-bodied people and 23% lower than the average peakVO_2 of males with spastic diplegia measured during cycle ergometry.[33] However, peakVO_2 is influenced by the amount of active muscle mass,[34-36] which may possibly be reduced due to paresis of lower extremity muscles. PeakVO_2 in non-ambulatory persons with MMC was lower than in ambulatory persons which may be caused by the lower amount of active muscle mass during arm ergometry compared to cycling. In able-bodied people, arm exercise induces a peakVO_2 ranging from 53 to 73%

of that achieved with lower extremity exercise.[37] If that ratio is also applicable to non-ambulatory persons who may be accustomed to using their arm and shoulder muscles, adapted values of average peakVO_2 would range between 1.76 and 2.43 l/min, which is of comparable range to the peakVO_2 in ambulatory persons with MMC. Furthermore, compared to males with spinal cord injuries with lesions below T10 measured during arm ergometry,[38] the peakVO_2 in non(functional)-ambulatory males with MMC was 22% lower. We therefore assumed that the poor aerobic capacity we found in persons with MMC may be influenced by reduced active muscle mass, but also deconditioning is likely to be present. This is supported by previous studies showing that adolescents and young adults with MMC were inactive, and that inactivity was associated with lower aerobic capacity,[39] particularly in non-ambulatory persons with MMC.[8]

VAT is another indicator of aerobic capacity, which was strongly correlated to peakVO_2 in the present study sample ($r = 0.84$; $p < 0.001$). In the general population, VAT roughly corresponds to 50–60% of VO_2max during leg exercise,[40] and values of 40–60% have been found in the able-bodied population during arm exercise.[41-43] Paradoxically, we found VAT to be at 82% of peakVO_2 suggesting high aerobic capacity. Comparably, Coutts and McKenzie found that persons with tetraplegia had lower VAT than persons with paraplegia caused by smaller muscle mass, but they had higher VAT%, and it was suggested that values of peakVO_2 were low in relation to VAT.[44]

Fifty-four percent of the non-ambulators and 79% of the community ambulators had subnormal strength in at least one of the major muscle groups. The relatively better performance of non-ambulators compared to ambulators in this respect might be explained by their relatively higher muscle strength in the upper extremities due to the habituation of using their arms for wheelchair propulsion. Furthermore, in contrast to lower extremity muscles, the neurological level of lesion will not disturb innervations of upper extremity muscles, which suggests that the weak upper extremity strength we found non-ambulators is due to atrophy of muscles as a consequence of disuse. Weak strength of lower extremity muscles in ambulatory persons with MMC corresponds to previous findings in children with MMC[9,12,13] and might be related to disturbed innervetions as well as to disuse of muscles.

Results of the present study support the findings of Verhoef and co-workers that a large proportion of persons with MMC have reduced joint mobility.[14] Similar to the results of Agre and co-workers restrictions in joint mobility in the lower extremities were mainly present in non-ambulatory persons with MMC.[9] We found high sum of four skin-folds, particularly in females and non-ambulatory, which is in agreement with high levels of body fat reported in previous studies.[15-17]

Relations between aerobic capacity and other components of health-related physical fitness

The results of the regression analyses indicated muscle strength was associated to peakVO_2 when controlling for gender and ambulatory status. This finding is in accordance with literature on persons with spinal cord injury.[18-20] In contrast to gender and ambulatory status, muscle strength is modifiable. Strength training may increase muscle mass and thus metabolizing mass contributing to higher peakVO_2.[19] In persons with spinal cord injury, strength training resulted in increased peakVO_2.[45] Because causality cannot be established with the cross-sectional design of the present study, future studies should confirm whether strength training results in increased aerobic capacity in persons with MMC. However, considering the small, but significant, contribution of muscle strength to the explained variance, we assume this specific effect to be small. In this respect, it seems that mainly aerobic training is needed in order to improve aerobic capacity; however, including strength training may have additional value.

Body composition tended to be positively related to aerobic capacity, indicating that persons with more body fat had higher values of peakVO_2. This relation may be caused by the greater fat-free mass of overweight persons concomitant with a greater body size.[46]

Joint mobility was not related to aerobic capacity. However, in non-ambulators we might have underestimated the relation because we did not measure mobility of upper extremity joints. Nevertheless, good flexibility of lower extremity is suggested to be important to prevent problems later in life, such as problems with personal hygiene and transfer capabilities.[9]

Limitations of the study

The methodology of the study had some limitations. First, because it is suggested that the primary mode of ambulation elicits the highest values of peakVO_2,[24] we used different exercise modes to assess aerobic capacity for ambulatory and non-ambulatory persons. However, due to large differences in active muscle mass, arm and leg exercise have different physiological responses. Therefore, we corrected for ambulatory status as proxy measure for type of ergometer when analysing relations between aerobic capacity and the other health-related fitness components. Furthermore, because the main mode of ambulation of ambulatory persons is walking rather than cycling, we may have underestimated peakVO_2 in some ambulatory persons with MMC who rarely cycle. However, in clinical practice, the cycle ergometer seems to be more practical because physically disabled people may experience severe balance problems on the treadmill, and on the cycle it is possible to strap the feet to the pedals.[33] Peripheral local fatigue may have caused exercise cessation before reaching maximum oxygen uptake, however, based on the objective (peakHR and peakRER) and subjective criteria of maximal exercise (RPE), it may be concluded that values of peakVO_2 are reasonable.

Furthermore, muscle strength was measured using handheld dynamometry. This method is known to be cheap, quickly applicable[47] and reliable.[32] However, for an average examiner, values above 250 N are considered too high to apply sufficient resistance,[30] which may lead to less accurate results in strong muscle groups. Using absolute values of strength measured with an isokinetic device, instead of expressing muscle strength as Z-score, might provide more detailed insight into the relation between strength peakVO_2. However, the current measurement method and use of Z-scores are considered adequate to determine weakness of major muscle groups.

Finally, the response rate was low, which hampers generalization of results. However, personal and disease-related characteristics did not differ between participants and non-participants. Moreover, the prevalence of middle-level (lumbosacral) and high-level (lumbar or thoracolumbar) lesions of the present study sample was comparable to the persons who participated in the national ASPINE study.[14]

Despite, a selection bias may have occurred since it could be that the more active and more fit people had higher interest in participating than the less active and less fit ones, which may have led to an overestimation of health-related physical fitness components.

In conclusion, the results of the present study show that both ambulatory and non-ambulatory adolescents and young adults with MMC have poor health-related physical fitness. A large part of the variance in aerobic capacity is explained by gender and ambulatory status. Results showed a small but significant relationship between peakVO_2 and muscle strength, suggesting that adding strength training to aerobic training may have additional value in increasing peakVO_2.

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References

1. Bowman RM, McLone DG, Grant JA, Tomita T, Ito JA. Spina bifida outcome: a 25-year prospective. Pediatr Neurosurg 2001; 34: 114–120.
2. Lamb KL, Brodie DA, Roberts K. Physical fitness and health-related fitness as indicators of a positive health state. Health Promot Int 1988; 3: 171–182.
3. Twisk JW, Kemper HC, van Mechelen W. The relationship between physical fitness and physical activity during adolescence and cardiovascular disease risk factors at adult age. The Amsterdam Growth and Health Longitudinal Study. Int J Sports Med 2002; 23(Suppl 1): S8–S14.
4. Noreau L, Shephard RJ. Spinal cord injury, exercise and quality of life. Sports Med 1995; 20: 226–250.
5. Stewart AL, Hays RD, Wells KB, Rogers WH, Spritzer KL, Greenfield S. Long-term functioning and well-being outcomes associated with physical activity and exercise in patients with chronic conditions in the Medical Outcomes Study. J Clin Epidemiol 1994; 47: 719–730.
6. Bruinings AL, van den Berg-Emons HJ, Buffart LM, van der Heijden-Maessen HC, Roebroeck ME, Stam HJ. Energy cost and physical strain of daily activities in adolescents and young adults with myelomeningocele. Dev Med Child Neurol 2007; 49: 672–677.
7. Caspersen CJ, Powell KE, Christenson GM. Physical activity, exercise, and physical fitness: definitions and distinctions for health-related research. Public Health Rep 1985; 100: 126–131.
8. Buffart LM, Roebroeck ME, Rol M, Stam HJ, van den Berg-Emons RJG. Triad of physical activity, aerobic fitness and obesity in adolescents and young adults with myelomeningocele. J Rehabil Med 2008; 40: 70–75.
9. Agre JC, Findley TW, McNally MC, Habeck R, Leon AS, Stradel L, Birkebak R, Schmalz R. Physical activity capacity in children with myelomeningocele. Arch Phys Med Rehabil 1987; 68: 372–377.
10. Sherman MS, Kaplan JM, Effgen S, Campbell D, Dold F. Pulmonary dysfunction and reduced exercise capacity in patients with myelomeningocele. J Pediatr 1997; 131: 413–418.
11. Van den Berg-Emons HJ, Bussmann JB, Meyerink HJ, Roebroeck ME, Stam HJ. Body fat, fitness and level of everyday physical activity in adolescents and young adults with meningomyelocele. J Rehabil Med 2003; 35: 271–275.
12. McDonald CM, Jaffe KM, Mosca VS, Shurtleff DB. Ambulatory outcome of children with myelomeningocele: effect of lower-extremity muscle strength. Dev Med Child Neurol 1991; 33: 482–490.
13. Schoenmakers MA, Gulmans VA, Gooskens RH, Helders PJ. Spina bifida at the sacral level: more than minor gait disturbances. Clin Rehabil 2004; 18: 178–185.
14. 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: 420–427.
15. Bandini LG, Schoeller DA, Fukagawa NK, Wykes LJ, Dietz WH. Body composition and energy expenditure in adolescents with cerebral palsy or myelodysplasia. Pediatr Res 1991; 29: 70–77.

16. 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: 305–311.
17. Shepherd K, Roberts D, Golding S, Thomas BJ, Shepherd RW. Body composition in myelomeningocele. Am J Clin Nutr 1991; 53: 1–6.
18. Haisma JA, Bussmann JB, Stam HJ, Sluis TA, Bergen MP, Dallmeijer AJ, de Groot S, van der Woude LH. Changes in physical capacity during and after inpatient rehabilitation in subjects with a spinal cord injury. Arch Phys Med Rehabil 2006; 87: 741–748.
19. Janssen TW, van Oers CA, Hollander AP, Veeger HE, van der Woude LH. Isometric strength, sprint power, and aerobic power in individuals with a spinal cord injury. Med Sci Sports Exerc 1993; 25: 863–870.
20. Zoeller RF Jr, Riechman SE, Dabayebeh IM, Goss FL, Robertson RJ, Jacobs PL. Relation between muscular strength and cardiorespiratory fitness in people with thoracic-level paraplegia. Arch Phys Med Rehabil 2005; 86: 1441–1446.
21. 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.
22. Donkervoort M, Roebroeck M, Wiegerink D, van der Heijden-Maessen H, Stam H, The Transition Research Group South West Netherlands. Determinants of functioning of adolescents and young adults with cerebral palsy. Disabil Rehabil 2007; 29: 453–463.
23. Bar-Or O. Pediatric sports medicine for the practioner. From physiologic principles to clinical applications. Springer, New York; 1983.
24. Bhambhani YN, Holland LJ, Steadward RD. Maximal aerobic power in cerebral palsied wheelchair athletes: validity and reliability. Arch Phys Med Rehabil 1992; 73: 246–252.
25. Vos JA. Ergometrie en Trainingsbegeleiding. Nederlands Paramedisch Instituut, Amersfoort; 2001.
26. Reinhard U, Muller PH, Schmulling RM. Determination of anaerobic threshold by the ventilation equivalent in normal individuals. Respir Int Rev Thorac Dis 1979; 38: 36–42.
27. Wassermann K, Hansen JE, Sue DY, Whipp BJ, Casaburi R. Principles of exercise testing and interpretation. Lippincot Williams and Wilkins, Philadelphia; 1999.
28. Borg GA. Psychophysical bases of perceived exertion. Med Sci Sports Exerc 1982; 14: 377–381.
29. Mahler DA, Rosiello RA, Harver A, Lentine T, McGovern JF, Daubenspeck JA. Comparison of clinical dyspnea ratings and psychophysical measurements of respiratory sensation in obstructive airway disease. Am Rev Respir Dis 1987; 135: 1229–1233.
30. Van der Ploeg RJ, Fidler V, Oosterhuis HJ. Hand-held myometry: reference values. J Neurol Neurosurg Psychiatry 1992; 54: 244–247.
31. Phillips BA, Lo SK, Mastaglia FL. Muscle force measured using “break” testing with a hand-held myometer in normal subjects aged 20 to 69 years. Arch Phys Med Rehabil 2000; 81: 653–661.
32. Bohannon RW. Reference values for extremity muscle strength obtained by hand-held dynamometry from adults aged 20 to 79 years. Arch Phys Med Rehabil 1997; 78: 26–32.
33. Lundberg A. Maximal aerobic capacity of young people with spastic cerebral palsy. Dev Med Child Neurol 1978; 20: 205–210.
34. Davies CT, Sargeant AJ. Indirect determination of maximal aerobic power output during work with one or two limbs. Eur J Appl Physiol Occup Physiol 1974; 32: 207–215.
35. Davies CT, Sargeant AJ. Effects of training on the physiological responses to one- and two-leg work. J Appl Physiol 1975; 38: 377–375.

36. Lewis SF, Taylor WF, Graham RM, Pettinger WA, Schutte JE, Blomqvist CG. Cardiovascular responses to exercise as functions of absolute and relative work load. J Appl Physiol 1983; 54: 1314–1323.
37. Bar-Or O, Zwiren LD (1975) Maximal oxygen consumption test during arm exercise—reliability and validity. J Appl Physiol 1975; 38: 424–426.
38. Janssen TW, Dallmeijer AJ, Veeger DJ, van der Woude LH (2002) Normative values and determinants of physical capacity in individuals with spinal cord injury. J Rehabil Res Dev 2002; 39: 29–39.
39. Van den Berg-Emons HJ, Bussmann JB, Brobbel AS, Roebroeck ME, van Meeteren J, Stam HJ. Everyday physical activity in adolescents and young adults with meningomyelocele as measured with a novel activity monitor. J Pediatr 2001; 139: 880–886.
40. Davis JA, Storer TW, Caiozzo VJ. Prediction of normal values for lactate threshold estimated by gas exchange in men and women. Eur J Appl Physiol Occup Physiol 1997; 76:157–164.
41. Flandrois R, Grandmontagne M, Gerin H, Mayet MH, Jehl JL, Eyssette M. Aerobic performance capacity in paraplegic subjects. Eur J Appl Physiol Occup Physiol 1986; 55: 604–609.
42. Lin KH, Lai JS, Kao MJ, Lien IN. Anaerobic threshold and maximal oxygen consumption during arm cranking exercise in paraplegia. Arch Phys Med Rehabil 1993; 74: 515–520.
43. Schneider DA, Sedlock DA, Gass E, Gass G. VO_2peak and the gas-exchange anaerobic threshold during incremental arm cranking in able-bodied and paraplegic men. Eur J Appl Physiol Occup Physiol 1999; 80: 292–297.
44. Coutts KD, McKenzie DC. Ventilatory thresholds during wheelchair exercise in individuals with spinal cord injuries. Paraplegia 1995; 33: 419–422.
45. Cooney MM, Walker JB. Hydraulic resistance exercise benefits cardiovascular fitness of spinal cord injured. Med Sci Sports Exerc 1986; 8: 522–525.
46. Unnithan VB, Houser W, Fernhall B. Evaluation of the energy cost of playing a dance simulation video game in overweight and non-overweight children and adolescents. Int J Sports Med 2006; 27: 804–809.
47. Van der Ploeg RJ, Oosterhuis HJ, Reuvekamp J. Measuring muscle strength. J Neurol 1984; 231: 200–203.

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Chapter 4

Energy Cost and Physical Strain in Adolescents and Young Adults with Myelomeningocele

Dev Med Child Neurol 2007; 49: 672-677

Abstract

The aim of this study was to assess the energy cost and physical strain of daily activities in adolescents and young adults with myelomeningocele (MMC) compared with peers without a disability.

Eighteen participants with MMC aged between 16 and 30 years (13 males, five females; mean age 21y 4mo [SD 4y 8mo]) and 18 age- and sex-matched nondisabled participants performed several standardized activities. Energy cost was assessed by oxygen uptake expressed per unit time (all activities) and per metre (walking and wheelchair use at preferred speed). Physical strain was calculated by dividing energy cost by aerobic capacity.

For all activities no difference was found in energy cost per unit time between ambulatory participants with MMC and comparison participants. Energy cost per metre walking at preferred speed in participants with MMC was 0.26ml/kg/m (SD 0.08), and in comparison participants was 0.20ml/kg/m (SD 0.03); $p = 0.08$. Non-ambulatory participants with MMC had lower energy cost (per unit time and per metre) during wheelchair use than comparison participants during walking ($p < 0.05$). For most activities, physical strain was 1.4 to 2 times higher in participants with MMC than in comparison participants ($p < 0.05$).

In conclusion, energy cost per unit time of daily activities was not increased in participants with MMC. However, energy cost per metre during walking at preferred speed and physical strain were higher than in peers without disability.

Introduction

Myelomeningocele (MMC) is a consequence of failed closure of the neural tube in the vertebral column, occurring in approximately 1 out of 1000 live births.[1,2] MMC is associated with abnormalities in the brain, including hydrocephalus and Arnold-Chiari type II malformations. Other important medical problems are urinary and faecal incontinence, reduced mobility, pressure sores, and orthopaedic problems such as scoliosis and contractures.[3]

The literature shows that individuals with MMC are hypoactive,[4,5] which may have a negative effect on physical fitness,[6] social participation, and quality of life.[7] Furthermore, a hypoactive lifestyle may increase the chance of developing cardiovascular disease and diabetes mellitus type II later in life.[8] A study by van den Berg-Emons et al. found that adolescents and young adults with MMC performed, on average, 1.5 hours of dynamic activities (such as walking and wheelchair use) per day, whereas their non-disabled peers performed, on average, 3 hours of dynamic activities per day.[5] Bandini et al. reported that total daily energy expenditure and resting metabolic rate were subnormal in adolescents with MMC, particularly in non-ambulators.[4] Lower fat-free body mass and lower activity level were proposed as main causes for this finding.[4]

It has been suggested that ambulatory individuals with MMC have a high energy cost during walking due to disturbed equilibrium caused by hip-abductor weakness leading to an energy-expensive lateral trunk sway[9] or increased pelvic obliquity.[10] It may be hypothesized that the high energy cost of daily activities, such as walking, leads to increased fatigue and discomfort and, consequently, to a hypoactive lifestyle, as identified in those with MMC.[4,5] In addition to the high-energy cost of daily activities, the physical strain (energy cost relative to aerobic capacity) of daily activities is expected to be high in individuals with MMC, particularly because aerobic capacity is subnormal in these individuals.[6,11–13]

The literature on energy cost in individuals with MMC is scarce, inconclusive, and focuses primarily on children. Energy cost per metre walking (oxygen [O_2] cost in ml/kg/metre) was found to be higher in children with MMC than in children without disabilities.[9,10,14] However, contradictory results were found for energy cost per unit time (O_2 cost in ml/kg/min) during walking at a self-selected velocity. Duffy et al.[9] found similar energy costs in children with MMC and peers without disabilities, whereas Bare et al.[10] found higher energy cost in children with MMC. Williams et al. found similar energy cost per unit time at self-selected velocity, but at faster walking velocities energy cost was higher in children with MMC compared with non-disabled peers.[14] Lower energy cost during wheelchair use compared with walking, allowing long-distance locomotion at normal velocity is also reported.[12,14,15]

In adolescents and young adults with MMC, little is known about energy cost or physical strain during locomotion and no study is available on energy cost and physical strain of activities other than locomotion. The aim of this study was to assess the energy cost and physical strain of daily activities in ambulatory and non-ambulatory adolescents and young adults with MMC compared with non-disabled peers.

Method

Participants

The study sample consisted of 18 consecutively-selected participants from a cohort of 51 participants with MMC aged between 16 and 30 years who participated in a large cross-sectional study on daily physical activity in MMC. Participants in the cohort were recruited from university hospitals in Rotterdam, Leiden, Utrecht, and Amsterdam, and all rehabilitation centres in the southwest of the Netherlands. Exclusion criteria for the cohort were: (1) complete dependence on an electric wheelchair; (2) inability to understand instructions necessary for the study; (3) presence of disorders other than MMC interfering with daily physical activity (e.g. rheumatoid arthritis); and (4) disorders that contraindicate a maximal exercise test (e.g. exercise-induced ischemia or arrhythmias, uncontrolled hypertension, and exercise limitation due to chronic obstructive pulmonary disease). Mean age of the cohort was 21 years (SD 4y 6mo; 26 males, 25 females).

Ambulatory status was defined according to the Hoffer classification:[16] 15 participants were community ambulators walking indoors and outdoors, eight were household ambulators walking only indoors, and 28 were wheelchair dependent. Twenty-one participants agreed to take part in the current study, of whom 18 completed the assessment successfully (13 males, five females; mean age 21y 4mo [SD 4y 8mo]). In three participants, assessments failed due to technical problems. Information about neurological lesion level, the presence of hydrocephalus, and history of scoliosis and tethered cord was collected from medical records.

Eighteen non-disabled comparison participants, matched for sex and age (plus or minus 3y) participated in the study. They were recruited from nearby schools and a university. All participants were informed about the study and written consent was obtained. The Medical Ethics Committee of the Erasmus Medical Center approved the study.

Participant characteristics are presented in Table 1. In participants with MMC, height was measured with a flexible tape while lying on a bed. For participants with contractures, measurements were performed from joint to joint. In the comparison group, height was measured with a static measure while participants were standing against a wall. Body mass of non-ambulators was obtained while sitting on an electronic scale (Cormier Paribel [FH Balances]; Cormier, Romanville, France); body mass of ambulators with MMC and of the comparison participants was measured while standing on a Seca scale (Seca GmbH and Co., Hamburg, Germany).

Table 1 Physical characteristics of participants with myelomeningocele (MMC) and comparison groups.

	MMC[a] ambulatory (*n*= 8)	**Comparison group** (*n*= 8)	***p*-value[b]**	**MMC non-ambulatory** (*n*= 10)	**Comparison group** (*n*= 10)	***p*-value[a]**
Age, y:m	22:6 (4:10)	23.9 (4:8)		20:4 (4:7)	21:6 (3:8)	
Males/Females	6/2	6/2		7/3	7/3	
Neurological level of lesion						
Thoracolumbar	0			1		
Lumbar	2			3		
Lumbosacral	2			6		
Sacral	4			0		
Hydrocephalus	5			9		
History of scoliosis	1			6		
Tethered cord	4			3		
Ambulatory status						
Community	8			0		
Household	0			1		
Non(functional)	0			9		
Height, cm	169.4 (9.8)	181.4 (5.7)	0.03[b]	156.6 (9.1)	181.9 (8.8)	<0.001[b]
Body mass, kg	63.6 (6.5)	74.0 (9.7)	0.03[b]	68.7 (15.3)	73.5 (8.2)	0.25
PeakVO_2, ml/kg/min	31.4 (8.3)	47.9 (11.2)	0.02[b]	23.5 (5.3)	51.0 (9.6)	<0.001[b]

Data are presented as mean (standard deviation, SD) or *n*. [a]Participants with MMC versus non-disabled comparison participants analyzed with independent t-test. [b]$p < 0.05$. VO_2, oxygen uptake.

Measurements

Activity protocol

In a laboratory setting, participants performed a circuit of several activities of daily life, according to a standard protocol (Table 2). Activities were mainly related to locomotion (walking, wheelchair use, cycling) and to housekeeping. For ambulatory participants, the circuit consisted of 12 activities and for non-ambulatory participants there were five activities. The circuit started with a 3-minute rest period in a (wheel)chair, supported with the back of the chair and armrests. Each activity was performed over 3 minutes to achieve a steady state with one minute rest between activities. Participants were asked to perform the activities continuously and at a constant speed. Walking and wheelchair use were performed at several fixed speeds and at the participant's preferred speed on a trajectory of 12 metres (smooth turns at the corners). For fixed speeds, a researcher determined the speed using a one-wheeled distance metre to which a calibrated real-time cycle-speed computer was attached. The researcher walked at the prescribed speed and participants were asked to follow the researcher. For the housekeeping activities ironing and washing dishes, the height of the workplace was adjusted for ambulators and non-ambulators. Cycling was performed on an electronically-braked cycle ergometre (Jaeger ER800; Jaeger Toennies, Breda, the Netherlands). As in their daily lives, most ambulatory participants with MMC used orthopaedic shoes or orthoses during the protocol, but none of them used crutches, sticks, or walking frames.

Table 2 Standard activity protocol for myelomeningocele (MMC) subgroups and the comparison groups.

Activities	Community ambulatory MMC and comparison participants	Non-ambulatory MMC
Standing	+	
Walking at preferred speed	+	
Walking at 2.5 km/h	+	
Walking at 4.5 km/h	+	
Wheelchair use at preferred speed		+
Wheelchair use at 2.5 km/h		+
Wheelchair use at 4.5 km/h		+
Ironing	+	+
Washing dishes	+	+
Vacuum cleaning	+	
Cycling at 20 watt	+	
Cycling at 40 watt	+	
Cycling at 60 watt	+	
Cycling at 80 watt	+	
Cycling at 100 watt	+	

+, denotes activities that were performed.

Energy cost of daily activities

During the activity protocol, energy cost of daily activities was assessed by measuring oxygen uptake (VO_2). Gas volume and gas concentrations were determined continuously with a portable breath-by-breath gas analyzer and a latex-free face mask ($K4b^2$; Cosmed, Rome, Italy). Gas analyses were made by a galvanic fuel cell (oxygen) and an infrared carbon dioxide analyzer. Energy cost of each activity per unit time was defined as mean VO_2 during the last minute of the activity (VO_2ml/kg/min). In addition, energy cost per metre was calculated for walking and wheelchair use at preferred speed (VO_2ml/kg/m).

Physical strain of daily activities

Physical strain was defined as VO_2 (per unit time) during activity divided by aerobic capacity (×100%). In participants with MMC, aerobic capacity was measured during a progressive maximal exercise test on an electronically-braked arm or cycle ergometer (Jaeger ER800SH and ER800 respectively; Jaeger Toennies, Breda, the Netherlands), depending on the main mode of ambulation. In a study of participants with cerebral palsy who were partly wheelchair-dependent, Bhambhani et al. concluded that maximal exercise testing during the main mode of ambulation elicits the highest VO_2.[17] Therefore, 10 participants with MMC performed an arm crank ergometer test while sitting in their own immobilized wheelchair with cranks at shoulder height, and eight participants with

MMC performed a leg cycle ergometer test. The test was based on the McMaster All-out Progressive Continuous Cycling and Arm Test.[18] The test was preceded by a 3-minute warm-up (5 watts (W) for the arm ergometer, 20W for the cycle ergometer), followed by a resting period of 5 minutes. During the test, resistance was increased every 2 minutes with a variable load, depending on the ability of the participant.

Individual protocols were constructed so that total exercise time ranged from 8 to 12 minutes. The pedal/crank rate for the arm and leg tests was 60rpm; strong verbal encouragement was given throughout the test. The test was terminated when the participant voluntarily stopped due to exhaustion, or when the participant was unable to maintain the initial pedal/crank rate. VO_2 was measured with a portable breath-by-breath gas analyzer and a latex-free face mask. Aerobic capacity was defined as mean VO_2 during the last minute of exercise (peakVO_2) and was expressed per kilogram of body mass.

For practical reasons, maximal exercise tests were only performed in participants with MMC. In the non-disabled comparison group, aerobic capacity was extrapolated from VO_2 and heart rate as measured during cycling at 100W (activity protocol) using the nomogram presented by Astrand and Rodahl.[19]

Statistical analysis

All data are presented as mean (SD) or *n* for the ambulatory and non-ambulatory MMC subgroups and the comparison groups. Differences were tested between MMC and comparison groups with an independent *t*-test. Because the main mode of ambulation in the non-ambulatory participants with MMC was wheelchair use, energy cost during wheelchair use in this group was compared with energy cost during the main mode of ambulation (walking) in the non-disabled comparison participants. Statistical significance was set at $p < 0.05$.

Results

Ambulatory participants with MMC were, on average, 12cm shorter (7%, $p = 0.03$) than non-disabled comparison participants; non-ambulatory participants with MMC were, on average 25.3cm shorter (14%, $p < 0.001$) than non-disabled participants (Table 1). Body mass of ambulatory participants with MMC was 10.4kg lower (14%, $p = 0.03$) than that of their non-disabled peers.

Energy cost of daily activities

Energy cost of daily activities per unit time is presented in Table 3. For all activities, no significant difference was found in energy cost per unit time between ambulatory participants with MMC and the comparison group. Preferred walking speed of ambulatory participants with MMC (3.2 [SD 0.8] km/h) was 14% lower ($p = 0.22$) than the preferred speed of comparison participants (3.7 [SD 0.8] km/h).

Table 3 Energy cost of daily activities per unit time, expressed as oxygen uptake (VO_2) in ml/kg/min.

Activity	Ambulatory				
	MMC (*n*= 8)	Comparison (*n*= 8)	Difference	Confidence interval (95%)	*p*-value[a]
	Mean (SD)	**Mean (SD)**			
Sitting	4.2 (1.0)	4.0 (1.2)	0.16	-1.06 to 1.37	0.78
Standing	5.0 (1.2)	4.2 (1.1)	0.74	-0.47 to 1.95	0.21
Walking preferred speed	13.2 (3.4)	12.7 (3.4)	0.52	-3.09 to 4.13	0.76
Walking 2.5 km/h	10.9 (2.0)	10.4 (2.0)	0.45	-1.70 to 2.60	0.66
Walking 4.5 km/h	15.1 (2.9)	14.8 (1.2)	0.31	-2.04 to 2.65	0.78
Ironing	9.7 (3.0)	8.8 (1.8)	0.94	-1.77 to 3.67	0.48
Washing dishes	8.8 (2.5)	9.5 (2.0)	0.62	-3.02 to 1.77	0.59
Vacuum cleaning	12.0 (2.6)	11.3 (2.9)	0.72	-2.22 to 3.65	0.61
Cycling; 20 watt	12.0 (2.9)	10.1 (1.9)	1.96	-0.66 to 4.58	0.13
Cycling; 40 watt	15.7 (4.5)	13.3 (2.5)	2.38	-1.52 to 6.28	0.21
Cycling; 60 watt	19.0 (3.9)	16.3 (2.7)	2.88	-0.76 to 6.49	0.11
Cycling; 80 watt (*n*= 6)[b]	21.9 (4.5)	19.7 (3.2)	2.22	-2.78 to 7.23	0.35
Cycling; 100 watt (*n*= 4)[b]	24.1 (1.8)	23.2 (3.2)	0.91	-3.58 to 5.40	0.64
Activity	**Non-ambulatory**				
	MMC (*n*= 10)	Comparison (*n*= 10)	Difference	Confidence interval (95%)	*p*-value[b]
	Mean (SD)	**Mean (SD)**			
Sitting	3.8 (1.1)	4.1 (1.1)	-0.34	-1.44 to 0.77	0.53
Walking or wheelchair use; preferred speed[c]	9.7 (3.8)	12.8 (3.1)	-3.15	-6.46 to 0.16	0.06
Walking or wheelchair use; 2.5 km/h[c]	6.4 (1.7)	9.4 (1.3)	-3.02	-4.47 to -1.57	< 0.001[d]
Walking or wheelchair use; 4.5 km/h[c]	8.7 (2.3)	12.5 (2.0)	-3.81	-5.83 to -1.79	< 0.001[d]
Ironing	7.5 (2.4)	7.3 (1.8)	0.13	-1.87 to 2.12	0.90
Washing dishes	7.7 (2.2)	7.9 (1.2)	-0.21	-1.92 to 1.50	0.80

[a]Participants with myelomeningocele (MMC) versus non-disabled comparison participants analyzed with independent t-test. [b]Four ambulatory participants with MMC stopped cycling at higher loads, two participants stopped after performing at 60W, and two participants stopped after performing at 80W. In statistical analysis their matched comparison participants were also excluded from cycling at 80 and 100W. [c]Wheelchair use in participants with MMC versus walking in non-disabled comparison participants. [d]$p< 0.05$.

Table 4 Physical strain of daily activities, calculated as VO_2 (ml/kg/min) during activity divided by peakVO_2 (ml/kg/min, x100%).

Activity	Ambulatory				
	MMC (*n*= 8)	Comparison (*n*= 8)	Difference	Confidence interval (95%)	*p*-value[a]
	Mean (SD)	**Mean (SD)**			
Sitting	14.1 (4.9)	8.7 (2.4)	5.48	1.33 to 9.61	0.01[b]
Standing	16.7 (5.5)	9.0 (2.1)	7.69	3.00 to 12.37	0.01[b]
Walking preferred speed	44.8 (15.5)	27.1 (6.3)	17.76	4.47 to 31.05	0.01[b]
Walking 2.5 km/h	36.9 (11.3)	22.3 (3.8)	14.64	5.01 to 24.26	0.01[b]
Walking 4.5 km/h	51.5 (17.0)	32.4 (7.9)	19.07	4.29 to 33.86	0.02[b]
Ironing	31.9 (11.1)	18.8 (4.3)	8.86	0.44 to 16.93	0.03[b]
Washing dishes	29.2 (9.7)	20.3 (4.3)	13.12	3.61 to 22.15	0.01[b]
Vacuum cleaning	40.2 (11.8)	24.3 (7.0)	18.84	5.40 to 26.28	0.01[b]
Cycling; 20 watt	41.1 (14.7)	21.6 (4.1)	19.49	7.05 to 31.92	0.01[b]
Cycling; 40 watt	53.6 (20.5)	28.6 (5.3)	25.02	7.73 to 42.30	0.01[b]
Cycling; 60 watt	65.3 (23.9)	34.7 (6.4)	30.64	10.46 to 50.82	0.01[b]
Cycling; 80 watt (*n* = 6)[c]	70.2 (28.7)	41.3 (9.6)	28.90	-1.23 to 59.08	0.06
Cycling; 100 watt (*n* = 4)[c]	66.6 (16.8)	48.6 (9.5)	18.04	-5.54 to 41.61	0.11
Activity	**Non-ambulatory**				
	MMC (*n*= 10)	Comparison (*n*= 10)	Difference	Confidence interval (95%)	*p*-value[b]
	Mean (SD)	**Mean (SD)**			
Sitting	16.2 (3.3)	8.5 (2.7)	7.68	4.82 to 10.54	< 0.001[b]
Walking or wheel-chair use; preferred speed[d]	41.0 (11.9)	26.5 (10.3)	14.50	4.04 to 24.95	0.01[b]
Walking or wheel-chair use; 2.5 km/h[d]	27.3 (4.4)	19.2 (4.6)	8.17	3.92 to 12.41	< 0.001[b]
Walking or wheel-chair use; 4.5 km/h[d]	37.8 (8.5)	25.5 (6.3)	12.32	5.30 to 19.34	0.002[b]
Ironing	32.7 (5.0)	16.1 (4.0)	16.50	10.67 to 22.41	< 0.001[b]
Washing dishes	31.7 (7.2)	15.1 (5.2)	16.60	12.37 to 20.87	< 0.001[b]

[a]Participants with myelomeningocele (MMC) versus non-disabled comparison participants analyzed with independent t-test. [b]$p < 0.05$. [c]Four ambulatory participants with MMC stopped cycling at higher loads. Two stopped after performing at 60W, two stopped after performing at 80W. In statistical analysis their matched comparison participants were also excluded from cycling at 80 and 100W. [d]Wheelchair use in participants with MMC versus walking in non-disabled comparison participants.

In non-ambulatory participants with MMC, energy cost during wheelchair use per unit time, both at the preferred speed (p= 0.06) and at fixed speeds (p<0.001), was lower than energy cost during walking at corresponding speeds in the comparison group. Preferred speed of wheelchair use of non-ambulatory participants with MMC was 3.9 (0.6) km/h, whereas walking at preferred speed of their comparison participants was 4.1 (0.7) km/h (p= 0.36).

Energy cost per metre walking at preferred speed was 0.26 (0.08) ml/kg/m in ambulatory participants with MMC and 0.20 (0.03) ml/kg/m in comparison participants (p= 0.08). Energy cost per metre wheelchair use at preferred speed was 0.15 (0.04), which was 21% lower than energy cost per metre walking in the comparison group (p= 0.03).

Physical strain of daily activities

PeakVO_2 was 16.5ml/kg/min (34%, p= 0.02) and 27.5ml/kg/min (54%, p< 0.001) lower in ambulatory and non-ambulatory participants with MMC respectively than in comparison participants (Table 1). Table 4 presents data on the physical strain of daily activities. Ambulatory and non-ambulatory participants with MMC had higher (1.4–2 times, p< 0.05) physical strain than non-disabled comparison participants in all activities, except for cycling at 80W and 100W.

Discussion

This study compared individuals with MMC and non-disabled comparison peers with regard to energy cost and physical strain during daily activities. Results on energy cost in comparison participants are in agreement with rates of energy expenditure presented in the 'Compendium of Physical Activities', a coding scheme that classifies specific physical activities in the general population by rate of energy expenditure.[20] Thus, the comparison group in the present study is considered to be representative of the general population.

Results show that the energy cost of daily activities per unit time in participants with MMC is comparable with, or even lower (for wheelchair use) than in non-disabled comparison participants. However, per metre walking at preferred speed, energy cost tended to be higher in participants with MMC than in non-disabled comparison participants, which is in agreement with the literature.[9,10,14] Results indicate that participants with MMC perform daily activities using a considerably higher percentage of their aerobic capacity than non-disabled comparison participants, indicating a higher physical strain. This high physical strain is mainly caused by a decreased aerobic capacity. The low aerobic capacity found in participants with MMC is in agreement with previous reports.[6,11–13]

High energy cost per metre walking at preferred speed and high levels of physical strain during daily activities may restrict activities of daily life, primarily as a consequence of increased fatigue and discomfort,[21] and may, therefore, contribute to a hypoactive lifestyle.

Physical rehabilitation programmes aimed at improving aerobic capacity may be effective in reducing the energy cost per metre walking at preferred speed and physical strain in daily life of participants with MMC. As a consequence, those with MMC participating in physical rehabilitation may break through the negative spiral of hypoactivity as found in this patient group.[4,5] However, this has to be confirmed in future studies.

Considering that hypoactivity is found to be more pronounced in non-ambulatory than in ambulatory participants with MMC,[5] and that physical strain during wheelchair use was lower than during walking, factors other than physical strain seem to contribute to the hypoactive lifestyle in nonambulatory participants with MMC; for example, restricted mobility in small rooms might play a role.

The finding that energy cost per unit time during walking was comparable between participants with MMC and non-disabled peers is in accordance with the results of Duffy et al.[9] but in contrast with the results of Bare et al.[10] (possibly as a result of differences in lower-limb muscle strength between participants in the current study and those in the study by Bare et al.). Although differences between ambulatory participants with MMC and comparison participants in preferred walking speed were not statistically significant, it appears that, by modifying walking velocity the energy cost per unit time could remain close to normal in participants with MMC.

Although ambulatory participants with MMC had a higher oxygen cost per metre walking at preferred speed than comparison participants, energy cost during walking at fixed speeds did not differ, contrary to the researchers' expectations. This is also in contrast to results of Williams et al. which indicate that children with MMC have higher energy costs per metre at faster velocities.[14]

The energy cost (per unit time and per metre) of the main mode of ambulation in the non-ambulatory participants with MMC (wheelchair use) was lower than the energy cost of walking in the comparison group. This suggests that wheelchair use is more energy-efficient than walking, and allows participants with MMC to have a lower energy cost during locomotion.

Some limitations of this study were identified. First, because of the relatively small sample size, some differences between groups may have failed to show statistical significance. However, the small study sample does not appear to have greatly affected the main results of the study.

Second, household ambulators were not included. In the cross-sectional study on daily physical activity in MMC, of which the current study is a part, only eight participants were household ambulators, and these participants were not available during the study period or were not able to walk continuously for 3 minutes. It may be hypothesized that, during adolescence, household ambulators tend to become more wheelchair dependent. Bartonek et al. found a downward transition in ambulatory status in a 12-year follow-up study of 60 ambulatory participants with MMC (median age 23y; range 12–54y), which was mainly due to deterioration of neurological lesion level, spasticity, and knee and hip flexion contractures.[22] Asher and Olson found that motivation, obesity, and musculoskeletal deformity are also factors causing this downward transition in ambulatory status.[23] It seems likely, therefore, that at the age of young adulthood, relatively good walkers (community ambulators) are more likely to remain ambulatory, and more restricted ambulators (household ambulators) tend to be wheelchair dependent. Future research should focus on the energy cost of daily activities in adolescents and young adults with MMC who are household ambulatory.

Third, in the non-disabled comparison group, peakVO_2 was estimated from submaximal VO_2 by the nomogram as described by Astrand and Rodahl,[19] whereas in the participants with MMC, peakVO_2 was measured directly. However, because the method of Astrand and Rodahl is shown to be reliable and valid for estimating peakVO_2 in non-disabled participants,[24,25] it can be assumed that the discrepancy in testing methods between the study groups has not greatly affected our results.

Finally, the activity protocol included activities related to locomotion and some housekeeping activities performed in a laboratory setting over only 3 minutes. It should also be noted that participants with MMC rarely cycled in their daily life. Future studies should focus on energy cost of daily activities in a more natural setting.

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References

1. Figoni S. Spinal cord disabilities: paraplegia and tetraplegia. In: Durstine JL, Moore GE, editors. ACSM's Exercise Management for Persons with Chronic Diseases and Disabilities. 2nd edn. Champaign, IL: Human Kinetics 2002; p 247–253.
2. Botto LD, Moore CA, Khoury MJ, Erickson JD. Neural-tube defects. N Engl J Med 1999; 341: 1509–1519.
3. 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: 420–427.
4. Bandini LG, Schoeller DA, Fukagawa NK, Wykes LJ, Dietz WH. Body composition and energy expenditure in adolescents with cerebral palsy or myelodysplasia. Pediatr Res 1991; 29: 70–77.
5. Van den Berg-Emons HJ, Bussmann JB, Brobbel AS, Roebroeck ME, van Meeteren J, Stam HJ. Everyday physical activity in adolescents and young adults with meningomyelocele as measured with a novel activity monitor. J Pediatr 2001; 139: 880–886.
6. Van den Berg-Emons HJ, Bussmann JB, Meyerink HJ, Roebroeck ME, Stam HJ. Body fat, fitness and level of everyday physical activity in adolescents and young adults with meningomyelocele. J Rehabil Med 2003; 35: 271–275.
7. Stewart AL, Hays RD, Wells KB, Rogers WH, Spritzer KL, Greenfield S. Long-term functioning and well-being outcomes associated with physical activity and exercise in patients with chronic conditions in the Medical Outcomes Study. J Clin Epidemiol 1994; 47: 719–730.

8. Crespo CJ. Exercise and the prevention of chronic disabling illness. In: Dawson DM, Slovik DM, editors. Exercise in Rehabilitation Medicine. Champaign, IL: Human Kinetics 1999; p 151–172.
9. Duffy CM, Hill AE, Cosgrove AP, Corry IS, Graham HK. Energy consumption in children with spina bifida and cerebral palsy: a comparative study. Dev Med Child Neurol 1996; 38: 238–243.
10. Bare A, Vankoski SJ, Dias L, Danduran M, Boas S. Independent ambulators with high sacral myelomeningocele: the relation between walking kinematics and energy consumption. Dev Med Child Neurol 2001; 43: 16–21.
11. Winninck JP, Short FX. The physical fitness of youngsters with spinal neuromuscular condition. Adapt Phys Act Q 1984; 1: 297–314.
12. Agre JC, Findley TW, McNally MC, Habeck R, Leon AS, Stradel L, Birkebak R, Schmalz R. Physical activity capacity in children with myelomeningocele. Arch Phys Med Rehabil 1987; 68: 372–377.
13. Sherman MS, Kaplan JM, Effgen S, Campbell D, Dold F. Pulmonary dysfunction and reduced exercise capacity in patients with myelomeningocele. J Pediatr 1997; 131: 413–418.
14. Williams LO, Anderson AD, Campbell J, Thomas L, Feiwell E, Walker JM. Energy cost of walking and of wheelchair propulsion by children with myelodysplasia: comparison with normal children. Dev Med Child Neurol 1983; 25: 617–624.
15. Findley TW, Agre JC. Ambulation in the adolescent with spina bifida. II. Oxygen cost of mobility. Arch Phys Med Rehabil 1988; 69: 855–861.
16. 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.
17. Bhambhani YN, Holland LJ, Steadward RD. Maximal aerobic power in cerebral palsied wheelchair athletes: validity and reliability. Arch Phys Med Rehabil 1992; 73: 246–252.
18. Bar-Or O. Pediatric Sports Medicine for the Practitioner. From Physiologic Principles to Clinical Applications. New York: Springer Verlag 1983; p 315–341.
19. Astrand PO, Rodahl K. Evaluation of physical performance on the basis of tests. In: Astrand PO, Rodahl K, editors. Textbook of Work Physiology: Physiological Bases of Exercise. Singapore: McGraw-Hill Book Co. 1986; p 354–390.
20. Ainsworth BE, Haskell WL, Whitt MC, Irwin ML, Swartz AM, Strath SJ, O'Brien WL, Bassett DR, Jr., Schmitz KH, Emplaincourt PO, et al. Compendium of Physical Activities: an update of activity codes and MET intensities. Med Sci Sports Exerc 2000; 32: S498–504.
21. Hoffman MD. Cardiorespiratory fitness and training in quadriplegics and paraplegics. Sports Med 1986; 3: 312–330.
22. Bartonek A, Saraste H, Samuelsson L, Skoog M. Ambulation in patients with myelomeningocele: a 12-year follow-up. J Pediatr Orthop 1999; 19: 202–206.
23. Asher M, Olson J. Factors affecting the ambulatory status of patients with spina bifida cystica. J Bone Joint Surg Am 1983; 65: 350–356.
24. Macsween A. The reliability and validity of the Astrand nomogram and linear extrapolation for deriving VO_2max from submaximal exercise data. J Sports Med Phys Fitness 2001; 41: 312–317.
25. Cink RE, Thomas TR. Validity of the Astrand-Ryhming nomogram for predicting maximal oxygen intake. Br J Sports Med 1981; 15: 182–185.

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Chapter 5

Sport Participation in Adolescents and Young Adults with Myelomeningocele and its Role in Total Physical Activity Behaviour and Fitness

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Abstract

Objective: To assess sports participation in young adults with myelomeningocele and its association with personal, disease-related and psychosocial factors, physical activity and fitness.
Design: Cross-sectional study.
Subjects: Fifty-one persons (26 males) with myelomeningocele aged 21.1 (standard deviation 4.5) years.
Methods: We assessed self-reported sports participation, ambulatory status, presence of hydrocephalus, functional independence, social support, perceived competence, exercise enjoyment, objective and self-reported physical activity, peak oxygen uptake, muscle strength and body fat. Associations were studied using regression analyses.
Results: Thirty-five subjects (69%) participated in sports. Sports participation was not associated with disease-related characteristics, but was associated with social support from family, perceived athletic competence and physical appearance ($p \leq 0.05$), and tended to be associated with global self-worth ($p = 0.10$). Sports participants had higher self-reported physical activity levels than non-participants ($p \leq 0.05$); objective results did not support this. Furthermore, they tended to be less likely to have subnormal muscle strength (odds ratio= 0.26; $p = 0.08$) and their peak oxygen uptake was 0.19 l/min higher but not statistically significant ($p = 0.13$).
Conclusion: Sports participation seems to be a due to personal preferences rather than physical ability, it could benefit from improving social support and perceived competence, and is associated with higher self-reported physical activity.

Introduction

In people with a disability, there is a shift from disability prevention to prevention of secondary conditions.[1] This is emphasized by the *Healthy People 2010* report, which contains a separate chapter on improving health of persons with disabilities that includes specific goals and objectives for increasing physical activity and fitness.[2] In the disabled population, optimizing physical activity may be even more important than in the general population, as disabilities commonly cause a cycle of deconditioning, in which physical functioning deteriorates, leading to further reduction in physical activity levels.[3] Furthermore, regular physical activity, sports participation and active recreation are important for disease prevention and the maintenance of functional independence, aerobic capacity, participation, social integration, and life satisfaction.[4]

Several barriers for engaging in sport activities have been recognized in people with a variety of disabilities, i.e. lack of transportation and accessible facilities, lack of equipment suited to their needs, lack of knowledge of where to obtain a programme, inability to perform physical activities, lack of time, money and social support, as well as poor self-efficacy and motivation.[5-7] With the increased emphasis on healthy lifestyles, there is a strong need to understand physical activity of various subgroups of the disabled population. Furthermore, in the context of physical activity promotion, insight into modifiable determinants is warranted, since these can be targeted in interventions.

Myelomeningocele (MMC) is a common neural tube defect with an incidence of 1:1000 live births. MMC is often associated with paralysis and loss of sensation in the lower limbs, incontinence and hydrocephalus.[8] Many persons with MMC now survive into adulthood, which raises the importance of a healthy lifestyle in this population. Previous research showed that adolescents and young adults with MMC have an inactive lifestyle compared to their able-bodied peers.[9,10] These studies have investigated daily physical activity by assessing total energy expenditure or total duration of dynamic activities (e.g. walking, cycling, wheelchair-driving, general non-cyclic movement); however, they did not specifically describe sports participation. Sports and exercise can be seen as a subcategory of physical activity which is planned, structured and repetitive.[11] Sports-related physical activities can easily be provided through rehabilitation services and could increase physical activity behaviour in persons with MMC. Furthermore, participating in sports may be important to improve their fitness, which was low in many.[12]

Due to the lack of knowledge on sports participation in people with MMC, the present study aimed to investigate sports participation and its association with disease-related, personal and social factors in adolescents and young adults with MMC. Furthermore, this study focussed on how sports participation might influence total physical activity behaviour and fitness.

Methods

Study sample

We recruited adolescents and young adults with MMC, aged between 16 and 30 years, from 4 university hospitals in the Western part of the Netherlands (Rotterdam, Leiden, Utrecht and Amsterdam) and all rehabilitation centres in the Southwest of the Netherlands. Exclusion criteria were complete dependence on an electric wheelchair, presence of disorders other than MMC that affect daily physical activity (e.g. rheumatoid arthritis), and presence of disorders that contra-indicate a maximal exercise test (e.g. exercise induced ischemia or arrhythmias, uncontrolled hypertension and exercise limitation due to chronic obstructive pulmonary disease). We invited 171 persons of whom 51 participated in the study (30%). The main reasons for non-participation were no interest, lack of time, or duration of the measurements. We found no differences between participants and non-participants regarding age, gender, level of lesion and presence of hydrocephalus as assessed by independent *t*-test or χ^2 test.[10] All participants and parents of adolescents aged less than 18 years gave written informed consent before participating in the study. The medical ethics committee of the Erasmus MC Rotterdam, and of all other participating institutes approved the study.

In total, 26 males and 25 females participated. Information about the neurological level of lesion was obtained from the medical records; they were registered as sacral (*n*= 7), lumbosacral (*n*= 21), lumbar (*n*= 15), thoracolumbar (*n*= 7) and thoracic (*n*= 1).

Sports participation

The main outcome measure was self-reported current sports participation (yes/no). We also registered reasons for sports participation or non-participation, and among sports participants we assessed the kind of sport and average self-reported number of hours spent on each sport per week.[13] Each sport was allocated to an intensity category using the physical activity compendium[14] using 4 intensity categories according to metabolic equivalents (METs: 1 MET = 4.184 kJ/kg body weight/h): < 3 METs, 3-6 METs, 6-9 METs and 9-12 METs. Activities in these categories were given intensity scores of 1.5, 4.5, 7.5 and 10.5 METs, respectively.[13] From these intensity scores and from the average amount of time per week spent on each sport we calculated a total sport score expressed in kJ/kg body weight/day for each sports participant.

Correlates of sports participation

Personal factors

Educational level. This was categorized as low (pre-vocational practical education or lower level, *n*= 17) or high (secondary theoretical educational and higher education, *n*= 34).

Disease-related factors

Ambulatory status was defined according to the classification of Hoffer.[15] Participants were categorized as ambulatory (community ambulators, *n*= 15, or household ambulators,*n*= 8) or non-ambulatory (non-(functional) ambulators, *n*= 28).

Hydrocephalus. The presence of hydrocephalus was obtained from medical records. Hydrocephalus was considered to be present when a shunt was placed (82%). For 2 participants, hydrocephalus was mentioned in the medical record at some time, but no shunt was placed. This was assumed to be only minor hydrocephalus and these participants were categorized as not having hydrocephalus.[8]

Functional independence. This was assessed in a semi-structured interview based on the functional independence measure FIM™ and functional assessment measure (FAM)[16]: In total, 30 items were assessed addressing 18 FIM™ items on functioning in basic physical and cognitive abilities and 12 FAM items on cognitive and psychosocial functioning. The items are divided into 7 subscales, i.e. self-care, sphincter control, transfers, locomotion, communication, psychosocial adjustment and cognitive functioning. Each item is scored on a 7-level scale from 1 (total dependence) to 7 (total independence). Total motor score is the sum of a person's scores on the first 4 subscales and total cognitive score includes the last 3 subscales. We used these respective scores as indicator of motor and cognitive functioning. The FIM™ + FAM has shown to have high internal consistency, Cronbach's alpha = 0.99, and good reliability.[17,18]

Psychosocial factors

Social support. The Dutch version of the scales for measuring Social Support for Diet and Exercise Behaviors by Sallis and co-workers[19] was used to measure social support from family and friends. The Dutch scale consists of three subscales: (i) the exercising together subscale from the Friend Support for Exercise Habits Scale (5 items), (ii) the participation and involvement subscale (10 items) and (iii) the rewards and punishment subscale (3 items). The latter two subscales are both from the Family Support for Exercise Habits scale.[20] Items were rated on a 5-point scale (1= 'never' to 5= 'very often') and possible sum scores ranged from 18 to 90. Respective Cronbach alpha values for the subscales were 0.69, 0.71 and 0.26, indicating sufficient internal consistency for the first two scales but low internal consistency for the latter, which is most likely due to the small number of items.[20]

Perceived competence. The Dutch version of Harter's Social Perception Profile for Adolescents scale (SPPA) was used to assess perceived competences and global self worth.[21,22] The Dutch SPPA is a 35-item instrument covering seven subscales (5 items per subscale), i.e. scholastic competence, social acceptance, athletic competence, physical appearance, behavioural conduct, close friendship and global self-worth. Since the subscales job competence and romantic appeal of the original SPPA were not applicable for Dutch adolescents, these were not included in the Dutch version.[22] Items are scored on a 4-point scale, with a higher score corresponding to a more positive perception of a specific competence. Scale scores were calculated by summing the items (maximum score 20). The Dutch SPPA has shown good internal consistency, test-retest reliability and construct validity in able-bodied young adults.[22]

Enjoyment. Enjoyment in exercise was measured with the Groningen Enjoyment Questionnaire (GEQ).[23] This consists of 10 statements with which subjects were asked to rate their agreement on a 7-point scale (1= 'absolutely disagree' to 7= 'absolutely agree'). Possible sum scores ranged from 10 to 70. Cronbach alpha for this scale was 0.88, indicating sufficient internal consistency.[23]

Physical activity

Objective dynamic activity. The duration of dynamic activities (composite measure of the separately detected activities walking, including walking stairs and running, cycling, general movement and wheelchair-driving) was measured objectively during 2 randomly selected weekdays using an accelerometry-based activity monitor (AM, Temec Instruments, Kerkrade, the Netherlands). Participants were fitted with the AM at their homes and instructed to perform their usual daily activities, but not to swim, or take a shower or bath during activity monitoring. To avoid measurement bias, the principles of the AM were explained to the participants after the measurements. All participants agreed with this procedure. Detailed methods are described elsewhere.[10] Since no differences existed between the first and the second measurement day, we used average results, expressed in min/day, for further analyses. The AM has shown to be a valid and reliable instrument to quantify mobility-related activities, including wheelchair driving.[24,25]

Self-reported physical activity. Self-reported physical activity was assessed by interview using the physical activity scale for individuals with physical disabilities (PASIPD) to obtain insight in the composition of total physical activity behaviour.[26] The Dutch version of the PASIPD is a 12 item, seven day recall questionnaire which consists of questions on leisure time, household and work related physical activities (Cronbach's alpha = 0.60), from which a total physical activity score in kJ/kg body weight/day was calculated. We also presented duration of total physical activity and of separate types of activities presented in min/day. A previous study reported acceptable test-retest reliability and criterion validity which was comparable to other questionnaires.[27]

Physical fitness

Aerobic capacity. This was measured during a progressive maximal exercise test, based on the McMaster All-Out Progressive Continuous Cycling and Arm test.[28] 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) or an arm ergometer (Jaeger ER800SH; Jaeger Toennies, Breda, the Netherlands), respectively. Detailed descriptions of the test can be found elsewhere.[12] We defined aerobic capacity as the mean oxygen uptake during the last 30 seconds of exercise (peakVO_2, in l/min).

Body fat. Thickness of 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 of the 4 skin-folds (in mm) was used as indicator of body fat.

Muscle strength. The strength of hip flexors and knee extensors was measured in persons whose main mode of ambulation was walking, and the strength of shoulder abductors and elbow extensors in persons whose main mode of ambulation was wheelchair driving, using the "break" testing method and a hand-held dynamometer (Microfet, Hoggan Health Industries Inc. West Jordan, USA). The positions and performance of the measurements were according to Van der Ploeg and co-workers.[29] The average value of 3 repetitions of the dominant side was used for further analyses. Detailed descriptions of the testing method can be found elsewhere.[12] To assess whether muscle strength was subnormal, values were normalized to Z-scores using reference values of healthy males and females.[30,31] A person's lowest Z-score of upper or lower extremity was used as indicator of muscle strength, dichotomizing it into normal (Z-score > -2) or subnormal (Z-score ≤ -2) muscle strength.

Statistical analyses

Data on sports participation were presented as mean (standard deviation SD) for the total group and for subgroups regarding gender and ambulatory status. Logistic regression analyses were performed to study correlates of sports participation and linear regression analyses to study whether participating in sports was associated with higher levels of physical activity and physical fitness. Gender and ambulatory status were potential confounders adjusted for in all multivariable analyses. Due to high collinearity between type of ergometer and ambulatory status, ambulatory status was used as a proxy measure for type of ergometer.[12] Odds ratios (OR) were presented for the logistic regressions and regression coefficients (RC) for the linear regressions, including 95% confidence intervals. p-values ≤ 0.05 were considered significant and p= 0.05-0.10 as a trend.

Results

Sports participation

Results on sports participation are presented in Table 1. Thirty-five (69%) adolescents and young adults participated in sports, for an average of 30 (SD 24) min/day, which on average accounted for 3.3 (SD 2.5) kJ/kg/day. The top 5 sports were hand biking, (cardio) fitness, swimming, wheelchair basketball, and wheelchair tennis. The majority of the sports participants mentioned fun and improving fitness as main reasons for participating in sports. Other reasons were to improve health, for social contacts or cosmetic reasons. Lack of time and lack of interest were the most frequently reported reasons for non-participation; others mentioned lack of energy, lack of transport, physical inability, not having a sports mate or fear of being criticized.

Table 1 Descriptive results for sports participation of adolescents and young adults with myelomeningocele.

	Total group (*n*= 51)	**Males** (*n*= 26)	**Females** (*n*= 25)	**Ambulators** (*n*= 23)	**Non-ambulators** (*n*= 28)
Sport, *n* (%)	35 (69)	21 (81)*	14 (56)*	15 (65)	20 (71)
Time spent on sports for doers† (min/day), mean (SD)	30 (24)	32 (27)	27 (17)	31 (29)	29 (20)
Total sport score for doers† (kJ/kg/day), mean (SD)	3.3 (2.5)	3.5 (2.8)	3.1 (2.0)	3.2 (2.6)	3.4 (2.4)

*Difference between males and females, $p= 0.08$. †Persons reporting participation in sports. SD= standard deviation.

Table 2 Association between sports participation and personal, disease-related, and psychosocial factors.

Independent variable	**Dependent variable**		**Logistic regression analyses**	
	Sports (*n*= 35)	No Sports (*n*= 16)		
Personal and disease-related factors	***n* (%)**	***n* (%)**	**OR (95% CI)**	***p*-value**
Gender, male	21 (60)	5 (31)	0.30 (0.09, 1.06)	0.06†
Ambulators	15 (43)	8 (50)	0.75 (0.23, 2.46)	0.64
Hydrocephalus	26 (77)	15 (94)	0.16 (0.02, 1.54)[1]	0.11
Educational level, low	12 (34)	5 (31)	0.93 (0.24, 3.53)[1]	0.91
	mean (SD)	**mean (SD)**	**OR[1] (95% CI)**	***p*-value**
Age	20.8 (4.8)	21.8 (4.1)	0.97 (0.86, 1.13)	0.84
Functional independence				
Motor functioning	6.03 (0.95)	6.21 (0.49)	0.81 (0.31, 2.09)	0.66
Cognitive functioning	6.43 (0.70)	6.58 (0.49)	0.65 (0.19, 2.18)	0.49
Psychosocial factors				
Social support				
Friends	11.7 (4.3)	8.8 (4.5)	1.17 (0.97, 1.40)	0.09†
Family participation	24.4 (7.0)	19.4 (7.1)	1.12 (1.00, 1.25)	0.05*
Family rewarding	13.2 (1.3)	11.5 (2.9)	2.12 (1.16, 3.86)	0.01*
Perceived competence				
Scholastic competence	12.9 (3.0)	11.7 (3.6)	1.08 (0.88, 1.33)	0.47
Social acceptance	15.2 (2.9)	14.4 (3.2)	1.05 (0.85, 1.31)	0.66
Athletic competence	13.5 (3.4)	8.4 (3.3)	1.47 (1.17, 1.86)	0.001*
Physical appearance	13.5 (3.3)	10.4 (4.3)	1.24 (1.02, 1.50)	0.03*
Behavioural conduct	15.4 (2.9)	15.2 (3.7)	1.07 (0.87, 1.33)	0.51
Close friendships	16.3 (3.6)	15.6 (3.3)	1.05 (0.88, 1.26)	0.60
Global self-worth	16.0 (2.9)	13.9 (3.9)	1.20 (0.98, 1.48)	0.10†
Enjoyment	56.8 (10.9)	49.4 (13.3)	1.05 (0.99, 1.11)	0.10†

[1]Adjusted for gender and ambulatory status. *$p \leq 0.05$. †p=0.05-0.10. OR= odds ratio. CI= confidence interval. SD= standard deviation.

Correlates of sports participation

Table 2 shows correlates of sports participation. Females tended to be less likely to participate in sports than males (OR= 0.30, *p*= 0.06). Ambulatory status, the presence of hydrocephalus, educational level, age, and motor and cognitive functioning were not related to sports participation.

People who received more social support from family were more likely to participate in sports (OR= 1.12 to 2.12, *p*≤ 0.05). In addition, those who received social support from friends (OR= 1.17, *p*= 0.09) and those who reported higher enjoyment of exercise (OR= 1.05, *p*= 0.10) tended to be more likely to participate in sports. Furthermore, sports participation was associated with athletic competence (OR= 1.47, *p*= 0.001) and physical appearance (OR= 1.24, *p*= 0.03), and appeared to have some association with global self-worth (OR= 1.20, *p*= 0.10).

Table 3 Association between sport participation and physical activity and physical fitness parameters.

Dependent variable	Sports (*n*= 35)	No Sports (*n*= 16)	Linear regression analyses	
	Mean (SD)	**Mean (SD)**	**RC (95% CI)[1]**	***p*-value**
Objective physical activity, AM				
Dynamic activity (min/day)	84 (65)	74 (57)	12 (-23, 47)	0.49
Self-reported physical activity, PASIPD				
Total score (kJ/kg/day)	58.6 (37.5)	29.7 (23.6)	29.6 (7.7, 51.6)	0.009*
Total duration (min/day)	285 (191)	179 (141)	115 (0.6, 230)	0.05*
Leisure activity (min/day)				
Non-exercise related walk/wheel	122 (84)	73 (83)	56 (4, 109)	0.04*
Light exercise	16 (50)	6 (19)	4 (-23, 32)	0.75
Moderate+vigorous+strength exercise	44 (45)	3 (7)	39 (14, 63)	0.002*
Household activity (min/day)	41 (67)	46 (59)	5 (-36, 45)	0.82
Occupational activity (min/day)	61 (67)	51 (96)	11 (-51, 74)	0.72
Physical fitness				
PeakVO_2 (l/min)	1.58 (0.53)	1.27 (0.50)	0.19 (-0.06, 0.44)	0.13
Sum 4 skin-folds (mm)	74.0 (38.6)	75.5 (39.4)	7.78 (-11.3, 26.9)	0.42
	% subnormal	**% subnormal**	**OR (95% CI)[1]**	***p*-value**
Muscle strength, normal (0) versus subnormal (1)	52	81	0.26 (0.06, 1.15)	0.08†

[1]Adjusted for gender and ambulatory status. *$p \leq 0.05$. †p=0.05-0.10. SD= standard deviation. RC= linear regression coefficient. OR= odds ratio. CI= confidence interval.

Influence on physical activity behaviour and fitness

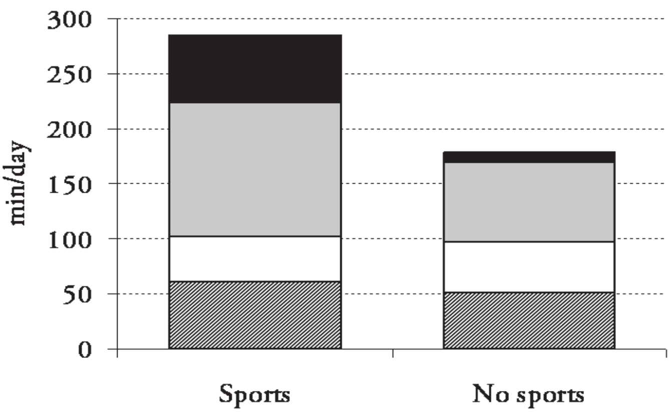
Table 3 presents results on physical activity and physical fitness and the association with sports participation. The 2-day objective measurements with the AM showed that participants spent on average 84 (SD 65) minutes on dynamic activities per day. According to the AM, after adjusting for gender and ambulatory status, sports participants spent 12 min longer on dynamic activities during the day than persons who did not participate in sports; however, this difference was not significant (p= 0.49).

Self-reported physical activity recalled over the previous 7 days with the PASIPD, showed that sport participants spent on average 285 (SD 191) minutes combined in leisure, household and occupational activities, corresponding with an average total score of 58.6 (SD 37.5) kJ/kg/day. Results of the PASIPD showed that persons who engaged in sports were more physically active during the day, as indicated by total score (p= 0.009) and by total duration of physical activities expressed in min per day (p= 0.05). In addition, besides spending on average 39 minutes more on sports and exercise per day (p= 0.002), they also reported 56 minutes more on non-exercise related walking or wheelchair driving (p= 0.04).

Fig. 1 shows the contribution of occupational tasks, household tasks, sports and exercise, and non-exercise related walking or wheelchair driving to total self-reported physical activity as measured with the PASIPD. In sports participants, the contribution of sports and exercise to total physical activity was 16%. Time spent on household and occupational activities did not differ between persons who participated in sports and those who did not (Fig. 1; Table 3).

Average peakVO_2 was 0.19 l/min higher in persons who participated in sports compared to those who did not; however this difference was not significant (p= 0.13). The percentage of participants with subnormal muscle strength tended to be lower in those who participated in sports (52%) compared to those who did not (81%; OR= 0.26, p= 0.08). Sports participation was not associated with body fat.

Figure 1 Contribution of sports to total physical activity behaviour as measured with the PASIPD.



Black: sports and exercise. Grey: non-exercise related walking or wheeling. White: household taks. Dashed: occupational tasks.

Discussion

Sports and physical activity behaviour

The present study contributes to the understanding of sport behaviour in adolescents and young adults with MMC. Two-third of the participants reported to be engaged in sports, which accounted for 30 min each day on average. Comparably, 6 out of 10 able-bodied persons aged 18-24 years participate in sports in the Netherlands.[32] Of people with various physical disabilities (including persons after stroke, back disorders, neurological disorders, chronic pain and orthopaedic disorders), 53% participated in sports one year after rehabilitation.[33] Although the proportion of adolescents and young adults with MMC participating in sports was not lower than in people with other physical disabilities, the estimated energy expended per day due to self-reported sports activities, as indicated by total sport score, was almost half as low; 3.3 kJ/kg/day in people with MMC vs 6.3 kJ/kg/day in people with other physical disabilities.[33] It is unclear whether this difference is due to less time spent on sports or due to lower intensity sports. In addition, self-reported levels of physical activity, as measured with the PASIPD were considerably lower than those mentioned in persons with a variety of physical disabilities: average PASIPD scores reported by Washburn and co-workers[26] and by Van der Ploeg and co-workers[33] – these were 84.5 kJ/kg/day and 63.7 kJ/kg/day respectively, whereas the current study showed an average score of 58.6 kJ/kg/day in sport participants and 29.7 kJ/kg/day in non-participants. This is in line with result from previous studies reporting that persons with MMC have inactive lifestyles.[9,10]

Correlates of sports participation

Results showed that ambulatory status, the presence of hydrocephalus and functional independence were not related to sports participation. This is an important contribution to knowledge on physical activity behaviour in persons with MMC. In addition to lack of time and lack of interest being the most frequently mentioned barriers for sports participation, it supports the view that engaging in sports seems to be due to personal preferences rather than physical ability. Therefore, persons with MMC should be encouraged to become more physically active, and sports participation may be a way to accomplish this. Rehabilitation centres may provide a good setting to increase physical activity levels.[13,33]

A large proportion of persons with MMC show cognitive impairments of some sort,[34] which may influence sports participation. In the present study, however, sports participation was not associated with educational level or cognitive functioning. Nevertheless, impairments in executive functioning rather than general intelligence might be associated with physical activity levels.[35] Future studies are necessary to investigate the association between sports participation and cognitive impairments.

In the context of promoting physical activity behaviour, modifiable environmental and personal determinants, are the most interesting targets in interventions. In able-bodied adults, the association between social support and physical activity behaviours is inconclusive.[36] However, the present results support the view that social support is important for adolescents and young adults with MMC to participate in sports. This study did not include other environmental influences,[7] however in 113 adults with mobility impairments, Kinne and co-workers found that exercise maintenance was not significantly associated with environmental barriers, whereas motivational barriers and self efficacy were.[5] In contrast, other studies reported lack of transportation and accessibility facilities to be important barriers to exercise.[6,7] Future studies should provide insights in the role of such environmental barriers to sports participation and physical activity in general in persons with MMC. In line with results of Kinne and co-workers,[5] the present study showed that adolescents and young adults with MMC who perceived higher athletic competence were more likely to participate in sports than those who perceived lower athletic competence. In addition, sports participation was associated with perceived physical appearance and tended to be associated with global self worth. Due to the cross-sectional study design, it is unclear whether persons with higher athletic competence, perceived physical appearance and global self-worth are more likely to participate in sports or whether participating in sports results in higher perceived competence for these aspects.

Furthermore, the present study showed that persons who perceived higher enjoyment during sports tended to be more likely to participate in sports. This is in line with previous results in persons with spinal cord injuries, that enjoyment was an important reason for participating in sports.[37] Therefore, interventions aiming to increase physical activity levels should be tailored individually ensuring that each person finds a type of physical activity they enjoy.

Sports participation and physical activity

The present study found some evidence that persons who participated in sports had more active lifestyles than those who did not, regardless of gender and ambulatory status. Sports participation contributed to 16% of the total daily physical activity in persons who participated in sports. This is an important contribution to total physical activity behaviour because apparently, non-participants do not seem to compensate with other physical activities, such as household and occupational. In addition, those who participated in sports reported that they also spent more time on non-exercise-related walking or wheeling. Objective measurements of physical activity with the AM pointed in the same direction, i.e. persons who participated in sports spent on average 12 minutes more on dynamic activities, but it was not significantly different. In addition, there was a substantial difference in the total duration of physical activity measured with both instruments; the AM registered an average duration of 84 minutes per day whereas the PASIPD reported 285 minutes per day. Both measures have limitations, which may have contributed to the discrepancy in results. Questionnaires are prone to overestimation of physical activity levels due to social desirability and recall difficulties.[38] Furthermore, objective measurements with the AM covered 2 consecutive weekdays, whereas the self-reported PASIPD was a 7 day-recall, also including weekend-days. Although in this study the measurement days overlapped, the PASIPD covered a period more representative for usual physical activity patterns and probably recorded usual activities that may be missed by the AM, especially in the weekend. Moreover, both instruments measure somewhat different aspects of physical activity. The AM measures mobility-related activities whereas the PASIPD includes several physical activity domains, i.e. leisure, household and occupational activities, of which not all are necessarily mobility-related and therefore not detected with the AM. We therefore consider both measures to be complementary using the PASIPD to describe the contribution of specific activity domains to total physical activity behaviour.

Sports participation and physical fitness

Although the present study failed to show significant correlations, data showed higher values of peakVO_2 and higher muscle strength in sports participants. Fitness is known to be an independent risk factor for lifestyle related diseases, but low values in persons with MMC are reported.[12] Therefore, increasing sport participation may be important to increase and maintain health of persons with MMC. The correlation between sports participation and fitness has been found previously in persons with spinal cord injuries.[39] Due to the relatively small sample sizes in the present study, correlations may have failed to show significances. There did not seem to be a correlation between sport participation and body fat levels.

Considering that the primary mode of ambulation elicits the highest values of peakVO_2 we used different exercise modes to assess aerobic capacity for ambulatory and non-ambulatory persons. Arm and leg exercise are known to have different physiological responses. In the analyses, we adjusted for ambulatory status as proxy measure for type of ergometer.[12]

Study limitations

Some limitations should be taken into account when interpreting the results of the study. The response rate of the study was low, which may have led to selection bias with people who were more interested in physical activity being more likely to participate in the study. This may have resulted in the high proportion of sports participation. However, it is unlikely that this selection bias has influenced the relevant correlates of sports participation as well as its relation with fitness. Another limitation was the relatively small sample size and low statistical power, which might have led to overlooking some of the relevant relationships.

Furthermore, both the sport score and the PASIPD score were based on MET values derived from the general population. It is possible that MET values for the same activity differ between the general population and the persons with MMC. Consequently, the absolute values of the sport score and the PASIPD score might not be completely accurate, and comparability with the general population may be limited. However, during several daily activities, oxygen uptake did not differ between persons with MMC and aged-matched peers, although physical strain was higher in persons with MMC.[40]

In conclusion, sports participation seems to be due to personal preferences rather than physical ability, it could benefit from social support (particular from family) and perceived competence, and is associated with higher self-reported physical activity. Conclusive evidence is needed on whether sports participation improves aerobic capacity. Improving physical activity behaviour and fitness in persons with MMC is important for health and increasing sports participation may contribute.

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References

1. Rimmer JH. Health promotion for people with disabilities: the emerging paradigm shift from disability prevention to prevention of secondary conditions. Phys Ther 1999; 79: 495-502.
2. United States Department of Health and Human Services. Healthy People 2010: Understanding and improving health. 2nd ed. Washington DC: US Government Printing Office, Nov 2000. Available from: http://www.health.gov/healthypeople.
3. Cooper RA, Quatrano LA, Axelson PW, Harlan W, Stineman M, Franklin B et al. Research on physical activity and health among people with disabilities: a consensus statement. J Rehabil Res Dev 1999; 36: 142-154.
4. Heath GW, Fentem PH. Physical activity among persons with disabilities--a public health perspective. Exerc Sport Sci Rev 1997; 25: 195-234.
5. Kinne S, Patrick DL, Maher EJ. Correlates of exercise maintenance among people with mobility impairments. Disabil Rehabil 1999; 21: 15-22.
6. Rimmer JH, Riley B, Wang E, Rauworth A, Jurkowski J. Physical activity participation among persons with disabilities: barriers and facilitators. Am J Prev Med 2004; 26: 419-425.
7. Ellis R, Kosma M, Cardinal BJ, Bauer JJ, McCubbin JA. Physical activity beliefs and behaviour of adults with physical disabilities. Disabil Rehabil 2007; 29: 1221-1227.

8. 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: 420-427.
9. Bandini LG, Schoeller DA, Fukagawa NK, Wykes LJ, Dietz WH. Body composition and energy expenditure in adolescents with cerebral palsy or myelodysplasia. Pediatr Res 1991; 29: 70-77.
10. 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.
11. Caspersen CJ, Powell KE, Christenson GM. Physical activity, exercise, and physical fitness: definitions and distinctions for health-related research. Public Health Rep 1985; 100: 126-131.
12. 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: 181-188.
13. Van der Ploeg HP, Streppel KR, van der Beek AJ, van der Woude LH, Vollenbroek-Hutten MM, van Harten WH et al. Counselling increases physical activity behaviour nine weeks after rehabilitation. Br J Sports Med 2006; 40: 223-229.
14. Ainsworth BE, Haskell WL, Whitt MC, Irwin ML, Swartz AM, Strath SJ et al. Compendium of physical activities: an update of activity codes and MET intensities. Med Sci Sports Exerc 2000; 32: S498-504.
15. 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.
16. Hall KM, Hamilton BB, Gordon WA, Zasler ND. Characteristics and comparisons of functional assessment indices: Disability Rating Scale, Functional Independence Measure and Functional Assessment Measure. J Head Trauma Rehabil 1993; 8: 60-74.
17. McPherson KM, Pentland B, Cudmore SF, Prescott RJ. An inter-rater reliability study of the Functional Assessment Measure (FIM+FAM). Disabil Rehabil 1996; 18: 341-347.
18. Hawley CA, Taylor R, Hellawell DJ, Pentland B. Use of the functional assessment measure (FIM+FAM) in head injury rehabilitation: a psychometric analysis. J Neurol Neurosurg Psychiatry 1999; 67: 749-754.
19. Sallis JF, Grossman RM, Pinski RB, Patterson TL, Nader PR. The development of scales to measure social support for diet and exercise behaviors. Prev Med 1987; 16: 825-836.
20. Stevens M, Bakker van Dijk A, de Greef MH, Lemmink KA, Rispens P. A Dutch version of the Social Support for Exercise Behaviors Scale. Percept Mot Skills 2000; 90: 771-774.
21. Harter S. Manual for the Self-Perception Profile for Adolescents. Denver, CO: University of Denver; 1988.
22. Treffers PDA, Goedhart AW, van den Bergh BRH, Veerman JW, Ackaert L, de Rycke L. Competentie Belevingsschaal voor Adolescenten (Dutch edition of the Self-perception Profile for Adolescents). Lisse: Hartcourt Test Publishers; 2002.
23. Stevens M, Moget P, de Greef MH, Lemmink KA, Rispens P. The Groningen Enjoyment Questionnaire: a measure of enjoyment in leisure-time physical activity. Percept Mot Skills 2000; 90: 601-604.
24. 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: 349-356.

25. 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: 550-557.
26. Washburn RA, Zhu W, McAuley E, Frogley M, Figoni SF. The physical activity scale for individuals with physical disabilities: development and evaluation. Arch Phys Med Rehabil 2002; 83: 193-200.
27. Van der Ploeg HP, Streppel KR, van der Beek AJ, van der Woude LH, Vollenbroek-Hutten M, van Mechelen W. The Physical Activity Scale for Individuals with Physical Disabilities: test-retest reliability and comparison with an accelerometer. J Phys Act Health 2007; 4: 96-100.
28. Bar-Or O. Pediatric sports medicine for the practioner. From physiologic principles to clinical applications. New York: Springer Verlag; 1983.
29. Van der Ploeg RJ, Fidler V, Oosterhuis HJ. Hand-held myometry: reference values. J Neurol Neurosurg Psychiatry 1991; 54: 244-247.
30. Bohannon RW. Reference values for extremity muscle strength obtained by hand-held dynamometry from adults aged 20 to 79 years. Arch Phys Med Rehabil 1997; 78: 26-32.
31. Phillips BA, Lo SK, Mastaglia FL. Muscle force measured using "break" testing with a hand-held myometer in normal subjects aged 20 to 69 years. Arch Phys Med Rehabil 2000; 81: 653-661.
32. Centraal Bureau voor de Statistiek. Jeugd 2003, cijfers en feiten [Statistics Netherlands. Youth 2003, numbers and facts]. Voorburg/Heerlen: Centraal Bureau voor de Statistiek; 2003 (in Dutch).
33. Van der Ploeg HP, Streppel KR, van der Beek AJ, van der Woude LH, Vollenbroek-Hutten MM, van Harten WH et al. Successfully improving physical activity behavior after rehabilitation. Am J Health Promot 2007; 21: 153-159.
34. Barf HA, Verhoef M, Jennekens-Schinkel A, Post MW, Gooskens RH, Prevo AJ. Cognitive status of young adults with spina bifida. Dev Med Child Neurol 2003; 45: 813-820.
35. Roebroeck ME, Hempenius L, van Baalen B, Hendriksen JG, van den Berg-Emons HJ, Stam HJ. Cognitive functioning of adolescents and young adults with meningomyelocele and level of everyday physical activity. Disabil Rehabil 2006; 28: 1237-1242.
36. Prochaska JJ, Rodgers MW, Sallis JF. Association of parent and peer support with adolescent physical activity. Res Q Exerc Sport 2002; 73: 206-210.
37. Wu SK, Williams T. Factors influencing sport participation among athletes with spinal cord injury. Med Sci Sports Exerc 2001; 33: 177-182.
38. Sallis JF, Saelens BE. Assessment of physical activity by self-report: status, limitations, and future directions. Res Q Exerc Sport 2000; 71: S1-14.
39. Dallmeijer AJ, Hopman MT, van As HH, van der Woude LH. Physical capacity and physical strain in persons with tetraplegia; the role of sport activity. Spinal Cord 1996; 34: 729-735.
40. Bruinings AL, van den Berg-Emons HJ, Buffart LM, van der Heijden-Maessen HC, Roebroeck ME, Stam HJ. Energy cost and physical strain of daily activities in adolescents and young adults with myelomeningocele. Dev Med Child Neurol 2007; 49: 672-677.

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Chapter 6

Lifestyle, Participation and Health-Related Quality of Life in Adolescents and Young Adults with Myelomeningocele

Submitted for Publication

Abstract

We aimed to study relationships between lifestyle-related factors, participation and health-related quality of life (HRQoL) in adolescents and young adults with myelomeningocele (MMC).

Fifty-one persons aged 21.1 ± 4.5 years participated in this cross-sectional study. Participation was assessed with the Life Habits questionnaire and HRQoL with the Short Form 36. We assessed physical activity using an accelerometry-based activity monitor, fitness (peak oxygen uptake) during a maximal exercise test, and the sum of 4 skin-folds as indicator of body fat. Relationships were studied using logistic regression analyses.

Of the participants, 63% perceived 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 perceived less difficulty in daily activities ($p \leq 0.05$) and a higher physical HRQoL ($p \leq 0.05$). Persons with higher fitness tended to perceive less difficulty in social roles (OR= 5.3; $p = 0.08$). Body fat was not related with participation and HRQoL.

In conclusion, higher levels of physical activity and fitness are related with less difficulty in participation and higher HRQoL. Therefore, promoting healthy lifestyles, by improving physical activity and aerobic fitness, deserves attention in health care in persons with MMC.

Introduction

The increased life expectancy of people who are born with a physical disability such as myelomeningocele (MMC) raises new challenges in providing optimal health care and preventing secondary conditions. Many adolescents and young adults with MMC have medical problems such as urinary and faecal incontinence, reduced joint mobility, pressure sores, orthopaedic problems, i.e. scoliosis and contractures,[1] and many are physically inactive, 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 functioning and health in people with physical disabilities such as MMC.

Functioning and health can objectively be studied by assessing participation in society, whereas health-related quality of life (HRQoL) is a subjective or self-perceived outcome.[3] Participation can be evaluated by assessing life habits, i.e. activities of daily living and social roles, addressing difficulty of performance and the assistance required.[4] Many adolescents and young adults with MMC are independent in most functional activities but report dependency for sphincter control, locomotion and self-care,[5,6] particularly those with hydrocephalus and higher lesion levels.[6] Furthermore, many adolescents with MMC showed limited responsibilities in household tasks, decision making, money management, and engagement in activities with peers.[5]

HRQoL refers to an individual's perception of various aspects of his or her life that are affected by a specific condition. Results on HRQoL of adolescents and young adults with MMC are equivocal. Some studies showed that they perceived lower levels of HRQoL compared to the able-bodied population, particularly regarding aspects of physical functioning, including self-care, continence and mobility,[7,8] but also on emotional, social and school domains.[8] Other studies showed overall good HRQoL.[9,10]

Little information is available on whether improving lifestyle may benefit participation and HRQoL in adolescents and young adults with MMC. Studies in persons with other chronic conditions showed that physical activity and aerobic fitness are associated with participation and HRQoL.[11] In able-bodied adolescents and young adults, obesity was associated with lower HRQoL, but no differences were found in HRQoL between obese and non-obese persons with MMC.[8]

The present study assessed participation and HRQoL in a sample of adolescents and young adults with MMC. To study whether lifestyle interventions have the potential to improve participation and HRQoL in persons with MMC, we explored whether physical activity, aerobic fitness and body fat were associated with participation and HRQoL.

Methods

Participants

Fifty-one adolescents and young adults (aged 21.1 ± 4.5 years) participated in the study of whom 26 (51%) were male. Recruitment of participants is described elsewhere.[2] All participants and parents of adolescents aged less than 18 years gave written informed consent before participating in the study. The Medical Ethics Committee of Erasmus MC and of all participating institutes approved the study.

Information regarding lesion level and presence of hydrocephalus was obtained from medical records. Lesion levels were registered as sacral (*n*= 7), lumbosacral (*n*= 21), lumbar (*n*= 15), thoraco-lumbar (*n*= 7) and thoracic (*n*= 1). Hydrocephalus was considered to be present when a shunt was placed (82%).

According to the classification of Hoffer,[12] 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, *n*= 17) or high (secondary theoretical education and higher education, *n*= 34).

Participation

Functioning in daily activities and social roles was assessed using the Life Habits Questionnaire (LIFE-H).[13] The short version, the LIFE-H 3.0 consists of 69 life habits covering 12 categories: nutrition, fitness, personal care, communication, housing, mobility, responsibilities, interpersonal relationships, community life, education, employment and recreation. The first 6 categories refer to daily activities whereas the others are associated with social roles. The scoring is based on two specific elements: (i) the degree of difficulty in performing life habits (no difficulty, with difficulty, with substitution, or not accomplished); and (ii) 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 – 9, with 0 indicating total handicap (the activity or social role is not accomplished or achieved) and 9 indicating optimal participation (the activity is performed without difficulty and without help). Mean scores for the two sub-domains (i.e. daily activities and social roles) were calculated and a mean score below 8 was used to indicate difficulty in performance.[14]

Health-related quality of life

The Medical Outcomes Study 36-item Short-form Health survey (SF-36)[15,16] 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: a physical component summary (PCS) and a mental component summary (MCS). Higher domain and summary scores indicate higher levels of functioning or well-being.[15]

Lifestyle-related factors

Physical activity

We objectively measured the duration of dynamic activities (composite measure of the separately detected activities walking, including walking stairs and running, cycling, general movement and wheelchair-driving) during 2 consecutive weekdays using an accelerometry-based activity monitor (AM, Vitaport 3, Temec Instruments, Kerkrade, the Netherlands). The AM has shown to be a valid and reliable instrument to quantify mobility-related activities, including wheelchair-driving.[17,18] Participants were fitted with the AM at their homes and 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 of the first and second measurement day expressed in min/day.

Aerobic fitness

Aerobic fitness was measured during a progressive maximal exercise test, based on the McMaster All-Out Progressive Continuous Cycling and Arm test. According to Bhambhani and co-workers who studied persons with cerebral palsy, the main mode of ambulation elicits the highest oxygen uptake.[19] Therefore, 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) or arm ergometer (Jaeger ER800SH; Jaeger Toennies, Breda, The Netherlands) respectively. Detailed descriptions of the test can be found elsewhere.[20] Aerobic fitness was defined as the mean oxygen uptake during the final 30 seconds of exercise (peakVO_2, in l/min).

Body fat

Thickness of 4 skin-folds (biceps, triceps, subscapular, suprailiac) was measured twice on the right side of the body with a calliper (Harpenden, Burgess Hill, UK) and average sum (in mm) was used as indicator of body fat.

Statistical analyses

Results were expressed as mean and standard deviation (SD) for the total group. In addition, group percentages of adolescents and young adults with difficulties in daily activities and social roles (score < 8) were presented. Using a one sample t-test, we compared scores of the 8 domains of the SF-36 with the average scores of a Dutch reference population aged 16-40 years[16] and with persons with spina bifida aperta and hydrocephalus who participated in a national study on Adolescents with SPina Bifida in the Netherlands (ASPINE).[7] PCS and MCS were compared with a Dutch reference sample aged 20-29 years.[21] For all analyses, p-values ≤ 0.05 were considered significant and 0.05< p≤ 0.10 were reported as a trend.

Because data on participation and HRQoL were not normally distributed among our study sample, we dichotomized the outcome measures. For the sub-domains daily activities and social roles of the LIFE-H, individuals were classified as either perceiving no difficulties (1) or as perceiving difficulties (0). For the PCS and MCS of the SF-36, individuals were classified as high HRQoL (1) or low HRQoL (0), based on the median score (44.0 for PCS and 57.8 for MCS).

Using binary logistic regression analyses we built separate models to study whether daily physical activity, aerobic fitness and body fat were related to participation and HRQoL, and whether participation was related to HRQoL. The following variables were checked for potential confounding: age, gender, ambulatory status and level of education. Gender (0= female; 1= male) and ambulatory status (0= non-ambulator; 1= ambulator) were adjusted for in the analyses. In the relationships with aerobic fitness, ambulatory status was used as proxy for type of ergometer because of the high collinearity between the two variables.[20] Odds ratios (OR), including 95% confidence intervals (95% CI), and explained variances (R^2, Nagelkerke R square) were presented for relationships reporting significances or trends.

Results

Participants spent on average 81 ± 62 minutes on physical activities per day, the average peakVO_2 was 1.48 ± 0.52 l/min and average sum of 4 skin-folds was 74.4 ± 38.5 mm.

Table 1 Participation of adolescents and young adults with MMC, as measured with the Life-H.

Life-H	Mean ± SD	Perceived difficulties (%)
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

Participation

Overall, 63% of the participants perceived difficulties in daily activities, mainly in personal care, housing and mobility, and 59% perceived difficulties in social roles, and in particular with recreation and employment (Table 1). Several personal and condition-related characteristics were related to participation (Table 3). Males perceived less difficulty during daily activities than females (OR= 7.1; p= 0.004). Ambulators perceived less difficulty in daily activities (OR= 11.3; p< 0.001) and social roles (OR= 4.7; p< 0.001) than non-ambulators. Adjusted for gender and ambulatory status, individuals with a higher education level tended to perceive less difficulty with daily activities (OR= 4.1; p= 0.09) and perceived less difficulty with social roles (OR= 5.0; p=0.03).

Table 2 Health-related quality of life of adolescents and young adults with MMC and reference populations, as measured with the SF-36.

Domain SF-36	Total group	Dutch reference sample[16,21]		ASPINE[7]	
	Mean ± SD	Mean	*p*-value	Mean	*p*-value
Physical functioning	46.8 ± 23.7	93.1	< 0.001	34.4	0.001
Role physical	77.8 ± 34.1	86.4	0.08	68.9	0.06
Bodily pain	84.8 ± 20.1	80.9	0.18	75.6	0.002
General health	69.2 ± 23.7	78.2	0.01	64.9	0.20
Vitality	66.4 ± 15.9	70.7	0.06	64.3	0.36
Social functioning	81.1 ± 24.8	87.8	0.06	81.8	0.85
Role emotional	85.0 ± 29.3	85.4	0.92	85.2	0.96
Mental health	77.3 ± 16.7	78.7	0.54	76.8	0.85
Physical component summary	43.1 ± 9.0	53.2	< 0.001		
Mental component summary	55.0 ± 9.7	47.8	< 0.001		

Health-related quality of life

Average PCS was 43.1 (range 24.0 to 58.9) and average MCS was 55.0 (range 24.8 to 68.8), see Table 2. Adolescents and young adults with MMC perceived lower physical HRQoL (p< 0.001) than a Dutch reference sample, particularly on the domains physical functioning (p< 0.001) and general health (p= 0.01). They generally perceived higher mental HRQoL (p< 0.001) than a Dutch reference sample, but vitality and social functioning tended to be lower in persons with MMC (0.05< p≤ 0.10). Compared to persons with spina bifida aperta and hydrocephalus of the ASPINE study,[7] the participants of the present study perceived better physical functioning (p= 0.001) and less bodily pain (p= 0.002).

Males perceived a higher physical HRQoL than females (OR= 3.4; p= 0.04) and ambulators perceived a higher physical HRQoL than non-ambulators (OR= 4.1; p= 0.02), see Table 4. Adjusted for gender and ambulatory status, younger persons tended to perceive a higher physical HRQoL (OR= 0.87; p= 0.06) and perceived a higher mental HRQoL (OR= 0.86; p= 0.04).

Table 3 Relationships between personal, condition- and lifestyle-related factors and participation in adolescents and young adults with MMC.

	Daily Activities				Social roles			
	Difficulty (n= 32)	No difficulty (n= 19)	Logistic regression OR (95% CI)	R^2	Difficulty (n= 30)	No difficulty (n= 21)	Logistic regression OR (95% CI)	R^2
Personal and condition-related factors								
Univariate regression								-
Gender, *n* (%) male	11 (37)	15 (79)	7.1* (1.9, 26.9)	0.24	14 (47)	12 (57)	1.5 (0.5, 4.7)	0.17
Ambulatory status, *n* (%) non-ambulatory	24 (75)	5 (21)	11.3* (2.9, 43.9)	0.34	21 (70)	7 (33)	4.7* (1.4, 15.4)	
Multivariate regression[a]								
Education, *n* (%) low	14 (44)	3 (16)	4.1† (0.8, 24.4)	0.55	14 (47)	3 (14)	5.0* 91.1, 22.3)	0.29
Age, mean ± SD years	21.1 ± 4.7	21.1 ± 4.2	1.07 (0.89, 1.28)	-	20.3 ± 4.5	22.3 ± 4.4	1.13 (0.97, 1.33)	-
Lifestyle-related factors								
Physical Activity, mean ± SD min/day	56.8 ± 34.4	122.1 ± 77.4	8.8* (1.4, 54.2)[b]	0.61	61.8 ± 35.5	108.3 ± 81.2	2.2 (0.8, 5.6)[b]	-
Aerobic Fitness, (peakVO_2), mean ± SD l/min	1.21 ± 0.39	1.93 ± 0.42	29.7* (1.7, 506.7)	0.67	1.32 ± 0.43	1.72 ± 0.59	5.3† (0.8, 5.6)[b]	0.28
Body fat (sum of 4 skin-folds), mean ± SD mm	85.0 ± 37.6	57.7 ± 34.4	1.00 (0.97, 1.03)	-	78.3 ± 36.6	68.8 ± 41.3	1.00 (0.98, 1.03)	-

*$p \leq 0.05$. †$0.05 < p \leq 0.10$. OR= Odds ratio. CI= confidence interval. R^2= Nagelkerke R-square, represents explained variance. [a]Separate regression models for each factor, adjusted for gender and ambulatory status. [b]In the analyses, physical activity is expressed in hours per day.

Table 4 Relationships between personal, condition- and lifestyle-related factors, participation and health-related quality of life in adolescents and young adults with MMC.

	Physical component summary				Mental component summary			
	Low	High	Logistic regression		Low	High	Logistic regression	
	(*n*= 25)	(*n*= 26)	OR (95% CI)	R^2	(*n*= 25)	(*n*= 26)	OR (95% CI)	R^2
Personal and condition-related factors								
Univariate regression								
Gender, *n* (%) male	9 (36)	17 (65)	4.3* (1.1, 10.6)	0.11	12 (48)	14 (54)	1.1 (0.4, 3.3)	-
Ambulatory status, *n* (%) non-ambulatory	18 (72)	10 (39)	4.1* (1.2, 13.4)	0.15	11 (44)	17 (65)	0.5 (0.2, 1.5)	-
Multivariate regression[a]								
Education, *n* (%) low	8 (32)	9 (35)	0.7 (0.2, 2.7)	-	7 (28)	10 (39)	0.7 (0.2, 2.4)[a]	-
Age, mean ± SD years	22.4 ± 4.6	19.8 ± 4.1	0.87† (0.75, 1.00)	0.30	22.5 ± 4.6	19.7 ± 4.1	0.86* (0.74, 1.00)	0.18
Lifestyle-related factors								
Physical Activity, mean ± SD min/day	54.4 ± 33.4	108.2 ± 73.4	4.8 (1.3, 17.8)[b]	0.40	83.0 ± 74.9	78.6 ± 48.6	1.1 (0.6, 2.2)[b]	-
Aerobic Fitness, (peakVO_2), mean ± SD l/min	1.17 ± 0.39	1.78 ± 0.48	30.2* (2.7, 342.1)	0.47	1.53 ± 0.59	1.44 ± 0.48	0.81 (0.2, 33.7)	-
Body fat (sum of 4 skin-folds), mean ± SD mm	82.6 ± 35.0	67.2 ± 40.6	1.00 (0.98, 10.3)	-	69.7 ± 32.9	78.6 ± 43.0	1.00 (0.99, 1.03)	-
Participation								
Daily activities, *n* (%) difficulty	24 (96)	8 (31)	59.0* (4.7, 740.2)	0.55	15 (60)	17 (56)	1.2 (0.3, 5.5)	-
Social roles, *n* (%) difficulty	19 (76)	11 (42)	3.3† (0.9, 12.6)	0.29	14 (56)	16 (62)	1.1 (0.3, 3.8)	-

*$p \leq 0.05$. †$0.05 < p \leq 0.10$. OR= Odds ratio. CI= confidence interval. R^2= Nagelkerke R-square, represents explained variance. [a]Separate regression models for each factor, adjusted for gender and ambulatory status. [b]In the analyses, physical activity is expressed in hours per day.

Lifestyle, participation and health-related quality of life

Relationships between lifestyle and participation are presented in Table 3, and Table 4 presents relationships with HRQoL. Adjusted for gender and ambulatory status, persons with higher levels of physical activity perceived less difficulty in daily activities (OR= 8.8; p= 0.02) and a higher physical HRQoL (OR= 4.8; p= 0.02). Persons with higher peakVO_2 perceived less difficulty in daily activities (OR= 29.7; p= 0.02), they tended to perceive less difficulty in social roles (OR= 5.3; p= 0.08) and perceived a higher physical HRQoL (OR= 30.2; p= 0.006). Lifestyle-related factors were not associated with mental HRQoL.

Persons who perceived no difficulties in daily activities (OR= 59.0; p= 0.002) and social roles (OR= 3.3; p= 0.08) appeared to perceive a higher physical HRQoL, but participation was not related to mental HRQoL.

Discussion

Participation

Results showed that many adolescents and young adults with MMC perceived difficulties in daily activities and social roles, particularly regarding self care, housing, mobility, recreation and employment. Therefore, improving participation is one of the challenges in rehabilitation care for people with chronic conditions such as MMC.

The finding that a large proportion of persons with MMC perceived difficulties in daily activities and social roles is in line with previous reports of dependency in various activities of daily living[5,6] and subnormal levels of decision making and participation.[5] Also, compared to adolescents and young adults with cerebral palsy (CP) with normal intelligence, persons with MMC appeared to perceive more difficulties in participation; 21% of persons with CP perceived difficulties in daily activities and 24% in social roles[14] compared with 63% and 59% in persons with MMC.

Non-ambulators and persons with a low education level perceived more difficulties in participation. Comparably, in adolescents and young adults with CP, limitations in participation were mainly attributable to restricted gross motor functioning and low education level.[14] Since both ambulatory status and education level are irreversible characteristics associated with the severity of MMC, it might be relevant to focus on modifiable lifestyle-related factors.

Lifestyle and participation

Lower levels of physical activity and aerobic fitness were associated with perceiving difficulties in daily activities. Levels of physical activity and aerobic fitness did not differ among persons with low and high education level, indicating that these lifestyle-related factors are important for participation regardless of education level, which was also related with participation. Studies in adults with spinal cord injury showed higher levels of physical activity to be associated with lower levels of disability severity and handicap.[22]

Our study does not support the suggestion of Manns et al.[22] that physical activity may play a more important role in participation than aerobic fitness. On the contrary, our results indicated that aerobic fitness was not only associated with daily activities but also tended to have a relationship with social roles, whereas physical activity did not. Possibly, increasing aerobic fitness may reduce physical strain and associated fatigue, which may result in increased performance in daily activities and social roles. However, due to the cross-sectional study design causality can not 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.

Health-related quality of life

Physical component

Adolescents and young adults with MMC perceived lower physical HRQoL compared to a Dutch reference sample, particularly regarding mobility, which confirms previous results.[7,8] The finding that non-ambulators were more likely to have lower physical HRQoL confirms previous results that persons with MMC with lower mobility and self-care ability perceived lower physical HRQoL.[23] On the other hand, they perceived higher physical HRQoL compared to adults with paraplegia, who had an average PCS of 29.[24] In contrast to persons with MMC, 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; participants of our study were younger. We found that older individuals with MMC tended to perceive lower HRQoL, which underlines the need for paying attention towards improving HRQoL when growing into adulthood.

Mental component

Compared to a Dutch reference sample, adolescents and young adults with MMC generally perceived higher mental HRQoL, but they tended to perceive lower vitality and social functioning. Results on the different domains of mental HRQoL were comparable to results of young adults with spina bifida aperta and hydrocephalus from the national ASPINE study.[7]

Mental health was associated with age, but not with ambulatory status, nor with participation. This is in contrast with the study of Padua et al.[23] in a small sample of adolescents with spina bifida, who found that people with higher mobility and self-care ability perceived higher psychological distress and more severe role disability due to emotional problems.

Lifestyle and health-related quality of life

Results showed that higher levels of physical activity and aerobic fitness were related with higher perceived physical HRQoL, which supports the evidence for a strong focus in health care on increasing physical activity and fitness to maintain a healthy adult life in persons with chronic conditions such as MMC. However, causality needs to be established in future study, since it may also be that participants were more physically active because they perceived a higher physical HRQoL.

We found no relationship between mental HRQoL and body fat which is in accordance with previous findings in persons with MMC, but in contrast with findings in the able-bodied population.[8] 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.[9,10]

Study limitations

A selection bias may have occurred that the more active and fitter people had higher interest in participating in this study. This may have resulted in the perceiving of higher levels of physical functioning and less pain compared to participants of the ASPINE study. The hypotheses 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.

The strength of the monitoring physical activity with the AM is that it provides detailed objective information on mobility-related activities in both ambulatory and non-ambulatory persons. 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 the relationships from the present study were reasonable.

Furthermore, the present study focused on 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.[25]

In conclusion, a large proportion of adolescents and young adults with MMC, in particular non-ambulators and females, perceived difficulties in participation and a low physical HRQoL. We found some evidence that lifestyle can be important for participation and HRQoL; higher levels of physical activity and fitness were related with less difficulty in participation and higher physical HRQoL. Therefore, promoting a healthy lifestyle, by improving physical activity and aerobic fitness deserves attention in health care in persons with MMC.

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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: 420-427.
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. Colver A, Group S. Study protocol: SPARCLE--a multi-centre European study of the relationship of environment to participation and quality of life in children with cerebral palsy. BMC Public Health 2006; 6: 105.
4. Noreau L, Fougeyrollas P. Long-term consequences of spinal cord injury on social participation: the occurrence of handicap situations. Disabil Rehabil 2000; 22: 170-180.
5. Buran CF, Sawin KJ, Brei TJ, Fastenau PS. Adolescents with myelomeningocele: activities, beliefs, expectations, and perceptions. Dev Med Child Neurol 2004; 46: 244-252.
6. 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: 114-119.
7. 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: 192-197.
8. 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: S112-118.
9. Kirpalani HM, Parkin PC, Willan AR, Fehlings DL, Rosenbaum PL, King D et al. Quality of life in spina bifida: importance of parental hope. Arch Dis Child 2000; 83: 293-297.
10. 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.
11. Tomas-Carus P, Hakkinen A, Gusi N, Leal A, Hakkinen K, Ortega-Alonso A. Aquatic Training and Detraining on Fitness and Quality of Life in Fibromyalgia. Med Sci Sports Exerc 2007; 39: 1044-1050.
12. 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.

13. Fougeyrollas P, Noreau L, St-Michel G. Life Habits measure - shortened version (LIFE-H 3.0). Lac St-Charles, Quebec, Canada: COCIDIH; 2001.
14. 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: 453-463.
15. Ware JE. SF-36 Physical and Mental Health Summary Scales: A User's Manual. Boston, MA: The Health Institute; 1994.
16. Aaronson NK, Muller M, Cohen PD, Essink-Bot ML, Fekkes M, Sanderman R 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: 1055-1068.
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: 349-356.
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: 550-557.
19. Bhambhani YN, Holland LJ, Steadward RD. Maximal aerobic power in cerebral palsied wheelchair athletes: validity and reliability. Arch Phys Med Rehabil 1992; 73: 246-252.
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: 181-188.
21. 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.
22. 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: 1566-1571.
23. 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: 1384-1388.
24. 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: 2200-2207.
25. 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: 458-463.

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Chapter 7

Cardiovascular Disease Risk Factors and the Relationships with Physical Activity, Aerobic Fitness, and Body Fat in Adolescents and Young Adults with Myelomeningocele

Arch Phys Med Rehabil. in press

Abstract

Objective: To describe cardiovascular disease (CVD) risk factors in adolescents and young adults with myelomeningocele (MMC) and to explore relationships with physical activity, aerobic fitness and body fat.
Design: Cross-sectional study.
Setting: Outpatient clinic.
Participants: Adolescents and young adults (*n*= 31) with MMC (58% males) aged 16 through 30 years; 13 were ambulatory and 18 were nonambulatory.
Interventions: Not applicable.
Main outcome measures: We studied biological and lifestyle-related CVD risk factors, including lipid and lipoprotein profiles, blood pressure, aerobic fitness (peakVO_2), body fat, daily physical activity and smoking behavior. We considered subjects at increased CVD risk when 2 or more of the following risk factors clustered: systolic blood pressure, total serum cholesterol (TC), high-density lipoprotein cholesterol (HDL-C) and cigarette smoking. Relationships were studied using regression analyses.
Results: Levels of TC, low-density lipoprotein cholesterol, and triglycerides were elevated in respectively 29%, 38% and 3% of the participants. HDL-C was reduced in 19%. Hypertension was found in 20% and 19% were current cigarette smokers. Based on the clustering of risk factors, 42% of the participants were at increased CVD risk; 15% of ambulatory participants and 61% of non-ambulatory (*P*= .03). Adjusted for sex and ambulatory status, individuals with higher aerobic fitness tended to be more likely to have no CVD risk (OR= 13.0; *P*= .07). CVD risk was not associated to physical activity and body fat.
Conclusions: A large proportion of the study sample was at CVD risk, indicated by clustering of risk factors. Improving aerobic fitness in young adults with MMC may contribute in reducing CVD risk; this needs to be confirmed in future studies.

Introduction

Lifestyle-related diseases such as cardiovascular diseases (CVD) are of major concern in industrialized countries.[1,2] Although the clinical manifestations of CVD typically appear in adulthood, the process of atherosclerosis, causing CVD, lies early in childhood[3,4] and seems to increase rapidly during adolescence and young adulthood.[5] During the transition from adolescence to adulthood, people develop their own lifestyle. It may therefore be important to encourage healthy lifestyle behavior at these ages with the aim of delaying the development of atherosclerosis and reducing the incidence of CVD later in life.

The Framingham Heart Study has substantially contributed to the understanding of the causes of CVD and has identified numerous risk factors, such as cigarette smoking, hypertension, high total serum cholesterol (TC), and low levels of high-density lipoprotein cholesterol (HDL-C).[1,6-8] It has been suggested to focus on multiple risk factors rather than on one specific risk factor because the severity of atherosclerosis increases as the number of CVD risk factors increases.[4] This clustering of risk factors has proved to be a better measure of cardiovascular health in youth than single risk factors.[9]

Other important risk factors for CVD are physical inactivity, obesity and low aerobic fitness[6,7,10-12] which can play a major role in the prevention of CVD.[6] Persons with chronic physical conditions may be at increased risk of developing CVD. Unfavorable CVD risk profiles in persons with a spinal cord injury (SCI) have been associated with inactive lifestyles and low aerobic fitness.[13-15] Furthermore, CVD is among the most important causes of death in persons with SCI,[16-18] occurring with a greater frequency compared to the general population.[19] Also persons with myelomeningocele (MMC) have inactive lifestyles, low aerobic fitness and excessive body fat.[20-24] Because they may be restricted in physical activity from birth, the risk of developing CVD may even be higher compared to persons with a SCI. Furthermore, deteriorating vascular properties in the lower extremities (small diameter, low flow, high shear stress), which are related to CVD, tend to be more pronounced in persons with MMC compared to persons with SCI.[25] Since many persons with MMC nowadays survive into adulthood,[26] CVD may be of increasing concern.

In contrast to the SCI population, no data are available on the number of deaths due to CVD in persons with MMC, and information on CVD risk factors other than physical inactivity, poor aerobic fitness and obesity is limited. Rendeli et al[27] found that girls with MMC (aged 1-16 y) had higher levels of TC and very low density lipoprotein compared to their peers, and TC was higher in nonambulatory than in ambulatory girls. In contrast, Nelson et al[28] did not find differences in lipid and lipoprotein profiles between adolescents with MMC and able-bodied peers. Because of the scarcity of studies, the present study aimed to describe CVD risk factors in adolescents and young adults with MMC, including ambulators and nonambulators. To study whether improving lifestyles might benefit cardiovascular health, we explored whether physical activity, aerobic fitness and body fat were related to other CVD risk factors.

Methods

Participants

Recruitment

Adolescents and young adults with MMC (aged 16-30 y) from 4 university hospitals in the western part of the Netherlands (Rotterdam, Leiden, Utrecht and Amsterdam) and 5 rehabilitation centers in the adjacent region were invited to participate in a cross-sectional study on physical activity and aerobic fitness. Exclusion criteria were complete dependence on an electric wheelchair, inability to understand the instructions necessary for the study, presence of disorders other than MMC that affect daily physical activity (e.g. rheumatoid arthritis), and presence of disorders that contra-indicate a maximal exercise test (e.g. exercise-induced ischemia or arrhythmias, uncontrolled hypertension and exercise limitation due to chronic obstructive pulmonary disease). Inclusion criterion was willingness to undergo blood tests. In total, 31 adolescents and young adults participated in the present study, which were a subset of a larger cross-sectional study (*n*= 51) on physical activity and fitness in adolescents and young adults with MMC.[24,29] All participants gave written informed consent. The Medical Ethics Committee of Erasmus MC and of all participating institutes approved the study.

Characteristics

Mean age of the participants was 21.4 ± 4.4 y and 18 (58%) were male. We obtained level of lesion from medical records. We defined ambulatory status according to the classification of Hoffer et al[30] and categorized participants as ambulators (community ambulators, *n*= 8, or household ambulators, *n*= 5) or nonambulators (*n*= 18, also including non-functional ambulators).[30]

Personal and condition-related characteristics (i.e. age, sex, level of lesion and presence of hydrocephalus), ambulatory status, blood pressure, sum of 4 skin-folds, aerobic fitness, daily physical activity and smoking behavior did not differ between those who were willing to yield a blood sample (*n*= 31) and those who were not (*n*= 20), as tested with an independent *t* test or chi square test (table 1). Furthermore, we found no differences in personal and disease-related characteristics of responders (*n*= 51) and nonresponders of the larger cross-sectional study (*n*= 108).[24]

Table 1 Descriptive results of personal and condition-related characteristics, and biological and lifestyle-related risk factors of cardiovascular disease of responders and nonresponders of this study.

	Responders (n= 31)	**Nonresponders** (n= 20)	
Personal and condition-related characteristics	***n*** **(%)**	***n*** **(%)**	***P***
Gender (female)	13 (42)	10 (50)	.77
Ambulatory status (non-ambulator)	18 (58)	10 (50)	.78
Lesion level			.37
Sacral	2 (7)	5 (25)	
Lumbosacral	14 (45)	7 (35)	
Lumbar	10 (32)	5 (25)	
Thoraco-lumbar	4 (13)	3 (15)	
Thoracic	1 (3)	0 (0)	
Hydrocephalus	25 (81)	17 (89)	.50
	Mean ± SD	**Mean ± SD**	***P***
Age (years)	21.4 ± 4.4	20.6 ± 4.8	.55
Biological risk factors			
Blood lipid and lipoproteins			
Total cholesterol (mmol/l)	4.59 ± 0.94	NA	
High-density lipoprotein (mmol/l)	1.27 ± 0.25	NA	
Low-density lipoprotein (mmol/l)	2.96 ± 0.93	NA	
Triglycerides (mmol/l)	1.09 ± 0.32	NA	
Blood pressure			
Systolic blood pressure (mmHg)	123.9 ± 13.2*	122.0 ± 37.5	.55
Diastolic blood pressure (mmHg)	80.9 ± 8.6*	80.5 ± 5.6	.88
Body fat, sum of 4 skin-folds (mm)	75.4 ± 40.6*	72.4 ± 37.5*	.70
Aerobic fitness, peakVO_2 (l/min)	1.47 ± 0.51	1.51 ± 0.56	.79
Lifestyle-related risk factors			
Daily physical activity (min/day)	77.4 ± 72.1†	85.7 ± 45.5	.65
Smoking behavior (cig/day)	2.0 ± 4.9	0.6 ± 2.4	.25

Abbreviations: NA, not applicable. *n – 1. †n – 2.

Biological risk factors

Blood lipids and lipoproteins

Nonfasting venous blood samples of approximately 10 ml were drawn from the vena antecubitus with a Vacutainer needle[a] and collected into an evacuated serum separator tube II (SST II tube[a]) while subjects were seated. TC and triglycerides (TG) were determined using an enzymatic calorimetric test (CHOD-PAP and GPO-PAP[b]). HDL-C and low-density lipoprotein cholesterol (LDL-C) were determined directly using a homogeneous enzymatic calorimetric test with polyethylene glycol modified enzymes and dextran sulfate (Roche/Hitachi 747, 902[b]).

Blood pressure

We measured blood pressure with an indirect method while subjects were seated for at least 10 minutes prior to the measurement. A standard pressure cuff was placed around the upper arm. Systolic blood pressure (SBP) and diastolic blood pressure (DBP) were measured twice using a sphygmomanometer (Maxi-Stabil 3[c]), and the lowest values were recorded.

Aerobic fitness

We measured aerobic fitness during a progressive maximal exercise test, based on the McMaster All-Out Progressive Continuous Cycling and Arm test.[31] According to Bhambhani et al[32] who studied persons with cerebral palsy, the main mode of ambulation elicits the highest oxygen uptake. Therefore, 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[d]) or an arm ergometer (Jaeger ER800SH[d]) respectively. Detailed descriptions of the test can be found elsewhere.[29] We defined aerobic capacity as the mean oxygen uptake during the last 30 seconds of exercise (peakVO_2, in l/min).

Body fat

We measured thickness of 4 skin-folds (biceps, triceps, subscapular, suprailiac) twice on the right side of the body with a Harpenden caliper[e] and used the average sum (in mm) as indicator of body fat.

Lifestyle-related risk factors

Level of daily physical activity

We objectively measured the duration of dynamic activities (composite measure of the separately detected activities walking, including walking stairs and running, cycling, general non-cyclic movement and wheelchair-driving) during 2 consecutive weekdays using an accelerometry based activity monitor (AM[f]; size: 15×9×3.5 cm; weight: 500 g). The AM has been shown to be a valid and reliable instrument to quantify mobility-related activities, including wheelchair-driving[33,34] We fitted participants with the AM at their homes and instructed them to perform their usual daily activities, but to swim or take a shower or bath during activity monitoring. To avoid measurement bias, we explained the principles of the AM to the participants after the measurements. All participants agreed with this procedure. Detailed procedures are described elsewhere.[24] We defined the level of daily physical activity as the average duration of dynamic activities of the first and second measurement day, expressed in min/day.

Smoking behavior

For each participant, we recorded the number of cigarettes smoked per day and designated them as a smoker (1; ≥ 1 cig/d) or nonsmoker (0).

Statistical analyses

We expressed results as mean and standard deviation (SD) or frequencies. We categorized values of TC, HDL-C, LDL-C and TG from normal to high according to the third report of the National Cholesterol Education, Adult Treatment Panel (NCEP ATP III)[7,8] (cut-offs are presented in table 2). SBP and DBP were categorized according to the seventh report of the Joint National Committee on prevention, detection, evaluation and treatment of high blood pressure (JNC 7 Report)[35] (see table 2). We tested differences in the proportion of males and females with subnormal levels and differences between ambulators and non-ambulators using a chi square test. To test differences in physical activity, aerobic fitness and body fat among subgroups, we used a *t* test for independent samples.

We selected the risk factors included in the Framingham Risk Assessment[8] (i.e. TC, SBP, HDL and smoking) to assess the degree of clustering. It was thus possible for a participant to have between 0 and 4 risk factors. We defined clustering of risk to be present in persons with two or more risk factors[36] and considered them being at increased CVD risk. Differences in the prevalence of an increased CVD risk between males and females and between ambulators and non-ambulators were assessed using a chi square test.

We conducted separate regression analyses to model the following outcome measures: each single risk factor from the Framingham Risk Assessment and the presence (0) or absence (1) of CVD risk. For each outcome measure, we included the following independent variables (continuous data) one at the time: (1) physical activity, (2) aerobic fitness, and (3) body fat. Sex (0= male; 1= female) and ambulatory status (0= nonambulator; 1= ambulator) were confounders, adjusted for in all regression analyses. In the regression analyses between aerobic fitness and the CVD risk factors, we used ambulatory status as proxy for type of ergometer because of the high collinearity between the two variables.[29] Probability to enter a factor was set at P less or equal to .05, and the probability to remove was set at P greater than or equal to .10. We calculated standardized regression coefficients (RC) and odds ratios (OR) of the models and their 95% confidence intervals. For all tests, we considered P less than or equal to .05 significant and reported $.05 < P \leq .10$ as a trend.

Table 2 Descriptive results and frequencies of cardiovascular disease (CVD) risk factors in adolescents and young adults with myelomeningocele.

CVD Risk factor		Total Group (*n*= 31)	Ambulators (*n*= 13)	Nonambulators (*n*= 18)		Male (*n*= 18)	Female (*n*= 13)	
		Mean ± SD	**Mean ± SD**	**Mean ± SD**	***P***	**Mean ± SD**	**Mean ± SD**	***P***
Physical activity (min/day)		77.4 ± 72.1	121.0 ± 93.2	46.6 ± 26.5	.02	99.1 ± 83.9	46.6 ± 35.5	.05
Aerobic fitness (peakVO_2, l/min)		1.47 ± 0.51	1.62 ± 0.59	1.36 ± 0.45	.18	1.76 ± 0.47	1.07 ± 0.27	.001
Body fat (sum 4 skin-folds, mm)		75.4 ± 40.6	59.1 ± 33.4	88.6 ± 42.0	.05	50.7 ± 24.2	110.4 ± 32.6	< .001
	Cut-offs	**%**	**%**	**%**		**%**	**%**	
Total cholesterol[a]	mmol/l				.32			.30
Desirable	< 5.2	71	85	61		78	61	
Borderline	5.2 - 6.2	26	15	33		17	39	
High	≥ 6.2	3	0	6		6	0	
Low-density lipoprotein[a]	mmol/l				.58			.31
Optimal	< 2.59	39	39	39		44	31	
Near/above optimal	2.59 - 3.34	23	31	19		22	23	
Borderline high	3.37 - 4.12	29	31	28		17	46	
High	4.14 - 4.86	6	0	11		11	0	
Very High	≥ 4.92	3	0	5		6	0	
High-density lipoprotein[a]	mmol/l				.43			.19
High (no risk)	≥ 1.56	16	8	22		11	23	
In between		65	77	56		78	46	
Low (major risk)	< 1.04	19	15	22		11	31	

Table 2 continued.

CVD Risk factor		Total Group (*n*= 31)	Ambulators (*n*= 13)	Nonambulators (*n*= 18)		Male (*n*= 18)	Female (*n*= 13)	
	Cut-offs	%	%	%	***P***	%	%	***P***
Triglycerides*	mmol/l				.42			.42
Normal	< 1.69	97	92	100		100	92	
Borderline high	1.69 - 2.25	3	8	0		0	8	
High	2.26 - 5.63	0	0	0		0	0	
Very high	≥ 5.64	0	0	0		0	0	
Blood pressure†	SBP/DBP (mmHg)				.10			.20
Normal	< 120/80	17	33	6		6	31	
Prehypertension	120-139/80-89	63	42	78		64	61	
Stage 1 hypertension	140-159/90-99	17	25	11		24	8	
Stage II hypertension	≥ 160/100	3	0	6		6	0	
Current cigarette smoker	yes	19	15	22	.99	17	23	.68
Number of CVD risk factors‡					.03			.41
0		3	8	0		0	8	
1		55	77	39		61	46	
2	Increased risk	42	15	61		39	46	

*According to the third report of the National Cholesterol Education Program[8]. †According to the seventh report of the Joint National Committee on prevention, detection, evaluation and treatment of high blood pressure[35]. ‡Number of CVD risk factors based on Framingham Risk Assessment (including total cholesterol, high-density lipoprotein, systolic blood pressure, smoking).

Results

Single CVD risk factors

Table 2 presents descriptive results and frequencies of CVD risk factors. Nonambulators had lower levels of daily physical activity (*P*= .02) and a higher sum of 4 skin-folds (*P*= .05) compared to ambulators. Females had lower levels of daily physical activity (*P*= .05), lower aerobic fitness (*P*< .001) and a higher sum of 4 skin-folds (*P*< .001) compared to males.

Based on the cut-off points of the NCEP ATP III, levels of TC, LDL-C, and TG were elevated in respectively 29%, 38% and 3% of the participants. HDL-C was reduced in 19%. Based on the cut-off points of the JNC 7 Report, 20% of the participants were hypertensive. Of all participants, 19% were current cigarette smokers. We found no significant differences in the proportion of participants with an unfavorable lipid and lipoprotein profile and smoking behavior among subgroups regarding gender and ambulatory status. The proportion of persons with a normal blood pressure tended to be higher in ambulators than in nonambulators (*P*= .10).

Clustering of CVD risk factors

Increased CVD risk, as indicated by clustering of risk factors, was present in 42% of the participants (see table 2). A larger proportion of non-ambulatory persons with MMC had increased CVD risk compared to ambulatory (*P*= .03).

Relations between CVD risk factors and physical activity, fitness and body fat

Physical activity, aerobic fitness and sum of 4 skin-folds were not associated with TC, HDL or smoking after adjusting for gender and ambulatory status (table 3). Results showed that individuals with higher values of peakVO_2 had higher values of SBP (RC= 12.4; *P*= .02) and individuals with higher sum of 4 skin-folds tended to have higher SBP (RC= 1.52; *P*= .07). Furthermore, we found that individuals with higher peakVO_2 tended to be more likely to have no CVD risk as indicated by clustering of risk factors (OR= 13.0; *P*= .07). Levels of physical activity and sum of 4 skin-folds were not associated with CVD risk.

Table 3 Relationships between cardiovascular disease risk factors and physical activity, aerobic fitness (peakVO_2) and body fat (sum of 4 skin-folds).

	Single risk factors				Clustering of risk factors[b]				
	TC	HDL	SBP	Smoking	No risk	Risk			
Independent variable[a]	RC (95% CI)	RC (95% CI)	RC (95% CI)	RC (95% CI)	Mean ± SD	Mean ± SD	OR (95% CI)	R^2	*P*
Physical activity (min/day)	0.17 (-0.22 - 0.55)	0.01 (-0.09, 0.12)	-2.02 (-6.91, 2.87)	1.13 (0.37, 3.47)	91 ± 84	60 ± 52	0.94 (0.37, 2.38)[d]	0.29	.90
PeakVO_2 (l/min)	-0.14 (-1.06, 0.79)	0.01 (-0.24, 0.26)	12.4* (1.8, 23.0)	0.11 (0.01, 1.79)	1.60 ± 0.58	1.30 ± 0.39	13.0 (0.8, 204.8)	0.41	.07#
Sum of 4 skin-folds (mm)	-0.05 (-0.19, 0.09)	-0.02 (-0.06, 0.01)	1.52# (-0.12, 3.16)	1.12 (-0.12, 3.16)	75 ± 44	76 ± 37	1.13 (0.81, 1.58)[e]	0.46	.46

* $P \leq 0.05$. # $0.05 < P \leq 0.10$. [a]For each risk factor, the independent variables were included in the model one at the time, adjusted for gender (0= male, 1= female) and ambulatory status (0= non-ambulator, 1= ambulator). [b]0= increased risk, 1= no risk. [c]0= smoker, 1= non-smoker. [d]In the logistic regression analyses, physical activity is expressed in hours per day. [e]In the logistic regression analyses, the sum of 4 skin-folds is expressed in cm; TC= total cholesterol. HDL= high-density lipoprotein; SBP= systolic blood pressure; RC= regression coefficient. CI= confidence interval. OR= odds ratio. R^2= Nagelkerke Rsquare.

Discussion

CVD risk factors

The present study provides an overview of biological and lifestyle-related CVD risk factors in adolescents and young adults with MMC. We did not find differences in blood lipids and lipoproteins values between subgroups regarding gender and ambulatory status.

Average values of lipid and lipoproteins were comparable to those reported in the general male and female Dutch population of similar ages.[37] This is in line with results from Nelson et al[28] who did not find differences in lipid and lipoprotein profiles between persons with MMC and able-bodied peers. However, although average values seem normal, according to the NCEP ATP III guidelines, we found unfavorable TC, LDL-C and HDL-C in 29%, 38% and 19% of the participants, respectively. TC values 5.2 mmol/l or higher were reported in approximately 4% and 15% of the general Dutch male and female population aged 16-19 years, respectively.[37] We found that 24% of the males were hypertensive, which is somewhat higher than the 13% reported in the general Dutch male population aged 20 to 29 years.[37] The proportion of hypertensive females with MMC (7%) was comparable to the general Dutch female population aged 20 to 29 years (8%).[37] In addition, in our study a large proportion of adolescents and young adults with MMC was prehypertensive and therefore at increased risk for progression to hypertension.[35] According to the JNC 7 Report, people with prehypertension require health-promoting lifestyle modifications. In the general population, 10 kg weight loss can reduce SBP by 5-20 mmHg[35,38,39] and regular aerobic physical activity can reduce SBP by 4-9 mmHg.[35,40,41] Considering the high body fat and the poor levels of physical activity and aerobic fitness found in persons with MMC,[20,22-24,29] sufficient reductions in blood pressure might be achieved from counseling aimed at reducing body fat and increasing physical activity and aerobic fitness. However, the present results do not support the above hypotheses that also in persons with MMC, health promoting lifestyle modifications will reduce SBP. Unexpectedly, after adjusting for sex and ambulatory status, we found that higher SBP was associated with higher fitness. Possibly, also factors other than sex and ambulatory status may play a role in the relationship between SBP and fitness in persons with MMC, but the small sample size limited further adjustments for confounders. We therefore remain inconclusive about the relation between blood pressure and fitness and future studies in fitness training with close monitoring of blood pressure are warranted in persons with MMC.

CVD risk

It is assumed that clustering of risk factors is a better measure of CVD risk than single risk factors.[4,9] Results showed that clustering was present in a large proportion (42%) of participating adolescents and young adults with MMC, particularly in non-ambulators. Berenson et al[4] reported that 2 or more risk factors were present in 23% and Andersen et al[36] reported clustering of risk to be present in approximately 15% of able-bodied adolescents and young adults. Although the proportion with multiple risk factors seems higher in persons with MMC, it is difficult to draw conclusions due to the summing of different risk factors among studies.

The only factor that tended to be associated with the clustering of risk factors was aerobic fitness. This implies that increasing aerobic fitness may lower the CVD risk in adolescents and young adults with MMC. It is known that aerobic exercise training may lead to a 10 to 20% increase in peakVO_2.[42] Results showed that individuals are 13 times more likely to have no CVD risk with every liter increase in peakVO_2. Correspondingly, with every 220ml increase in peakVO_2 (which is an increase of approximately 15%), people with MMC are 1.8 times more likely to have no CVD risk; future longitudinal studies are needed to confirm this relationship. In the general population, improvements in aerobic fitness over time have been shown to improve the prognosis of developing CVD.[11,43]

The finding that aerobic fitness was related to the CVD risk, whereas physical activity was not, is in line with the general conclusion emerging from six longitudinal observational studies in the general population[44-49] in which aerobic fitness was found to be predictive for a healthy CVD risk profile at a later age, whereas physical activity was not.[50] In these longitudinal studies, physical activity was assessed by means of questionnaires, and therefore, measurements of fitness offered greater objectivity compared to physical activity, which might have contributed to this conclusion. In contrast, the present study objectively assessed both aerobic fitness and physical activity. Because we found no relation with physical activity, this might contribute to the evidence that aerobic fitness seems to be more directly associated with the quality of the cardiovascular system than physical activity.[50] A possible explanation might be that physical activity is, in contrast to aerobic fitness, a behavior; therefore, measurements of physical activity could be momentary whereas aerobic fitness is an attribute that people have or achieve[51] and thus more or less the result of an active lifestyle over time. The history and intensity of physical activity was not assessed in the present study, but they may play a role in the relation with CVD risk.

Although obesity is considered a CVD risk factor, we found no relation between body fat and CVD risk. In persons with MMC, besides ambulatory status, perhaps factors such as aerobic fitness are more important for determining CVD risk than body fat. Similarly, Myers et al[12] found that aerobic fitness was the most important predictor of CVD mortality in able-bodied adult men and that it was more powerful than other established risk factors.

Limitations of the study

The present study had some limitations. First, the small sample size may hamper the generalization of the results, as indicated by large confidence intervals. However, because personal and disease-related characteristics did not differ between participants and nonparticipants of the present study or between participants and nonparticipants of the larger cross-sectional study, we consider the results to be sufficiently representative. Furthermore, results will have to be interpreted with caution because the sample size was rather small with regard to the number of independent variables (n= 3) in the regression models.

Second, for practical reasons, we did not obtain a fasting blood sample for the measurements of blood lipids and lipoproteins, which may have influenced the values. However, there is evidence that differences between nonfasting and fasting levels of TC, HDL-C and LDL-C are clinically irrelevant[52-54] and the NCEP ATP III guidelines consider nonfasting values for TC and HDL-C to be appropriate.[7] Nonfasting TG levels must be interpreted carefully because they are significantly higher than fasting TG.[52,53] Since we did not include TG in the clustering of CVD risk factors, we assumed that the relationships between CVD risk and levels of physical activity, aerobic fitness and body fat were not influenced by the non-fasting blood samples.

Third, we used the presence of clustering of risk factors to determine whether an individual was at increased CVD risk but, unlike the Framingham Risk Assessment, the risk factors were not weighted. However, because the Framingham Risk Assessment is only validated for individuals aged 20 years and older, we were restricted to study unweighted clustering of risk factors. Considering that the purpose of this study was not to predict future CVD events, but to study whether levels of physical activity, aerobic fitness and body fat were associated with CVD risk, we consider our approach valid.

Finally, objective monitoring of physical activity with the AM was restricted to 2 days. The strength of the AM measurements is that it provides detailed objective information on mobility-related physical activities in both ambulatory and non-ambulatory persons. However, we did not characterize an individual's actual activity pattern which needs at least 7 days of activity monitoring.[55] Therefore we may have underestimated the relation between physical activity and CVD risk. In a previous study, White et al[56] found evidence supporting that a 24 hrs measurement with the AM is of adequate duration to assess activity level. 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= .89). Therefore, we assumed that a sample of 2 random days during the week was sufficient to adequately assess an individual's physical activity level.

Conclusions

We found that a large proportion of the sample of adolescents and young adults with MMC, particularly the nonambulators, was at increased CVD risk as indicated by the clustering of risk factors. Individuals with higher aerobic fitness tended to be more likely to have no CVD risk. Improving aerobic fitness in persons with MMC deserves attention in health care as it may contribute in reducing cardiovascular risk. This needs to be confirmed in future studies.

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Suppliers

[a]Becton, Dickinson & Co, 1 Becton Drive, Franklin Lakes, New Jersey, 07417, USA. [b]Roche Diagnostics GmbH, D-68298; Sandhofer str 116, 68305 Mannheim, Germany. [c]Speidel & Keller, Zollerstrasse 2-4, 72417 Jungingen, Germany. [d]Jaeger ER800, Jaeger Benelux BV, Nikkelstraat 2, 4823 AB Breda, The Netherlands. [e]British indicators ltd, Burgess Hill, West Sussex, UK. [f]Temec Instruments BV, Spekhofstraat 2, 6466 LZ Kerkrade, The Netherlands.

References

1. Kannel WB. Contributions of the Framingham Study to the conquest of coronary artery disease. Am J Cardiol 1988; 62: 1109-1112.
2. Sans S, Kesteloot H, Kromhout D. The burden of cardiovascular diseases mortality in Europe. Task Force of the European Society of Cardiology on Cardiovascular Mortality and Morbidity Statistics in Europe. Eur Heart J 1997; 18: 1231-1248.
3. Berenson GS, Wattigney WA, Tracy RE, Newman WP, 3rd, Srinivasan SR, Webber LS et al. Atherosclerosis of the aorta and coronary arteries and cardiovascular risk factors in persons aged 6 to 30 years and studied at necropsy (The Bogalusa Heart Study). Am J Cardiol 1992; 70: 851-858.
4. Berenson GS, Srinivasan SR, Bao W, Newman WP, 3rd, Tracy RE, Wattigney WA. Association between multiple cardiovascular risk factors and atherosclerosis in children and young adults. The Bogalusa Heart Study. N Engl J Med 1998; 338: 1650-1656.
5. Strong JP, Malcom GT, McMahan CA, Tracy RE, Newman WP, 3rd, Herderick EE et al. Prevalence and extent of atherosclerosis in adolescents and young adults: implications for prevention from the Pathobiological Determinants of Atherosclerosis in Youth Study. Jama 1999; 281: 727-735.
6. Grundy SM, Balady GJ, Criqui MH, Fletcher G, Greenland P, Hiratzka LF et al. Primary prevention of coronary heart disease: guidance from Framingham: a statement for healthcare professionals from the AHA Task Force on Risk Reduction. American Heart Association. Circulation 1998; 97: 1876-1887.

7. Executive Summary of The Third Report of The National Cholesterol Education Program (NCEP) Expert Panel on Detection, Evaluation, And Treatment of High Blood Cholesterol In Adults (Adult Treatment Panel III). Jama 2001; 285: 2486-2497.
8. National Cholesterol Education Program Expert Panel on Detection Evaluation and Treatment of High Blood Cholesterol in Adults. Third Report of the National Cholesterol Education Program (NCEP) Expert Panel on Detection, Evaluation, and Treatment of High Blood Cholesterol in Adults (Adult Treatment Panel III) final report. Circulation 2002; 106: 3143-3421.
9. Andersen LB, Wedderkopp N, Hansen HS, Cooper AR, Froberg K. Biological cardiovascular risk factors cluster in Danish children and adolescents: the European Youth Heart Study. Prev Med 2003; 37: 363-367.
10. Grundy SM, Balady GJ, Criqui MH, Fletcher G, Greenland P, Hiratzka LF et al. Guide to primary prevention of cardiovascular diseases. A statement for healthcare professionals from the Task Force on Risk Reduction. American Heart Association Science Advisory and Coordinating Committee. Circulation 1997; 95: 2329-2331.
11. Pate RR, Pratt M, Blair SN, Haskell WL, Macera CA, Bouchard C et al. Physical activity and public health. A recommendation from the Centers for Disease Control and Prevention and the American College of Sports Medicine. Jama 1995; 273: 402-407.
12. Myers J, Prakash M, Froelicher V, Do D, Partington S, Atwood JE. Exercise capacity and mortality among men referred for exercise testing. N Engl J Med 2002; 346: 793-801.
13. Janssen TW, van Oers CA, van Kamp GJ, TenVoorde BJ, van der Woude LH, Hollander AP. Coronary heart disease risk indicators, aerobic power, and physical activity in men with spinal cord injuries. Arch Phys Med Rehabil 1997; 78: 697-705.
14. Dallmeijer AJ, Hopman MT, van der Woude LH. Lipid, lipoprotein, and apolipoprotein profiles in active and sedentary men with tetraplegia. Arch Phys Med Rehabil 1997; 78: 1173-1176.
15. Dallmeijer AJ, van der Woude LH, van Kamp GJ, Hollander AP. Changes in lipid, lipoprotein and apolipoprotein profiles in persons with spinal cord injuries during the first 2 years post-injury. Spinal Cord 1999; 37: 96-102.
16. Lidal IB, Snekkevik H, Aamodt G, Hjeltnes N, Biering-Sorensen F, Stanghelle JK. Mortality after spinal cord injury in Norway. J Rehabil Med 2007; 39: 145-151.
17. DeVivo MJ, Krause JS, Lammertse DP. Recent trends in mortality and causes of death among persons with spinal cord injury. Arch Phys Med Rehabil 1999; 80: 1411-1419.
18. Frankel HL, Coll JR, Charlifue SW, Whiteneck GG, Gardner BP, Jamous MA et al. Long-term survival in spinal cord injury: a fifty year investigation. Spinal Cord 1998; 36: 266-274.
19. Whiteneck GG, Charlifue SW, Frankel HL, Fraser MH, Gardner BP, Gerhart KA et al. Mortality, morbidity, and psychosocial outcomes of persons spinal cord injured more than 20 years ago. Paraplegia 1992; 30: 617-630.
20. Roberts D, Shepherd RW, Shepherd K. Anthropometry and obesity in myelomeningocele. J Paediatr Child Health 1991; 27: 83-90.
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: 305-311.
22. Van den Berg-Emons HJ, Bussmann JB, Brobbel AS, Roebroeck ME, van Meeteren J, Stam HJ. Everyday physical activity in adolescents and young adults with meningomyelocele as measured with a novel activity monitor. J Pediatr 2001; 139: 880-886.

23. Van den Berg-Emons HJ, Bussmann JB, Meyerink HJ, Roebroeck ME, Stam HJ. Body fat, fitness and level of everyday physical activity in adolescents and young adults with meningomyelocele. J Rehabil Med 2003; 35: 271-275.
24. 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.
25. Boot CR, van Langen H, Hopman MT. Arterial vascular properties in individuals with spina bifida. Spinal Cord 2003; 41: 242-246.
26. Bowman RM, McLone DG, Grant JA, Tomita T, Ito JA. Spina bifida outcome: a 25-year prospective. Pediatr Neurosurg 2001; 34: 114-120.
27. Rendeli C, Castorina M, Ausili E, Girardi E, Fundaro C, Caldarelli M et al. Risk factors for atherogenesis in children with spina bifida. Childs Nerv Syst 2004; 20: 392-396.
28. Nelson MD, Widman LM, Abresch RT, Stanhope K, Havel PJ, Styne DM et al. Metabolic syndrome in adolescents with spinal cord dysfunction. J Spinal Cord Med 2007; 30: S127-139.
29. 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: 181-188.
30. 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.
31. Bar-Or O. Pediatric sports medicine for the practioner. From physiologic principles to clinical applications. New York: Springer Verlag; 1983.
32. Bhambhani YN, Holland LJ, Steadward RD. Maximal aerobic power in cerebral palsied wheelchair athletes: validity and reliability. Arch Phys Med Rehabil 1992; 73: 246-252.
33. 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: 349-356.
34. 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: 550-557.
35. Chobanian AV, Bakris GL, Black HR, Cushman WC, Green LA, Izzo JL, Jr. et al. Seventh report of the Joint National Committee on Prevention, Detection, Evaluation, and Treatment of High Blood Pressure. Hypertension 2003; 42: 1206-1252.
36. Andersen LB, Hasselstrom H, Gronfeldt V, Hansen SE, Karsten F. The relationship between physical fitness and clustered risk, and tracking of clustered risk from adolescence to young adulthood: eight years follow-up in the Danish Youth and Sport Study. Int J Behav Nutr Phys Act 2004; 1: 6.
37. Viet AL, van den Hof S, Elvers LH, Ocke MC, Vossenaar M, Seidell JC et al. Risicofactoren En GezondheidsEvaluatie Nederlandse Bevolking, een Onderzoek Op GGD'en (Regenboog-project); jaarverslag 2001 [Risk Factors and Health in the Netherlands, a Survey by Municipal Public; annual report 2001]. Bilthoven: RIVM 2003 Report No.: 260854004.
38. Effects of weight loss and sodium reduction intervention on blood pressure and hypertension incidence in overweight people with high-normal blood pressure. The Trials of Hypertension Prevention, phase II. The Trials of Hypertension Prevention Collaborative Research Group. Arch Intern Med 1997; 157: 657-667.
39. He J, Whelton PK, Appel LJ, Charleston J, Klag MJ. Long-term effects of weight loss and dietary sodium reduction on incidence of hypertension. Hypertension 2000; 35: 544-549.

40. Kelley GA, Kelley KS. Progressive resistance exercise and resting blood pressure: A meta-analysis of randomized controlled trials. Hypertension 2000; 35: 838-843.
41. Whelton SP, Chin A, Xin X, He J. Effect of aerobic exercise on blood pressure: a meta-analysis of randomized, controlled trials. Ann Intern Med 2002; 136: 493-503.
42. Astrand PO, Rodahl K. Physical Training. In: Astrand PO, Rodahl K, editors. Textbook of work physiology: physiological bases of exercise. Singapore: McGraw-Hill Book Co; 1986. p 412-485.
43. Blair SN, Kohl HW, 3rd, Barlow CE, Paffenbarger RS, Jr., Gibbons LW, Macera CA. Changes in physical fitness and all-cause mortality. A prospective study of healthy and unhealthy men. Jama 1995; 273: 1093-1098.
44. Twisk JW, Kemper HC, van Mechelen W, Post GB. Tracking of risk factors for coronary heart disease over a 14-year period: a comparison between lifestyle and biologic risk factors with data from the Amsterdam Growth and Health Study. Am J Epidemiol 1997; 145: 888-898.
45. Lefevre J, Philippaerts R, Delvaux K, Thomis M, Claessens AL, Lysens R et al. Relation between cardiovascular risk factors at adult age, and physical activity during youth and adulthood: the Leuven Longitudinal Study on Lifestyle, Fitness and Health. Int J Sports Med 2002; 23: S32-38.
46. Boreham C, Twisk J, Neville C, Savage M, Murray L, Gallagher A. Associations between physical fitness and activity patterns during adolescence and cardiovascular risk factors in young adulthood: the Northern Ireland Young Hearts Project. Int J Sports Med 2002; 23: S22-26.
47. Hasselstrom H, Hansen SE, Froberg K, Andersen LB. Physical fitness and physical activity during adolescence as predictors of cardiovascular disease risk in young adulthood. Danish Youth and Sports Study. An eight-year follow-up study. Int J Sports Med 2002; 23: S27-31.
48. Janz KF, Dawson JD, Mahoney LT. Increases in physical fitness during childhood improve cardiovascular health during adolescence: the Muscatine Study. Int J Sports Med 2002; 23: S15-21.
49. Nicklas TA, von Duvillard SP, Berenson GS. Tracking of serum lipids and lipoproteins from childhood to dyslipidemia in adults: the Bogalusa Heart Study. Int J Sports Med 2002; 23: S39-43.
50. Twisk JW, Kemper HC, van Mechelen W. Prediction of cardiovascular disease risk factors later in life by physical activity and physical fitness in youth: general comments and conclusions. Int J Sports Med 2002; 23: S44-49.
51. Caspersen CJ, Powell KE, Christenson GM. Physical activity, exercise, and physical fitness: definitions and distinctions for health-related research. Public Health Rep 1985; 100: 126-131.
52. Schaefer EJ, Audelin MC, McNamara JR, Shah PK, Tayler T, Daly JA et al. Comparison of fasting and postprandial plasma lipoproteins in subjects with and without coronary heart disease. Am J Cardiol 2001; 88: 1129-1133.
53. Weiss R, Harder M, Rowe J. The relationship between nonfasting and fasting lipid measurements in patients with or without type 2 diabetes mellitus receiving treatment with 3-hydroxy-3-methylglutaryl-coenzyme A reductase inhibitors. Clin Ther 2003; 25: 1490-1497.

54. Craig SR, Amin RV, Russell DW, Paradise NF. Blood cholesterol screening influence of fasting state on cholesterol results and management decisions. J Gen Intern Med 2000; 15: 395-399.
55. Trost SG, Pate RR, Freedson PS, Sallis JF, Taylor WC. Using objective physical activity measures with youth: how many days of monitoring are needed? Med Sci Sports Exerc 2000; 32: 426-431.
56. White DK, Wagenaar RC, Del Olmo ME, Ellis TD. Test-retest reliability of 24 hours of activity monitoring in individuals with Parkinson's disease in home and community. Neurorehabil Neural Repair 2007; 21: 327-340.

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Chapter 8

Barriers and Facilitators of Physical Activity in Adolescents and Young Adults with Physical Disabilities

Abstract

Objective: To identify barriers and facilitators of physical activity (PA) in adolescents and young adults with a physical disability.
Methods: We reviewed the literature to obtain insight in barriers and facilitators of PA in young adults with a physical disability. Both quantitative and qualitative studies from 1998 to 2008 were included. In addition, we performed 3 focus groups including 16 participants aged 22.4 ± 3.4 years to study additional barriers and facilitators in adolescents and young adults with a childhood onset physical disability.
Results: People with physical disabilities perceived several personal and environmental barriers and facilitators of PA. Personal barriers and facilitators from the quantitative studies included 10 demographic and biological variables, 15 physical variables and 8 psychological variables. We identified 7 barriers or facilitators concerning the social environment and 11 concerning the physical environment. In addition, lack of time, money and information were barriers to PA. The literature review and focus groups showed that lack of energy, existing injuries or complications or fear for developing them, lack of motivation, facility limitations, or lack of information and knowledge appeared to be barriers to PA in people with a physical disability. Fun and social contacts were most frequently mentioned as facilitators of PA.
Conclusion: People with physical disabilities perceived several barriers and facilitators to PA. Rehabilitation has the potential to offer support in overcoming many of these barriers. Personalized tailored PA counselling is important to ensure that people have fun during PA, which was identified as an important facilitator of PA.

Introduction

Health promotion during adolescence and young adulthood

The majority of people who are born with a physical disability, such as myelomeningocele (MMC) or cerebral palsy (CP), or with a physical disability acquired early in childhood, such as traumatic brain injury (TBI) or spinal cord injury (SCI), now survive into adulthood.[1-3] This raises the importance of a healthy lifestyle in this population. As a consequence, health care shifts from disability prevention towards health promotion, to prevent secondary conditions and to ensure a healthy adult life.[4] Secondary conditions affecting people with a physical disability include osteoporosis, decreased balance, muscle strength and endurance, aerobic fitness, and flexibility, increased spasticity, overweight and depression.[4] With increasing age, lifestyle-related diseases, such as diabetes mellitus and cardiovascular diseases may also be of concern. Encouraging physical activity (PA) is an important health promoting activity, and is assumed to have positive effects on secondary conditions.

Physical activity

PA is defined as any bodily movement produced by skeletal muscles that results in energy expenditure.[5] PA can be divided into several categories such as leisure time, including sports and exercise, household tasks (e.g. cleaning, gardening and home repair), and occupation-related tasks. The Healthy People 2010 report emphasises that regular PA activity throughout life is important for maintaining a healthy body.[6] Health benefits include a lower risk of hypertension, diabetes type II, cardiovascular disease, as well as improvements in weight loss or maintenance, fitness and quality of life.[7-9]

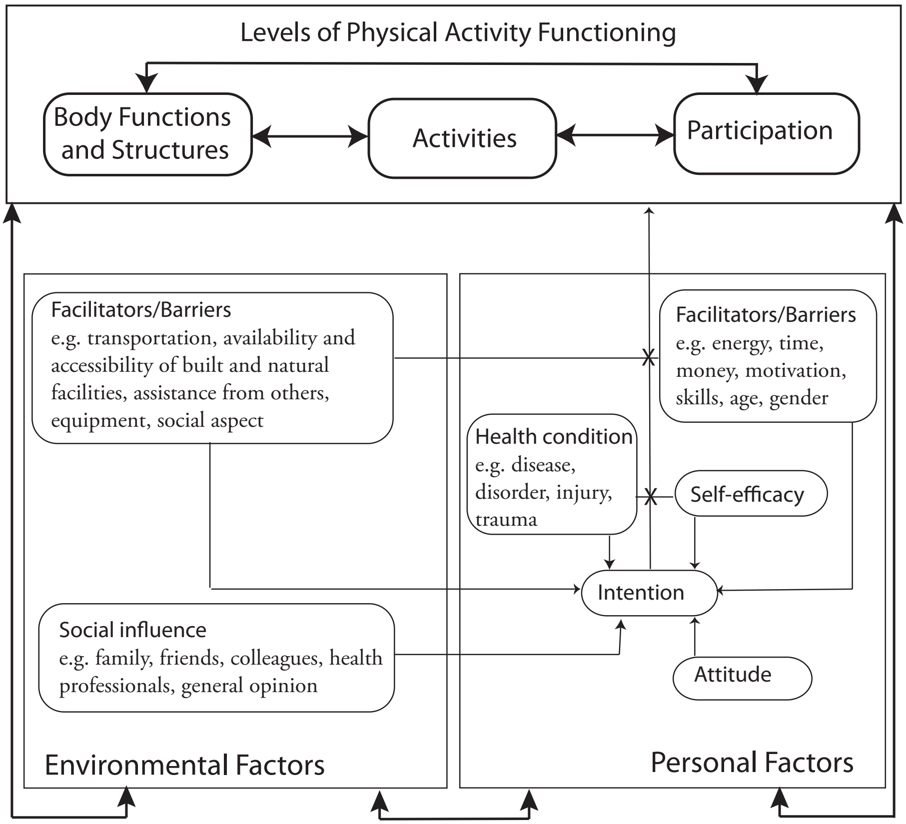
In the Netherlands, only 52% of the adult population meets the Dutch Public Health PA guideline,[10] i.e. 30 minutes of moderate PA for at least 5 days per week.[11] In people with a physical disability, an inactive lifestyle is even more common than in the general population.[6,7,12] Therefore, they are at risk to experience a vicious cycle: inactivity leading to a reduced fitness and an increase in body fat. This will result in higher physical strain during the performance of daily activities, which may lead to further physical inactivity.[13] Previous studies in adolescents and young adults with MMC and CP showed low levels of PA and low aerobic fitness, compared to able bodied people of the same age.[14-16] PA counselling can increase PA, and consequently reduce the deconditioning and associated reductions in physical functioning and health.[7]

Physical activity for people with a disability

In order to develop programs to promote PA in people with a childhood onset disability, it is important to understand the factors that determine PA behaviour in this population.[17,18] To describe relationships between PA, its correlates and functioning in people with a physical disability, Van der Ploeg and co-workers proposed the Physical Activity for people with a Disability (PAD) model (see Figure 1).[19] The model uses the International Classification of Functioning, disability and health (ICF) as starting point. The ICF model describes the functioning of an individual in a specific domain as a dynamic interaction or complex relationship with environmental and personal factors, given a certain health condition.[20] In the PAD model, PA behaviour and its correlates were integrated into the ICF model.[19] The model can be used as a theoretical framework for future interventions and research on PA promotion in people with a disability.[19]

According to the PAD model, several personal and environmental factors may influence PA behaviour. Whereas for healthy adolescents and young adults knowledge on barriers and facilitators of PA is increasing,[21-23] less information is available for people with a physical disability.[12] Therefore, we reviewed the literature to obtain insight in barriers and facilitators of PA in adolescents and young adults with a physical disability. In addition, we performed focus groups to study additional barriers and facilitators in adolescents and young adults with a childhood onset physical disability.

Figure 1 Physical Activity for people with a Disability model.[19]



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Methods

Literature review

We conducted a computer search (PubMed) in the English language using the following keywords: physical activity, exercise, sports participation, adolescents, (young) adults, physical disability, mobility limitation, determinants, correlates, barriers and facilitators. In addition, we conducted manual searches using the reference lists from relevant articles.

We included both quantitative and qualitative studies on barriers and facilitators of PA and sports participation in people with various types of physical disabilities from the past 10 years (1998-2008). We excluded studies that focused on people with primarily intellectual disabilities, learning disabilities, psychiatric disorders, visual impairments and hearing impairments, studies including people who were completely dependent on electric wheelchairs, and studies focusing on older adults (average age > 50 years) or parents of children with a disability. Furthermore, we excluded studies focusing on correlates in which disability or health benefits were used as an outcome measure of PA or those studying consequences of physical inactivity.

The quality of the selected studies was determined according to the study design and the instruments used to assess PA levels. We considered longitudinal studies to be of highest quality (quality A) because such design allows identification of true mediators of change, i.e. to study whether changing a correlate resulted in improvement in PA behaviour. In these studies, both PA and the determinant are assessed at several points in time. Cross-sectional studies assessing both PA as well as the determinant were considered as medium quality. We defined studies on barriers and facilitators of objectively assessed PA (quality B) as having higher quality than those of self-reported levels of PA (quality C). Finally, because qualitative studies did not assess actual PA levels, we considered the barriers and facilitators mentioned in these studies as having lower quality than the quantitative studies. A distinction was made between qualitative studies that used standardised questionnaires (quality D) and studies that used (semi-structured) interviews or focus groups (quality E). Qualitative studies were used as supporting evidence for relevant barriers and facilitators found in the quantitative studies. Studies with small sample sizes ($n < 50$) were interpreted with caution.

Similar to the PAD model,[19] this study focused on personal and environmental factors that may influence PA. Personal factors were subdivided into the following categories: demographic and biological factors, physical factors, and psychological factors (including a person's intention and attitude towards PA and self-efficacy). Environmental factors included social influences as well as factors from the physical environment. The factors obtained from the quantitative studies were coded as having a positive (+), inverse (-) or no significant (0) association with PA.

Focus groups

Studies on barriers and facilitators of PA in adolescents and young adults with a childhood onset physical disability are scarce. Therefore, focus groups were conducted to identify main barriers and facilitators of PA in these people.

Participants

Three focus groups were conducted with convenience samples of adolescents and young adults with childhood-onset physical disabilities from the Rehabilitation department of Erasmus MC and Rijndam Rehabilitation Centre, both in Rotterdam, and Sophia Rehabilitation in The Hague. The Medical Ethics Committee of Erasmus MC approved this study. In total, 16 (75% male) adolescents and young adults aged 22.4 ± 3.4 years participated, of whom 50% were wheelchair dependent. Seven participants were diagnosed with MMC, 4 with CP, 2 with acquired brain injury and 2 with rheumatoid arthritis.

Procedures

Focus groups were facilitated by two members of the research team (LB and TW). At the beginning of each focus group session, the purpose of the study was explained and written informed consent was obtained. A semi structured questioning route was used in the focus groups. Participants were asked about factors influencing their PA behaviour, including daily PA and sports; assessing reasons for participation or non-participation. Furthermore, participants were asked about their preferences on how to improve PA, which resulted in additional facilitators of PA. Groups ran for approximately 1.5 hour.

Data analyses

Focus groups were audio-taped, and tapes were transcribed verbatim to ensure systematic analyses of the interviews. Tape recordings of focus groups were content analysed by two members of the research team (LB and TW), which involved the systematic examination of the transcripts to identify major categories of barriers and facilitators of PA.

Table 1 Quantitative studies on correlates of physical activity in people with a physical disability.

Reference	*n*	*Age* mean± SD (range)	Gender (% male)	Main diagnoses	Quality	Physical activity measure
Boslaugh & Andresen 2006[37]	4038	18+	32	Back or neck problems (18%), Arthritis/rheumatism (14%), Bone/joint injury (11%)	C	Active yes/no (meet recommendation)
Buffart et al. *in press*[38]	51	21.1± 4.5 (16-30)	51	SB	C	Sports participation yes/no
Buffart et al. 2008[16]	51	21.1± 4.5 (16-30)	51	SB	B	Accelerometry-besed activity monitor 2d
Doerksen et al. 2007[30]	196	46.1 ± 9.8	12	MS	B + C	Pedometer 7d, Questionnaire IPAQ
Heller et al. 2002[33]	83	46.6 ± 11.0 (30-79)	47	CP	C	Exercise participation. Questionnaire PAQ-A
Kinne et al. 1999[32]	113	47 ± 1.4 (17-69)	42	Neuromuscular/skeletal (37%), MS (39%), SCI (16%)	C	Self-reported exercise maintenance (yes/no)
Maher et al. 2007[15]	112	13.9 ± 1.9 (11-17)	68	CP	C	Questionnaire PAQ-A
Maltais et al. 2005[34]	11	10-16	64	CP	C[a]	Questionnaire total EE/resting EE
Morris et al. 2005[31]	173	46.1	0	MS	B	ActiGraph accelerometer 7d
Motl et al. 2006[28]	196	46.1 ± 9.8 (22-67)	12	MS	B + C	Pedometer 7d
Motl et al. 2007 [29]	196	46.1 ± 9.8 (22-67)	12	MS	B	Questionnaire GLTEQ
Nosek et al. 2006[36]	386	47.1 ± 10.1 (18-65)	0	Joint and connective tissue disease (41%), Neuro-muscular disease, post polio (20%), SCI, SB (16%), MS (11%)	C	Self administered questionnaire
Roebroeck et al. 2006[27]	14	18 ± 4 (14-26)	57	SB	B[a]	Accelerometry-based activity monitor 2d
Van den Berg-Emons et al. 2001[26]	14	18 ± 4 (14-26)	57	SB	B[a]	Accelerometry-based activity monitor 2d
Van den Berg-Emons et al. *in press*[25]	16	42.2 ± 13.3	88	SCI	A[a]	Accelerometry-based activity monitor 2d
Van der Ploeg et al. 2005[35]	1007	46 ± 14	51	Stroke (25%), Neurological disorders (16%), Back disorders (15%), chronic pain/whiplash (13%), Rheumatic related disorders (10%)	C	Questionnaire PASIPD
Van der Ploeg et al. 2005[24]	165	46 ± 13	79	Stroke (34%), Chronic pain/whiplash (18%), rheumatic related disorders (14%), Neurological disorders (18%), amputation (10%)	A	Questionnaire PASIPD
Warms et al. 2007[18]	50	46.3 ± 13.6 (18-75)	54	SCI (50%), MS (8%), Amputation (8%)	B + C	Questionnaire PASIPD, ActiGraph accelerometer

CF= cystic fybrosis. CP= cerebral palsy. MS= multiple sclerosis. SB= spina bifida. SCI= spinal cord injury. EE= energy expenditure. GLTEQ= Godin Leisure-Time Exercise Questionnaire. PASIPD= Physical Activity Scale for Individuals with a Physical Disability. [a]$n < 50$.

Table 2 Qualitative studies on correlates of physical activity in people with a physical disability.

Reference	***n***	**Age** mean± SD (range)	**Gender** (% male)	**Main diagnoses**	**Quality**	**Method**
Ellis et al. 2007[42]	223	45.4 ± 10.8 (18-73)	29	SCI (21.5%), CP (18,8%), MS (17,9%)	E	6 open-ended questions
Kerstin et al. 2006[43]	16	36 ± 10.6 (21-61)	73	SCI	E[a]	Semi-structured interviews
Levins et al. 2004[44]	88	42 (24-59)	63	SCI	E[a]	Semi-structured interviews
Rimmer et al. 2000[19]	53	18-64	0	Arthritis (30%), Stroke (22%), MS (14%), Diabetes, heat disease (10%).	D	Questionnaire B-PED
Rimmer et al. 2004[45]	42	40.2 ± 12.8	55	SCI (53.5%), Limited leg ability (41.9%), Limited arm/hand ability (25.5%)	E[a]	Focus groups
Scelza et al. 2005[40]	72	44.1 ± 13.0 (18-80)	69	SCI	D	Questionnaire B-PED
Vissers et al. 2008[41]	32	45 ± 12	75	SCI	E[a]	Semi-structured interviews
Wu et al. 2001[46]	143	33.3	92	SCI	E	4-point importance rating scale

CF= cystic fybrosis. CP= cerebral palsy. MS= multiple sclerosis. SB= spina bifida. SCI= spinal cord injury. B/PED° Barriers to Physical Exercise and Disability.
[a]$n < 50$.

Results

Literature review

In total, 18 quantitative studies (Table 1) and 8 qualitative studies (Table 2) were included. There were two longitudinal studies that examined mediators of PA improvements.[24,25] One study objectively assessed levels of PA in people with SCI during inpatient rehabilitation and in the year after discharge. Lesion characteristics, both level of lesion and completeness of lesion, were determinants of PA improvements after discharge,[25] whereas age and gender were not. Another longitudinal study measured intervention induced improvements in self-reported PA behaviour in people with various types of physical disabilities using a questionnaire, and showed several determinants of PA change, which are worth targeting in interventions. These addressed psychosocial determinants: attitude and self-efficacy towards PA, social influences from family and friends concerning PA, the barriers: limited environmental possibilities, poor health conditions and lack of energy, and the perceived benefits: improved health and reduced risk of disease, better feeling about oneself, improved fitness and improved daily functioning.[24] Furthermore, 8 studies included objective PA measurements[16,18,26-31] and self-reported measurements of PA were included in 11 studies.[15,18,28,30,32-38] Three studies focused only on exercise or sports, which is a sub-category of PA that is planned and structured.[32,33,38]

Quantitative studies

Personal barriers and facilitators of physical activity

Table 3 presents personal barriers and facilitators of PA that were mentioned in the quantitative studies. The review identified 10 demographic and biological variables, of which 6 were studied more than 3 times, and included objective PA assessments. The most frequently studied variables were age, gender, education and marital status. Among the studies including different age ranges, results on whether older people were less physically active than younger people were inconsistent. The majority of the studies, including 3 studies with objective PA assessments, showed no differences in PA between males and females. Furthermore, one study using objective PA assessments showed no association between PA and education, which was supported by 3 of the 4 studies using self-reported PA assessments. The study with objective PA assessment showed no association between income and PA, which was supported by 2 of the 3 studies that used self-reported PA assessments. Marital status was not associated with PA.

The review identified 15 physical variables, of which 6 were included in more than 3 studies (Table 3). Studies using objective PA assessments showed that people with higher mobility were more physically active. This was supported by half of the studies using self-reported PA assessments. Furthermore, a better general health or health condition appeared to be associated with being more physically active, and improvements in this variable appeared to increase PA. Also reduced lack of energy or fatigue appeared to increase PA. This association was supported by 2 other studies using self-reported PA assessments. However the only study that assessed PA objectively did not report this association. Overweight or obesity was not associated with PA in all studies, except for one study using objective PA assessments. Inconsistent results were found regarding the association between PA and duration of disability or treatment, however, the only study that included objective PA assessments showed no association.

The review included 8 psychological variables of which self-efficacy was studied most often. Except for one study using both objective and self-reported PA assessments in people with a broad age range and various types of diagnoses, all studies reported a positive association between self-efficacy and PA. Also improving self-efficacy appeared to increase PA. One longitudinal study showed that mental health was a mediator of improved PA; however one study using objective PA assessment and 3 studies using self-reported PA assessments did not show an association between PA and mental health.

Environmental barriers and facilitators of physical activity

Table 4 presents environmental barriers and facilitators of PA mentioned in the quantitative studies. Lack of time and lack of money appeared to be barriers to PA. Furthermore, the review identified 7 variables of the social environment and 11 variables of the physical environment, of which social support and accessibility of facilities or services were studied most often. Studies on social support were inconsistent but the higher evidence studies showed no association with PA. Furthermore, studies that used objective PA assessments showed that better access to facilities or services was associated with higher PA levels; however this was not supported by studies that used self-reported PA assessments.

Table 3 Personal factors and the association with physical activity.

Personal factors	Association	References		
		Quality A	Quality B	Quality C
Demographic and biological variables				
Age	-		29	15, 18, 35, 37
	0	25[a]	18	32, 33, 36, 38
Gender, being male	+			38
	0	25[a]	16, 29	15, 32 , 37
Education level	+			35
	0		29	32 , 36, 37
Cognitive functioning	+		27[a]	
	0			33
Employment, having a job	+		29	35, 37
	0			36
Social economic status	0			15
Income	+			37
	0		29	32, 36
Ethnicity	+			37
	0		29	36
Marital status	0		29	32, 36, 37
Having children at home	+			35
	0		29	

Table 3 Continued.

	Association	References Quality A	Quality B	Quality C
Physical variables				
Type of disability	+		29	36
	0			32
Duration of disability/treatment	0		29	37
	-			35, 36
Functional status/independence	+		31	36
	0		26[a]	32, 35, 38
Daily functioning	+	24		
Arm-hand limitations	0			33
Lesion characteristics (level/completeness)	-	25[a]		
	0		26[a]	
Mobility	+		16, 26, 29	15, 36
	0			33, 38
Need for assistance ADL	-			36
General health, health condition	+	24		18, 33, 35, 37
	0		18	32
Lack of energy / fatigue	-	24		32, 35
	0			18
	-			34[a]
	0		18	18, 36
(Perceived) Fitness	0			35
	+	24	16	
Overweight-obesity	-		18	
	0		16	18, 32, 37, 38
Thermosensitivity	0		29	
Psychological variables				
Self-efficacy	+	24	28, 31	28, 32, 35, 36, 38
	0		18	18
Attitude towards PA	+	24		35
PA enjoyment	+		28	28, 38
Motivation	+			32
	0		18	18, 35
Stage of readiness to change	+			18
	0		18	
Health improvement, reduced risk of disease	0			35
	+	24		
Mental health, (better feeling, less depression)	0		18	18, 35, 36
	+	24		
Physical appearance	+			38

ADL= activities of daily living. PA= physical activity. [a]n< 50. - barrier. + facilitator . 0 no association.

Table 4 Environmental factors and the association with physical activity.

Environmental factors	Association	References Quality A	Quality B	Quality C
Lack of time	-			35
Costs, lack of money	-		30	35
	0			30, 32
Lack of information/education	0			32
Social environment				
Social support family/friends	+			18, 38
	0	24	18	32, 35, 36
Health professionals	+			18, 32
	0		18	
Social integration/meeting people	0			35, 36
Presence of active others	0		30	30
Personal assistance	+			36
Having insurance	+			36
	0			37
Caregiver's attitude	+			33
Physical environment				
Total physical environment/ aesthetics neighbourhood	0		18, 30	18, 30
Lack of transportation	0			32 , 35
Accessible facilities/services	+		30, 31	
	0			30, 32, 33
Proximity of services	+		30, 31	
	0			30
Proximity of public transportation	+		30	
	0			30
Environmental possibilities	0		30	30, 35
	-	24		
Dwelling density	0		30	30
Heavy traffic	0		30	30
Crime	0		30	30
Type of residence	+			33
Number of motor vehicles at home	0		30	30

- barrier. + facilitator. 0 no association.

Qualitative studies

Personal barriers and facilitators of PA mentioned in the qualitative studies are summarized in Table 5 and environmental barriers and facilitators are summarized in Table 6. The review showed that more studies focussed on barriers than on facilitators. Two studies used a standardized questionnaire to assess barriers to PA,[39,40] however among different diagnoses. This may have caused the differences in the percentages of people that perceived a certain variable as a barrier to PA.

Personal barriers and facilitators of physical activity

The physical variables health condition and fatigue, injuries, or lack of energy, were physical barriers to PA. Health was most frequently mentioned as a PA facilitator. Lack of motivation, lack of interest, boredom, being too lazy, or exercise being too difficult were most often mentioned as psychological barrier to PA. Most frequently mentioned psychological facilitator of PA was enjoyment. Furthermore, a large proportion of people with SCI reported emotional distress (69%) and physical appearance (66%) to be barriers to PA.[41]

Environmental barriers and facilitators of physical activity

Lack of time and high costs were frequently mentioned as environmental barriers to PA. Approximately one-third of the populations that were studied mentioned social support to be a facilitator of PA. Social contacts and competition were also perceived as important facilitators. Negative social attitude and lack of assistance were perceived as barriers to PA. Regarding the physical environment, many people perceived a lack of knowledge on how and where to exercise, lack of transportation and inaccessibility of environment and facilities as barriers to PA. Access to facilities, equipment and programmes were mentioned as important facilitators of the physical environment.

Table 5 Personal barriers and facilitators of physical activity mentioned in qualitative studies.

Personal Factors	% perceived	References Quality D	Quality E
Physical variables			
Barriers			
Self care problems	47		41[a]
Pain	25		41, 42[a]
Poor health condition	25-53	39, 40	41[a]
Exercise will not improve condition	8-15	39, 40	
Injury/falls/illness/ worsening condition	11-17	39, 40	42
Fatigue/ lack of energy	12-66	39, 40	42
Facilitators			
Functional independence			43[a]
Health			43, 46
Improving fitness			46
Psychological variables			
Barriers			
Mental health problems	47		41[a]
Emotional distress	69		41[a]
Physical appearance	66		41[a]
Lack of motivation	41-54	39, 40	
Lack of interest	18-33	39, 40	
Boredom	18-32	39, 40	
Fear of unknown		45[a]	
Being too lazy	24-32	39, 40	
Exercise is too difficult	15-32	39, 40	
Facilitators			
Enjoyment			43, 46
Goal setting			43[a]
Role model			43[a]
Creating Routine			43[a]

[a] $n < 50$.

Table 6 Environmental barriers and facilitators of physical activity mentioned in the qualitative studies.

Environmental factors	**% perceived**	**References Quality D**	**Quality E**
Barriers			
Lack of time	8-32	39, 40	42
Costs/finances	13-84	39, 40	41, 42, 45[a]
Facilitators			
More money, less expensive activities	12		42
Social influences			
Barriers			
Negative societal attitude	9		41, 44, 45[a]
Negative attitude family and friends	13		41[a]
Dissatisfaction social support	19		41[a]
Problems facility staffing			45[a]
Lack of/concerns about assistance/preparation	34		41, 42, 45[a]
Facilitators			
Social support family, friends, significant others	24-34		42, 43[a]
Social contacts			43, 46
Reference framework			43[a]
Support or assistance	16		42
Competition			43, 46
Physical environment			
Barriers			
Lack of programs	17		42
Lack of information	38		41, 45[a]
Not knowing how to exercise	28-37	39, 40	
Lack of knowledge where to exercise	36-58	39, 40	41, 45[a]
Lack of transportation	8-61	39, 40	42, 45
Inaccessibility to environment, facilities, equipment, programs	17-72		41, 42, 44, 45[a]
Movement possibilities in/around house	16-25		41[a]
Inclement weather/ harsh temperature	12		42
Facilitators			
Access to facilities, equipment, programs	22		42-44[a]

[a] *n* < 50.

Focus groups

Nearly all focus group participants had been involved in sports somewhere during childhood. Some of them stopped sports participation during adolescence or young adulthood due to injuries, complications associated to their condition, or other priorities. Results of this qualitative study showed that there were several personal and environmental barriers and facilitators to PA in adolescents and young adults with a childhood onset physical disability. These are listed in Table 7.

Personal barriers and facilitators

Several physical aspects were mentioned as barriers to PA, including having an injury or complication associated with the condition, and lack of energy or being fatigued. In addition, some people mentioned not perceiving health benefits as a barrier to PA, whereas others mentioned perceiving health benefits as a facilitator of PA. Furthermore, maintenance of fitness, muscle strength, or functional independence, and physical appearance were mentioned as reasons for engaging in PA.

Psychological barriers that were mentioned in the focus groups included feeling uncomfortable, or ashamed when engaging in PA, fear for complications or injuries and several motivational barriers. Fun was frequently mentioned as a reason to engage in sports or PA. Furthermore, attitude (If I want to be active, I will), wanting to win or achieve a goal, and clearing the mind were mentioned as reasons for being physically active. In addition, several motivational facilitators were mentioned, including being rewarded after exercise by for example social events or perceived benefits. Other reasons for being physically active were active transport, e.g. cycling to work or wheeling to the shops, and because people had done so since childhood.

Environmental barriers and facilitators

Lack of professional support was mentioned as a PA barrier of the social environment. Social contact was frequently given as a reason to be physically active. Furthermore, PA was often facilitated by others; they had an important role in stimulating the adolescents or young adult for being physically active, as well as facilitating transportation to facilities and having somebody to be active with.

Also aspects of the physical environment appeared to influence PA. Bad weather was mentioned as a barrier to PA, whereas good weather was a facilitator. Having a car was a barrier to active transport, but it appeared to facilitate the ability to go to sports and exercise facilities. Other frequently mentioned barriers were associated with facilities, equipment, lack of information and knowledge, and transportation. Possibilities to engage in PA (e.g. sports or exercise classes) appeared to be limited, particularly in rural areas, or they were focussed on the elderly population. Furthermore, people were restrained from purchasing expensive equipment unless they were very sure that it is worth the investment, and second-hand equipment is scarce. Also, the duration of acquiring equipment restricts people from purchasing. Finally lack of time due to school or work was mentioned as a barrier to PA.

Table 7 Barriers and facilitators of physical activity in adolescents and young adults with childhood-onset physical disabilities.

Personal factors	
Physical barriers	*Physical facilitators*
Injury/complication	Maintenance fitness/muscle strength
Lack of energy/fatigue	Maintenance of functional independence
	- Walking ability
	- Wheelchair skills
No perceived health benefits	Health benefits
	Physical appearance
	- Weight loss/maintenance
	- Muscles
Psychological barriers	*Psychological facilitators*
Feeling uncomfortable or ashamed	Fun
Fear for complications/injuries	Attitude
Motivational barriers	Motivational facilitators
- Wake up early	- Feelings of fulfilment
- Other priorities (e.g. tv, computer, social activities)	- Physical challenge
- Too much effort (e.g. changing clothes)	- Enjoyment
	- Rewarding
	- Goal setting
	Achieve goals/ wanting to win
	Clear the mind
Other personal barriers	*Other personal facilitators*
	Active transport
	Habit
Environmental factors	
Barriers of the social environment	*Facilitators of the social environment*
Lack of professional support	Social contacts
	- Social support
	- Parents
	- Professional
	- Peers/friends
	- Sports team
	- School
Barriers of the physical environment	*Facilitators of the physical environment*
Bad weather	Nice weather
Having a car	Having a car
Facilities	
- Limited possibilities, particularly for young people and in rural areas	
- Distance to facilities	
- Opening hours	
- Crowdedness	

Table 7. Continued.

Equipment	
- Expensive	
- Duration to purchase equipment	
Lack of information/knowledge on	
- Where to exercise	
- How to exercise	
- Physical and medical consequences	
Transportation problems	
- Duration (waiting for taxi)	
- Costs (public transport, taxi)	
- Dependency upon others	
- Carrying equipment in public transport	
Other environmental barriers	*Other environmental facilitators*
Financial/costs	
- Expensive equipment	
- Expensive entrance of facilities	
- Transportation to facilities	
Lack of time	

Discussion

The literature review showed that only few studies are available on barriers and facilitators of PA in people with childhood-onset physical disabilities, and often, these studies included small sample sizes.[26,27,34] Furthermore, the majority of the studies in these populations focused on non-modifiable factors such as age, gender, mobility, or functional independence. In regard to development of intervention, identification of such factors are important for defining subgroups who are at increased risk of developing an inactive lifestyle, or subgroups that need a different approach to promote PA. However, to change PA behaviour, it is relevant to focus more on modifiable correlates such as psychological factors and several physical factors. A study on sports participation in adolescents and young adults with MMC showed that sports participants perceived higher self-efficacy, exercise enjoyment, physical appearance than non-participants, and they had more social support.[38] However, sports participation is a planned and structured sub-category of PA and future studies should investigate whether these factors are also important barriers and facilitators of daily PA in general.

The literature review was extended to adolescents and young adults with other than childhood-onset physical disabilities, such as MS, SCI, and neuromuscular or neurological disorders. These studies showed that many barriers and facilitators of PA are related to attitude, motivation and self-efficacy, which is comparable to the general population.[21,23] Also for exercise maintenance, perceiving lower motivational barriers and self-efficacy seemed to be important.[32] However, in contrast to the general population, lack of energy, having injuries or complications associated with the condition, as well as fear for injury or complications, and lack of professional support were frequently identified as barriers to PA. The present focus group results support these findings for people with a childhood onset physical disability. However, quantitative data should provide more insight in barriers and facilitators of PA in people with a childhood onset physical disability.

Compared to personal factors, fewer studies have investigated environmental factors influencing PA. Many people with physical disabilities, including adolescents and young adults with a childhood onset physical disability as shown in the results of the focus groups, perceived a lack of knowledge on how and where to exercise, and perceived difficulties with access to facilities, equipment and programmes. However, most of these barriers were identified from qualitative studies and only one quantitative study showed that limited environmental possibilities was a barrier to PA.[24] Future insight in the role of the environment in PA behaviour is warranted.

For people with physical disabilities, Rehabilitation medicine can have an important role in promoting PA and overcoming barriers to PA. Rehabilitation medicine can inform patients on important health benefits of an active lifestyle and on where and how to exercise, it can facilitate in equipment and programmes, and supervise and assist during exercise. A previous study of van der Ploeg and co-workers in the Netherlands, showed that a rehabilitation intervention, providing both tailored sports advice and personalised tailored PA counselling, can successfully improve levels of PA of people who acquired a physical disability during adulthood on the short and long term.[47,48] However, future studies are warranted to obtain insight in whether such intervention also improves PA levels of people with a childhood onset physical disability. Individual tailoring is important to ensure that each person find the type of activity he or she enjoys. This is very important considering that fun or enjoyment is most frequently mentioned as a facilitator of PA in both quantitative and qualitative studies.

Qualitative studies, such as focus groups, are useful to examine reasons and motives behind people's behaviours, and they have the advantage to allow collection of a large amount of data in a short period of time.[49] However, they do not allow statistical testing necessary to differentiate between the importance of several barriers and facilitators. Therefore, we were unable to draw conclusions on which barriers and facilitators have the largest impact. To identify personal- and environmental correlates or determinants of PA, quantitative studies including simultaneous objective assessments of PA and variables are warranted. Therefore, there is a need to develop valid and reliable assessment instruments that measure both personal and environmental factors relevant to PA behaviour of people with physical disabilities.

Conclusion

People with physical disabilities perceived several barriers and facilitators to PA. First, several barriers related to attitude, motivation and self-efficacy were important, which is comparable to the general population. In addition, lack of energy, existing or fear for developing injuries or complications, facility limitations, and lack of information and knowledge appeared to be barriers to PA. Rehabilitation medicine has the potential to play an important role in health promotion. It can offer support in overcoming many perceived barriers to PA. Personalised tailored PA counselling is important to ensure that people have fun during PA, which was identified as an important facilitator of PA.

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References

1. Strauss DJ, Shavelle RM, Anderson TW. Life expectancy of children with cerebral palsy. Pediatr Neurol 1998; 18: 143-149.
2. Bowman RM, McLone DG, Grant JA, Tomita T, Ito JA. Spina bifida outcome: a 25-year prospective. Pediatr Neurosurg 2001; 34: 114-120.
3. Oakeshott P, Hunt GM. Long-term outcome in open spina bifida. Br J Gen Pract 2003; 53: 632-636.
4. Rimmer JH. Health promotion for people with disabilities: the emerging paradigm shift from disability prevention to prevention of secondary conditions. Phys Ther 1999; 79: 495-502.
5. Caspersen CJ, Powell KE, Christenson GM. Physical activity, exercise, and physical fitness: definitions and distinctions for health-related research. Public Health Rep 1985; 100: 126-131.
6. United States Department of Health and Human Services. Healthy People 2010: Understanding and improving health. 2nd ed. Washington DC: US Government Printing Office, Nov 2000. Available from: http://www.health.gov/healthypeople.
7. Durstine JL, Painter P, Franklin BA, Morgan D, Pitetti KH, Roberts SO. Physical activity for the chronically ill and disabled. Sports Med 2000; 30: 207-219.
8. US Department of Health and Human Services, 1996. Physical activity and health: a report of the Surgeon General. US Department of Health and Human Services, Centers of Disease Control and Prevention, National Center for Chronic Disease Prevention and Health Promotion. Atlanta, GA.
9. Haskell WL, Lee IM, Pate RR, Powell KE, Blair SN, Franklin BA et al. Physical Activity and Public Health: Updated Recommendation for Adults from the American College of Sports Medicine and the American Heart Association. Med Sci Sports Exerc 2007; 39: 1423-1434.
10. Kostalova B, Frenken F, Storm I. Norm gezond bewegen 2001-2004. In: Volksgezondheid Toekomst Verkenning, Nationale Atlas Volksgezondheid. Bilthoven: RIVM, <http://www.zorgatlas.nl> Beïnvloedende factoren\ Leefstijl\ Bewegen en sporten, 5 oktober 2005.
11. Kemper HGC, Ooijendijk WTM, Stiggelbout M. Consensus over de Nederlandse Norm voor Gezond Bewegen. Tijdschr Soc Gezondheidsz 2000; 78: 180-183.

12. Heath GW, Fentem PH. Physical activity among persons with disabilities--a public health perspective. Exerc Sport Sci Rev 1997; 25: 195-234.
13. Hoffman MD. Cardiorespiratory fitness and training in quadriplegics and paraplegics. Sports Med 1986; 3: 312-330.
14. Bandini LG, Schoeller DA, Fukagawa NK, Wykes LJ, Dietz WH. Body composition and energy expenditure in adolescents with cerebral palsy or myelodysplasia. Pediatr Res 1991; 29: 70-77.
15. Maher CA, Williams MT, Olds T, Lane AE. Physical and sedentary activity in adolescents with cerebral palsy. Dev Med Child Neurol 2007; 49: 450-457.
16. 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.
17. Bartholomew LK, Parcel GS, Kok G. Intervention mapping: a process for developing theory- and evidence-based health education programs. Health Educ Behav 1998; 25: 545-563.
18. Warms CA, Belza BL, Whitney JD. Correlates of physical activity in adults with mobility limitations. Fam Community Health 2007; 30: S5-16.
19. Van der Ploeg HP, van der Beek AJ, van der Woude LH, van Mechelen W. Physical activity for people with a disability: a conceptual model. Sports Med 2004; 34: 639-649.
20. WHO. International Classification of Functioning, Disability and Health (ICF). Geneva: World Health Organization; 2001.
21. Van Der Horst K, Paw MJ, Twisk JW, Van Mechelen W. A brief review on correlates of physical activity and sedentariness in youth. Med Sci Sports Exerc 2007; 39: 1241-1250.
22. Sallis JF, Prochaska JJ, Taylor WC. A review of correlates of physical activity of children and adolescents. Med Sci Sports Exerc 2000; 32: 963-975.
23. Trost SG, Owen N, Bauman AE, Sallis JF, Brown W. Correlates of adults' participation in physical activity: review and update. Med Sci Sports Exerc 2002; 34: 1996-2001.
24. Van der Ploeg HP. Underlying mechanisms of improving physical activity behaviour after rehabilitation. Promoting physical activity in the rehabilitation setting Amsterdam: Vrije Universiteit; 2005. p 115-134.
25. Van den Berg-Emons RJG, Bussmann JBJ, Haisma JA, Sluis TAR, van der Woude LHV, Bergen MP et al. Prospective study on physical activity levels after spinal cord injury during inpatient rehabilitation and the year after discharge. Arch Phys Med Rehabil in press.
26. Van den Berg-Emons HJ, Bussmann JB, Brobbel AS, Roebroeck ME, van Meeteren J, Stam HJ. Everyday physical activity in adolescents and young adults with meningomyelocele as measured with a novel activity monitor. J Pediatr 2001; 139: 880-886.
27. Roebroeck ME, Hempenius L, van Baalen B, Hendriksen JG, van den Berg-Emons HJ, Stam HJ. Cognitive functioning of adolescents and young adults with meningomyelocele and level of everyday physical activity. Disabil Rehabil 2006; 28: 1237-1242.
28. Motl RW, Snook EM, McAuley E, Scott JA, Douglass ML. Correlates of physical activity among individuals with multiple sclerosis. Ann Behav Med 2006; 32: 154-161.
29. Motl RW, Snook EM, McAuley E, Scott JA, Hinkle ML. Demographic correlates of physical activity in individuals with multiple sclerosis. Disabil Rehabil 2007; 29: 1301-1304.
30. Doerksen SE, Motl RW, McAuley E. Environmental correlates of physical activity in multiple sclerosis: A cross-sectional study. Int J Behav Nutr Phys Act 2007; 4: 49.
31. Morris KS, McAuley E, Motl RW. Self-efficacy and environmental correlates of physical activity among older women and women with multiple sclerosis. Health Educ Res 2007.

32. Kinne S, Patrick DL, Maher EJ. Correlates of exercise maintenance among people with mobility impairments. Disabil Rehabil 1999; 21: 15-22.
33. Heller T, Ying Gs GS, Rimmer JH, Marks BA. Determinants of exercise in adults with cerebral palsy. Public Health Nurs 2002; 19: 223-231.
34. Maltais DB, Pierrynowski MR, Galea VA, Bar-Or O. Physical activity level is associated with the O_2 cost of walking in cerebral palsy. Med Sci Sports Exerc 2005; 37: 347-353.
35. Van der Ploeg HP. Correlates of physical activity during outpatient rehabilitation: a study in 1007 people with disabilities. Promoting physical activity in the rehabilitation setting. Amsterdam: Vrije Universiteit; 2005. p 57-75.
36. Nosek MA, Hughes RB, Robinson-Whelen S, Taylor HB, Howland CA. Physical activity and nutritional behaviors of women with physical disabilities: physical, psychological, social, and environmental influences. Womens Health Issues 2006; 16: 323-333.
37. Boslaugh SE, Andresen EM. Correlates of physical activity for adults with disability. Prev Chronic Dis 2006; 3: A78.
38. Buffart LM, van der Ploeg HP, Bauman AE, van Asbeck FW, Stam HJ, Roebroeck ME, van den Berg-Emons RJ. Sports participation in adolescents and young adults with myelomeningocele and its role in total physical activity behaviour and fitness. J Rehabil Med *in press*.
39. Rimmer JH, Rubin SS, Braddock D. Barriers to exercise in African American women with physical disabilities. Arch Phys Med Rehabil 2000; 81: 182-188.
40. Scelza WM, Kalpakjian CZ, Zemper ED, Tate DG. Perceived barriers to exercise in people with spinal cord injury. Am J Phys Med Rehabil 2005; 84: 576-583.
41. Vissers M, van den Berg-Emons R, Sluis T, Bergen M, Stam H, Bussmann H. Barriers to and facilitators of everyday physical activity in persons with a spinal cord injury after discharge from the rehabilitation centre. J Rehabil Med 2008; 40: 461-467.
42. Ellis R, Kosma M, Cardinal BJ, Bauer JJ, McCubbin JA. Physical activity beliefs and behaviour of adults with physical disabilities. Disabil Rehabil 2007; 29: 1221-1227.
43. Kerstin W, Gabriele B, Richard L. What promotes physical activity after spinal cord injury? An interview study from a patient perspective. Disabil Rehabil 2006; 28: 481-488.
44. Levins SM, Redenbach DM, Dyck I. Individual and societal influences on participation in physical activity following spinal cord injury: a qualitative study. Phys Ther 2004; 84: 496-509.
45. Rimmer JH, Riley B, Wang E, Rauworth A, Jurkowski J. Physical activity participation among persons with disabilities: barriers and facilitators. Am J Prev Med 2004; 26: 419-425.
46. Wu SK, Williams T. Factors influencing sport participation among athletes with spinal cord injury. Med Sci Sports Exerc 2001; 33: 177-182.
47. Van der Ploeg HP, Streppel KR, van der Beek AJ, van der Woude LH, Vollenbroek-Hutten MM, van Harten WH et al. Counselling increases physical activity behaviour nine weeks after rehabilitation. Br J Sports Med 2006; 40: 223-229.
48. Van der Ploeg HP, Streppel KR, van der Beek AJ, van der Woude LH, Vollenbroek-Hutten MM, van Harten WH et al. Successfully improving physical activity behavior after rehabilitation. Am J Health Promot 2007; 21: 153-159.
49. Heary CM, Hennessy E. The use of focus group interviews in pediatric health care research. J Pediatr Psychol 2002; 27: 47-57.

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Chapter 9

Intervention 'Active Lifestyle and Sports Participation'.

Promoting Physical Activity in Adolescents and Young Adults with Childhood-Onset Physical Disabilities

Abstract

Objective: Previous studies showed that many people with childhood-onset physical disabilities have low levels of physical activity (PA) and fitness. This may have serious consequences for health. Nevertheless health promoting efforts are limited. This paper presents the design and procedures of the intervention 'Active Lifestyle and Sports Participation' which is offered in the outpatient rehabilitation department for young adults of Erasmus MC and Rijndam Rehabilitation Centre. Furthermore, two case reports are presented. The intervention aims to improve levels of physical activity and fitness.
Subjects: Adolescents and young adults with childhood-onset physical disabilities.
Methods: The main approach of the intervention consists of personalized tailored counselling sessions with a PA counsellor who serves as a personal coach. A tailored PA advice is given, and together with the personal coach, participants discuss their involvement in other components of the intervention, which are sports participation, by means of tailored sports advice and sport specific training, and fitness training.
Main outcome measures are self-reported levels of PA, using two self-reports, and fitness, as evaluated by a sub-maximal 6 minutes walk or wheel test, and a maximal exercise test on an arm or cycle ergometer. Furthermore, participants rate their satisfaction with the intervention on a numeric scale from 0 to 10.
Results: This paper presents case reports of a 17-year old man with myelomeningocele and a 23 year old woman with unilateral cerebral palsy. Both participants improved their PA levels, and one of them improved the level of fitness. Furthermore, both participants were satisfied with the intervention.
Conclusion: The intervention 'Active Lifestyle and Sports Participation' seems to be a promising intervention to increase levels of PA and fitness of adolescents and young adults with a childhood onset physical disability. Future studies on its effectiveness are warranted.

Introduction

The majority of children who is born with a physical disability, such as myelomeningocele (MMC) or cerebral palsy (CP), or who acquired a physical disability early in childhood, such as traumatic brain injury (TBI) or spinal cord injury (SCI), now survive into adulthood.[1-3] During adolescence and young adulthood, people develop their own lifestyle, and encouraging healthy lifestyles at these ages may be important to prevent secondary conditions and to ensure a healthy adult life.[4] Physical activity (PA) and fitness are important health components: sufficient levels of PA and fitness are important for disease prevention, maintenance of functional independence, participation, social integration and life satisfaction.[5-7] Low levels of PA and fitness were found in adolescents and young adults with MMC and CP.[8-10] Nevertheless, health promotion efforts by increasing PA among people with physical disabilities are limited.[4]

Health promotion in rehabilitation

Rehabilitation medicine can be an important partner in health promotion for people with physical disabilities. It can offer the collective expertise in client centeredness at the individual level, Rehabilitation professionals can be important educators and coaches for behavioural change, and many facilities for sports and other physical activities are available.[4,11]

In the Netherlands, a previous study of Van der Ploeg and co-workers showed that a rehabilitation intervention can successfully improve levels of PA on the short and long term.[12,13] They used two PA promotion programmes, focusing on people who acquired physical disabilities: (i) 'Rehabilitation and Sports' (R&S), which provided tailored sports advice including sports locations near peoples home, and aimed to improve sport participation after discharge from the rehabilitation centre, and (ii) 'Active after Rehabilitation' (AaR), which aimed to improve levels of PA in daily life and used personalised tailored counselling, based on the stages of behavioural change concept of the Transtheoretical model.[14] This model describes the stages a person goes through (precontemplation, contemplation, preparation, action, maintenance, relapse) when changing his or her health behaviour. Interventions focusing on the promotion of health behaviour should approach people in each stage of change in a different stage-specific way.[14,15] Van der Ploeg and co-workers showed that only the combination of R&S and AaR increased sport participation and daily PA behaviour.[12] Based on these results, we integrated both personalised tailored PA counselling and tailored sports advice into the 'Active Lifestyle and Sports Participation' intervention for adolescents and young adults with a childhood onset physical disability. Fitness training was also incorporated in the intervention to provide the opportunity to improve fitness levels in addition to PA levels. In this paper, we present the development and procedures, including standardized assessments, of the intervention 'Active Lifestyle and Sports Participation', which is offered in the outpatient rehabilitation department for young adults of Erasmus MC University Medical Centre and Rijndam Rehabilitation Centre, both in Rotterdam, the

Netherlands. The intervention aims to increase levels of PA and fitness of adolescents and young adults with a childhood onset physical disability, and attempts to overcome main barriers to PA that have been recognized in this population, such as lack of information on how and where to exercise, lack of motivation, and lack of muscle strength and endurance.[16] In addition, this paper presents case reports of two persons who received the intervention.

Methods

Development of the intervention 'Active Lifestyle and Sports Participation'

The intervention is developed for adolescents and young adults aged 15 to 25 years with childhood-onset physical disabilities without severe learning disabilities and without contra-indications for maximal exercise (e.g. cardiac arrhythmias). The aim of the intervention is to improve their levels of daily PA and physical fitness. Adolescents and young adults with physical disabilities prefer to participate in their own goal setting and to have their viewpoints and concerns taken seriously.[17] Therefore, in health care, it is important to take into account the wishes, desires, knowledge base, capabilities, and rights of the person involved.[18] Personalised tailored counselling provides a good method to accommodate this. Therefore, the main approach of the 'Active Lifestyle and Sports Participation' intervention consists of personalised tailored counselling sessions with a PA counsellor (e.g. physical therapist, movement therapist), who serves as a person's 'personal coach'. Together with the personal coach, participants discuss their involvements in other components of the intervention, which are sports participation (tailored sport advice and sport specific training) and fitness training, taking into account possible overuse injuries or cardiovascular limitations. This way, they will conduct a personalised programme, tailored to their needs.

Personalised tailored physical activity counselling

Similar to the AaR programme,[12,13] persons receive 4 individual counselling sessions. The AaR programme is based on the stages of change concept of the Transtheoretical model.[14] This concept divides participants into the precontemplation, contemplation, preparation, action, and maintenance stage of the PA behaviour change.[14] In the first session of approximately 45 minutes, possibilities, barriers and facilitators of PA are identified, leading to a tailored PA advice, supported by a brochure specific to stage of PA change according to materials from EMGO institute VUmc[a] and Health Partners and Health Behavior group[b].[12] Three other 15-20 minutes counselling sessions are completed during face-to-face contact (preferably), or by telephone at 4, 8 and 12 weeks after the first session. In these sessions, the current PA level is discussed and attention is paid to find possible solutions for barriers and to accommodate the use of new PA possibilities. The stage of PA change is assessed in all sessions and when individuals move to another stage, they additionally receive the brochure relating to that stage.[12] Table 1 shows a time schedule for the different parts of the intervention.

Sports participation

This part of the intervention includes tailored sports advice and sport specific training. Sports advice is based on the R&S programme which consists of 2 to 4 structured counselling sessions with a sport counsellor.[12,13] If a person requires sports advice, the personal coach refers him or her to the sports counsellor and an appointment is made within 2 weeks. During the first session of approximately 30 minutes, sport history, preferences, possibilities, barriers and facilitators are identified. This results in a tailored sports advice from the sport counsellor, including information on available, accessible and appropriate sports facilities in the person's living environment. Aspects as transportation, finances, and supervision are also taken into account to ensure long term effects. The second session is after 4 weeks, during which sports participation and the satisfaction with the advice are evaluated, and additional advice can be given. If needed, the participant can follow a third and fourth session. In sport specific training, we offer participants the opportunity to be introduced to several movement activities and different sports adjusted to their possibilities, as well as the ability to practice. This way, participants are able to experience which sports they like and prefer, and are able to perform.

Table 1 Time schedule of the 'Active Lifestyle and Sports Participation' intervention.

Week	Assessment	PA counselling	Sports advice	Fitness training	
				Department	*Home*
1	Pre-test				
2		PA session 1			
3			Sports Session 1		
4				Training 1	Training 2
5				Training 3	Training 4
6		PA session 2		Training 5	Training 6
7			Sports Session 2	Training 7	Training 8
8				Training 9	Training 10
9				Training 11	Training 12
10		PA session 3		Training 13	Training 14
11			(Sports Session 3)	Training 15	Training 16
12				Training 17	Training 18
13				Training 19	Training 20
14		PA session 4		Training 21	Training 22
15			(Sports session 4)	Training 23	Training 24
16	Post-test				

PA= physical activity.

Fitness training

This part of the intervention is considered important for people with a low aerobic fitness (i.e. peakVO_2). Besides being an important risk factor for secondary conditions as cardiovascular disease,[19-22] low aerobic fitness is associated with high physical strain during the performance of daily activities,[23] which may cause early fatigue and discomfort, exaggerating the barrier to increase levels of PA. Furthermore, higher fitness may benefit participation and quality of life.[7,24] In a previous study in adolescents and young adults with MMC, we found an association between levels of PA and aerobic fitness in those with a very low fitness.[10] This suggests that a minimum level of aerobic fitness may be a requirement for being physically active during the day.[10] Increased aerobic fitness may therefore break through the vicious cycle of low PA leading to low fitness and further reduction in PA levels. Furthermore, in people with physical disabilities, muscle strength was associated with aerobic fitness, and it has been suggested that adding strength exercises to aerobic training has additional value in increasing aerobic fitness.[25-29] Therefore, fitness training consisting of aerobic exercises, as well as strength exercises is offered in a 12 week programme, including 1 one-hour group session per week at the rehabilitation department and take home exercises for once a week.

Intensity of aerobic training is determined using the Karvonen formula[30] that uses the peak heart rate (peakHR) obtained from baseline maximal exercise test and the heart rate (HR) at rest (restHR) to calculate the training heart rate (trainingHR).[31] During the 12-week training programme, the duration of aerobic exercises increases from 15 to 30 minutes. The training intensity starts at a HR of HRrest + 40% of the maximum heart rate reserve (HRR= peakHR – restHR) to a maximum of 80%, using ergometers (cycle, arm, rowing or cross-country).

Strength exercises focus on the large muscle groups. The training load is based on a percentage of the baseline 1-Repetition Maximum (1-RM),[31] depending on the training status of the participant. The 1-RM refers to the maximum load that a person is able to lift once. Training intensity starts with one set of 10-15 repetitions of 30% of the 1-RM during the first week and increases towards 3 sets of 15 to 20 repetitions of 75% RM in week 12.

Several fitness equipments are available for strength exercises of the upper extremity (En-Q[c] for pull down and push-up, EN-Tree Pulley[c] for proprioceptive neuromuscular facilitation (PNF) exercises of the shoulder), lower extremity (EN-Dynamic Seated Leg Press[c]) and abdominal exercises (EN-Dynamic Abdominal Trainer[c]). Also hand weights are available for additional biceps and triceps exercises. At home, similar strength exercises can be performed with a Thera-Band[d]. Resistance can be increased by shortening the band or doubling the band. This basic framework can be adjusted to the individual capabilities to ensure that it is a challenge for each participant.

Procedures

Participants enrol into the program after referral by a Rehabilitation physician of the outpatient Rehabilitation department for young adults of Erasmus MC or Rijndam Rehabilitation Centre. Before referral, the Rehabilitation physician checks whether it is safe for a participant to undergo a maximal exercise test.

Individual tailoring of the intervention requires assessments of relevant information at the start of the intervention. Therefore, before the start of the programme, pre-test measurements of PA and fitness levels takes place in the human performance laboratory at the Rehabilitation department of Erasmus MC, in addition to assessments of personal and condition-related characteristics. Furthermore, before the maximal exercise test, the Checklist Physical Activity Readiness Questionnaire (PAR-Q) is administered in the laboratory.[32,33] Participants are requested to refrain from smoking or caffeine consumption 3 hours prior to the measurements. The Medical Ethics Committee of Erasmus MC and Rijndam Rehabilitation Centre approved the study design and procedures, and informed consent procedure.

The feasibility of the intervention is evaluated using primary outcome measures of the intervention, i.e. levels of PA and fitness. Furthermore, the participant's overall satisfaction with the intervention is rated on a numeric scale from 0 (very unsatisfied) to 10 (very satisfied). Secondary outcome measures include assessments of body composition and blood pressure.

Measurements

Ambulatory status

The Hoffer criteria are used to indicate level of ambulation.[34] Community ambulators walk indoors and outdoors for most of their activities and may use a wheelchair only for long trips. Household ambulators walk only indoors. Non-functional ambulators are the so-called exercise walkers, and only walk during therapy sessions. Non-ambulators are persons who are wheelchair bound. In addition, an additional category of normal ambulators[35] is added to define participants who do not use walking aids or a wheelchair at all. For people with CP, we used the Gross Motor Functioning Classification System (GMFCS),[36] in which levels of motor functions are identified based on functional limitations, the need for assistive devices and, to a lesser extent, quality of movement.[36,37]

Educational level

The highest educational level of each participant is recorded.

Body composition

Body height of non-ambulators is measured with a flexible tape while participants are lying. In case of contractures, measurements are performed from joint to joint. Body height of ambulators is measured during standing. Body weight of ambulatory persons is obtained while standing on a Seca scale, and of non-ambulatory persons while sitting on an electronic scale.[e] Body mass index (BMI) is calculated by dividing the body weight (in kg) by height (in m) squared.

Waist circumference of ambulatory and non-ambulatory participants is measured with a flexible tape during standing or lying, respectively.

Thickness of four skin-folds (biceps, triceps, subscapular, and suprailiac) is measured twice on the right side of the body with a Harpenden calliper,[f] and mean values are used for further analyses. If the two measurements of each skin-fold differ by more than 1 mm, a third measurement is performed, and skin-fold thickness is taken as the average over the three measurements.

Blood pressure

Blood pressure is measured with an indirect method while participants are seated for at least 10 minutes prior to the measurements. A standard pressure cuff is placed around the upper arm. Systolic blood pressure (SBP) and diastolic blood pressure (DBP) are measured twice using a sphygmomanometer (Maxi-Stabil 3[g]), and the lowest values are recorded.

Smoking behaviour

The number of cigarettes smoked per day is assessed by questionnaire.

Alcohol consumption

The average number of alcohol consumptions per week is assessed by questionnaire.

Physical activity

Self-reported PA is assessed during a structured interview using the physical activity scale for individuals with physical disabilities (PASIPD).[38] The Dutch version of the PASIPD is a 12 item, seven day recall questionnaire including leisure time, household and work-related physical activities, from which a total physical activity score in kJ/kg body weight/day, can be calculated. Previous studies reported a Cronbach alpha of 0.60 for total score, acceptable test-retest reliability and a criterion validity that was comparable to other questionnaires.[38,39]

Self-reported PA level will also be assessed on a horizontal Visual Analogue Scale.[40] Participants will be instructed to rate their own PA level on a 100mm line in which 0 means not active at all, and 100 means very active.

Aerobic Fitness

Ambulatory participants perform a sub-maximal 6 min walk test.[41] Participants will be instructed to walk, not run, as far as they can along a 30-m marked tape in an open gym area for 6 minutes. Non-ambulatory participants perform the same test in a wheelchair. Participants are allowed to stop and rest during the test, but are instructed to resume walking or wheeling as soon as they feel able to do so. The 6 min walk or wheel distance (6MWD) is used as indicator of sub-maximal exercise capacity.

After the 6 min walk or wheel test, aerobic fitness is measured during a progressive maximal exercise test. According to Bhambhani and co-workers who studied persons with cerebral palsy, the main mode of ambulation elicits the highest oxygen uptake.[42] Therefore, depending on whether the main mode of ambulation is walking or wheelchair driving, participants perform the test on electronically braked cycle (Jaeger ER800[h]) or arm ergometer (Jaeger ER800SH[h]).[29] The test is preceded 5 min rest while seated, to obtain resting values, followed by a 3 min warm-up (5 Watt for arm ergometry and 20 Watt for cycle ergometry). During the test, every minute, the resistance increases with 10, 12, 15, 17 or 20 W per minute during cycle ergometry and with 5, 7, 10, or 12 W per minute during arm ergometry, depending on the capabilities of the participant. The pedal/crank rate for both arm and cycle ergometry is 60 to 70 rpm and strong verbal encouragement is given throughout the test. The test terminates when the participant stops due to exhaustion or when the participant is unable to maintain the pedal/crank rate. Gas exchange is measured continuously using a breath-by breath measurement system (Oxycon Pro[j]). Calibration with reference gases are performed before each test. Heart rate is measured continuously using a heart rate (HR) monitor (Polar[k]) which is attached to the system, and participants are fitted with a transmitter belt around the chest. Aerobic fitness is defined as the mean oxygen uptake during the last 30 s of exercise (peakVO_2). Peak work load (peakW) is defined as the highest work load maintained for at least 30s. HR and respiratory exchange ratio (RER) are used as objective criteria for maximal exercise. Subjective strain is measured immediately after the final stage using the modified Borg scale of rating perceived exertion (RPE), which is a vertical scale labelled from 0 (no effort at all) to 10 (maximal effort).[43,44]

Results: Case Reports

This section describes case reports of two participants who enrolled into the intervention and underwent the assessment procedures as described above.

Participant A was a 17 year old man, diagnosed with MMC and hydrocephalus (non-functional ambulator). He lives with his parents and attends pre-vocational theoretical education. He smokes 6 cigarettes per day and has approximately 4 alcohol consumptions per week. Participant B was a 23 year old woman, diagnosed with CP (GMFCS level 1, walking indoors and outdoors without limitations). She attends university, and lives independently in a student house. She is non-smoker and has 2 alcohol consumptions per week, on average. Table 2 presents baseline assessments of both participants. Participant A performed the maximal exercise test on an electronically braked arm ergometer in which the resistance increased with 10 W per minute. Participant B performed the maximum exercise test on an electronically-braked cycle ergometer in which the resistance increased with 20 W per minute.

Table 2 Baseline assessments of participant A and B.

	Participant A	Participant B
Gender	Man	Woman
Age	17	23
Ambulatory status	Non-functional ambulator	GMFCS 1
Anthropometry		
Height (cm)	176	162
Weight (kg)	90.0	63
BMI (kg/m^2)	29.1	24
Waist circumference (cm)	92	77
Sum 4 skin folds (mm)	114	77
Blood pressure		
SBP (mmHg)	140	130
DBP (mmHg)	78	70
Physical activity		
PASIPD, total score kJ/kg/day	43.0	33.9
VAS (mm)	76	19
Fitness		
Distance 6MWD (m)	698	507
PeakVO_2 (l/min)	1.94	2.20
PeakPO (W)	120	180
Rest HR (bpm)	84	88
PeakHR (bpm)	184	179
PeakRER	1.17	1.14
% of peakHR predicted	101	91
Rate of perceived exertion	8	5

Intervention

After the baseline measurement, both participants had a consultation with the personal coach. It appeared that participant A plays wheelchair basketball once a week and wheelchair tennis twice a week (Stage 5 'maintenance'), but he was interested in other possibilities to increase his level of daily PA and physical fitness. Together, the participant and the personal coach decided that he would attend the fitness training and visit the sports counsellor for sports advice and sport-specific training. He trained weekly at the Erasmus MC. In the fitness training sessions, he performed aerobic exercises on an arm ergometer (MOTOmed Viva 2[l]), and strength exercises for the abdomen and upper extremity muscles, see Table 3. Because he already participated in other wheelchair sports he did not want to do a second training session at home. He visited the sports counsellor three times. During sports advice, he appeared to be interested in hand biking, which he had never tried before. He found the hand bike a nice opportunity for future active transportation. Furthermore, he was interested in skiing and he will thus be approached by the rehabilitation centre when they arrange a daytrip to practice indoor skiing.

Table 3 Fitness training as followed by participant A.

	Aerobic training		**Strength exercises**							
	Arm ergometer		Abdominal muscle		Pull down		Push up		PNF exercises	
week	min	%HRR	%1-RM	Rep	%1-RM	Rep	%1-RM	Rep	%1-RM	Rep
1	15	54	55	3 x 15	49	3 x 15	58	3 x 15	50	3 x 15
2	15	51	55	3 x 15	49	3 x 15	58	3 x 15	50	3 x 15
3	20	53	58	3 x 15	-	-	58	3 x 15	55	3 x 15
4	×	×	×	×	×	×	×	×	×	×
5	20	56	61	3 x 20	49	3 x 15	58	3 x 15	59	3 x 15
6	×	×	×	×	×	×	×	×	×	×
7	20	66	65	3 x 15	49	3 x 20	58	3 x 15	64	3 x 10
8	20	76	67	2 x 20	65	3 x 10	-	-	64	3 x 10
9	25	76	70	3 x 15	-	-	-	-	68	3 x 15
10	25	76	73	3 x 15	73	3 x 10	75	3 x 8	-	-

min= duration in minutes. %HRR= percentage of heart rate reserve. %1-RM= Percentage of 1-Repetition Maximum. Rep= number of repetitions. PNF= proprioceptive neuromuscular facilitation.

Participant B stopped judo at the age of 17 due to a lack of time. Now, she would like to be more active and play sports again, but she did not know where to go and wanted to have more information about her possibilities. During the counselling sessions, she started to realize that it is important to have an active lifestyle. She became more conscious of how to become more physically active during the day, and, whenever possible, she used the bicycle for active transport and the stairs instead of the elevator. She improved from stage 2 (i.e. 'contemplation') to stage 4 (i.e. 'action'). In addition to PA counselling, she attended the fitness training and visited the sports counsellor for sports advice and sport-specific training. During her weekly fitness training at the Erasmus MC, she performed aerobic exercises on a cycle ergometer, and additional sessions on a rowing ergometer and/or cross country ergometer, and she performed strength exercises for the abdomen, upper and lower extremity muscles (see Table 3). At home, she was advised to go for a bicycle ride of comparable duration as during the training on the ergometer and to use the Theraband for upper extremity strength exercises. For strength of the lower extremity, she was instructed to stand against the wall with her knees and hips in 90 degrees flexion. She had 3 sessions for sports advice in which possibilities of hockey, badminton and rowing were discussed. Together with the sports counsellor, she arranged meetings to practice these sports. She realized that it is not necessary to be a good athlete to practice sports, and that it is fun to do. She now considers going to a badminton club. She mentioned that she felt better being more physically active, and that she has more energy for other activities.

Table 4 Fitness training as followed by participant B.

	Aerobic training				Strength exercises					
	Cycling		Rowing	Cross country	Abdominal muscle		Leg press		PNF exercises	
week	min	%HRR	min	min	%1-RM	Rep	%1-RM	Rep	%1-RM	Rep
1	10	45	5	5	50	3 x 15	50	3 x 15	55	3 x 15
2	15	40	5	5	54	3 x 15	55	3 x 15	64	3 x 12
3	20	56	6	6	60	3 x 15	58	3 x 15	64	3 x 15
4	20	68	6	-	60	3 x 15	65	3 x 15	68	3 x 15
5	20	80	6	6	65	3 x 15	67	3 x 15	73	3 x 15
6	20	81	6	6	65	3 x 15	70	3 x 15	73	3 x 15
7	×	×	×	×	×	×	×	×	×	×
8	20	85	6	-	75	3 x 15	73	3 x 15	77	3 x 15
9	×	×	×	×	×	×	×	×	×	×
10	20	89	6	6	69	3 x 15	69	3 x 15	82	3 x 15
11	20	70	6	-	69	3 x 15	73	3 x 15	82	3 x 15
12	20	78	6	-	69	3 x 15	75	3 x 15	82	3 x 15

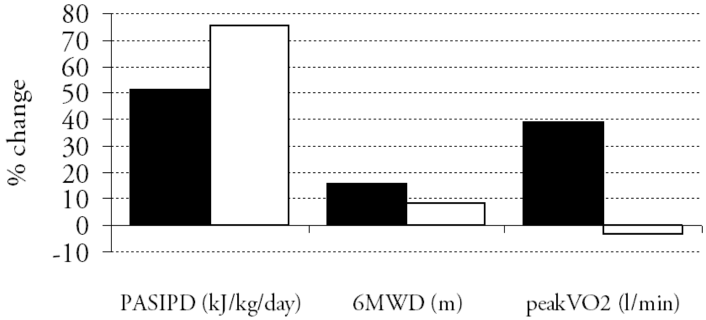
min= duration in minutes. %HRR= percentage of heart rate reserve. %1-RM= Percentage of 1-Repetition Maximum;. Rep= number of repetitions. PNF= proprioceptive neuromuscular facilitation.

Outcome

Changes in the main outcome measures after the intervention are presented in Figure 1. Self-reported PA as measured with the PASIPD increased from 43.0 to 65.0 kJ/kg/day in participant A and from 33.9 to 59.4 kJ/kg/day in participant B. Self-reported PA as rated on the VAS, increased from 76 to 97 in participant A and from 19 to 92 in participant B. Furthermore, the 6MWD increased with 16.0% in participant A and with 8.5% in participant B. Participant A also showed increased PeakVO_2 from 1.94 to 2.70 l/min. At baseline, his level of peakVO_2 was higher than the average values assessed during arm exercise of non-ambulatory males with MMC (1.57 l/min)[10] and males with paraplegic spinal cord injuries with lesions below T10 (1.98 l/min).[45] His PeakPO increased from 120 to 160W. Baseline peakVO_2 of participant B was close to reference values of females with the same age (2.3 l/min) assessed during cycle ergometry,[46] and did not improve after intervention. The secondary outcome measures body composition and blood pressure remained unchanged in both participants.

Both participants were satisfied with the intervention. Ratings were 10 for participant A and 7 for participant B. Both participants missed several training sessions (see Table 3 and 4) due to time constraints, transportation problems, illness, or holiday. However, participant A completed 67% of the training sessions at Erasmus MC and participant B 83%, indicating reasonable compliance.

Figure 1 Changes in physical activity and fitness after the intervention.



Black bars: Participant A. White bars: participant B.

Discussion

It appeared to be feasible to offer the intervention 'Active Lifestyle and Sports Participation' in the outpatient Rehabilitation department for young adults with a childhood onset physical disability. Regarding the main outcome measures, levels of self-reported PA, and sub-maximal exercise capacity seemed to have improved after the intervention in both participants, and aerobic fitness increased in one of them. Furthermore, both participants were satisfied with the intervention.

The substantial increase in aerobic fitness of participant A may be a direct result of the fitness training, but it may have also motivated him to train harder during his other sports. For participant B, the frequency of fitness training may not have been sufficient to improve aerobic fitness. Although self-reported levels of PA have increased, the results should be interpreted with caution because questionnaires are prone to overestimation of PA levels.[47] Furthermore, participant A may not have been a typical example of the population that needs PA counselling, as he was already physically active before he started the intervention. Nevertheless, he seemed to have increased his fitness, and for him it was a worthwhile experience to try out the hand bike.

Future directions

In the future, this intervention will also be offered in other rehabilitation centres in the Southwest of the Netherlands. In future evaluation of the intervention, we will include pre- and post-test assessments of a larger study sample and a control sample not receiving the intervention, to evaluate whether the intervention increases levels of PA and fitness. In addition, follow-up assessments are needed to obtain insight in long-term effectiveness of the intervention. Our future evaluation of the intervention will also include objective PA monitoring during 7 days using the GTM1 ActiGraph[m], which is a uni-axial accelerometer. Previous studies in a laboratory setting and in free-living conditions established its validity for ambulatory adults[48,49] and children,[50-52] and for wheelchair-use.[53,54]

Studies showed that endurance training may positively influence respiratory function.[55,56] Therefore, future evaluation will also include spirometry tests to assess respiratory function, focusing on the small airways (forced vital capacity (FVC) and forced expiratory volume in one second, FEV_1) and the large airways (peak expiratory flow, PEF). Furthermore, cost-effectiveness studies of the intervention are needed.

In conclusion, the intervention 'Active Lifestyle and Sports Participation' seems to be a promising intervention to increase levels of daily PA and physical fitness of adolescents and young adults with childhood-onset physical disabilities. Future studies on its effectiveness are warranted.

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Suppliers

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References

1. Strauss DJ, Shavelle RM, Anderson TW. Life expectancy of children with cerebral palsy. Pediatr Neurol 1998; 18: 143-149.
2. Bowman RM, McLone DG, Grant JA, Tomita T, Ito JA. Spina bifida outcome: a 25-year prospective. Pediatr Neurosurg 2001; 34: 114-120.
3. Oakeshott P, Hunt GM. Long-term outcome in open spina bifida. Br J Gen Pract 2003; 53: 632-636.
4. Rimmer JH. Health promotion for people with disabilities: the emerging paradigm shift from disability prevention to prevention of secondary conditions. Phys Ther 1999; 79: 495-502.

5. Heath GW, Fentem PH. Physical activity among persons with disabilities--a public health perspective. Exerc Sport Sci Rev 1997; 25: 195-234.
6. Durstine JL, Painter P, Franklin BA, Morgan D, Pitetti KH, Roberts SO. Physical activity for the chronically ill and disabled. Sports Med 2000; 30: 207-219.
7. Buffart LM, van den Berg-Emons RJG, van Meeteren J, Stam HJ, Roebroeck ME. Lifestyle, participation and health-related quality of life in adolescents and young adults with myelomeningocele. *Submitted*.
8. Bandini LG, Schoeller DA, Fukagawa NK, Wykes LJ, Dietz WH. Body composition and energy expenditure in adolescents with cerebral palsy or myelodysplasia. Pediatr Res 1991; 29: 70-77.
9. Maher CA, Williams MT, Olds T, Lane AE. Physical and sedentary activity in adolescents with cerebral palsy. Dev Med Child Neurol 2007; 49: 450-457.
10. 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.
11. Teague ML, Capriano RF, McGhee VL. Health promotion as a rehabilitation service for people with disabilities. J Rehabil 1990; 56: 52-56.
12. Van der Ploeg HP, Streppel KR, van der Beek AJ, van der Woude LH, Vollenbroek-Hutten MM, van Harten WH, van Mechelen W. Counselling increases physical activity behaviour nine weeks after rehabilitation. Br J Sports Med 2006; 40: 223-229.
13. Van der Ploeg HP, Streppel KR, van der Beek AJ, van der Woude LH, Vollenbroek-Hutten MM, van Harten WH, van Mechelen W. Successfully improving physical activity behavior after rehabilitation Am J Health Promot 2007; 21: 153-159.
14. Prochaska JO, DiClemente CC. Stages and processes of self-change of smoking: toward an integrative model of change. J Consult Clin Psychol 1983; 51: 390-395.
15. Marcus BH, Selby VC, Niaura RS, Rossi JS. Self-efficacy and the stages of exercise behavior change. Res Q Exerc Sport 1992; 63: 60-66.
16. Buffart LM, Westendorp T, van den Berg-Emons RJG, van Mechelen W, Stam HJ, Roebroeck ME. Barriers and facilitators of physical activity in adolescents and young adults with a physical disability. *Thesis: Chapter 8.*
17. Britto MT, DeVellis RF, Hornung RW, DeFriese GH, Atherton HD, Slap GB. Health care preferences and priorities of adolescents with chronic illnesses. Pediatrics 2004; 114: 1272-1280.
18. Michaud PA, Suris JC, Viner R. The adolescent with a chronic condition. Part II: healthcare provision. Arch Dis Child 2004; 89: 943-949.
19. Janssen TW, van Oers CA, van Kamp GJ, TenVoorde BJ, van der Woude LH, Hollander AP. Coronary heart disease risk indicators, aerobic power, and physical activity in men with spinal cord injuries. Arch Phys Med Rehabil 1997; 78: 697-705.
20. Dallmeijer AJ, Hopman MT, van der Woude LH. Lipid, lipoprotein, and apolipoprotein profiles in active and sedentary men with tetraplegia. Arch Phys Med Rehabil 1997; 78: 1173-1176.
21. Dallmeijer AJ, van der Woude LH, van Kamp GJ, Hollander AP. Changes in lipid, lipoprotein and apolipoprotein profiles in persons with spinal cord injuries during the first 2 years post-injury. Spinal Cord 1999; 37: 96-102.

22. Buffart LM, van den Berg-Emons RJG, Burdorf A, Janssen WGM, Stam HJ, Roebroeck ME. Cardiovascular disease risk factors and the relations with physical activity, aerobic fitness, and body fat in adolescents and young adults with myelomeningocele. Arch Phys Med Rehabil *in press*.
23. Bruinings AL, van den Berg-Emons HJ, Buffart LM, van der Heijden-Maessen HC, Roebroeck ME, Stam HJ. Energy cost and physical strain of daily activities in adolescents and young adults with myelomeningocele. Dev Med Child Neurol 2007; 49: 672-677.
24. Tomas-Carus P, Hakkinen A, Gusi N, Leal A, Hakkinen K, Ortega-Alonso A. Aquatic Training and Detraining on Fitness and Quality of Life in Fibromyalgia. Med Sci Sports Exerc 2007; 39: 1044-1050.
25. Cooney MM, Walker JB. Hydraulic resistance exercise benefits cardiovascular fitness of spinal cord injured. Med Sci Sports Exerc 1986; 18: 522-525.
26. Janssen TW, van Oers CA, Hollander AP, Veeger HE, van der Woude LH. Isometric strength, sprint power, and aerobic power in individuals with a spinal cord injury. Med Sci Sports Exerc 1993; 25: 863-870.
27. Zoeller RF, Jr., Riechman SE, Dabayebeh IM, Goss FL, Robertson RJ, Jacobs PL. Relation between muscular strength and cardiorespiratory fitness in people with thoracic-level paraplegia. Arch Phys Med Rehabil 2005; 86: 1441-1446.
28. Haisma JA, Bussmann JB, Stam HJ, Sluis TA, Bergen MP, Dallmeijer AJ, de Groot S, van der Woude LH. Changes in physical capacity during and after inpatient rehabilitation in subjects with a spinal cord injury. Arch Phys Med Rehabil 2006; 87: 741-748.
29. 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: 181-188.
30. Karvonen J, Vuorimaa T. Heart rate and exercise intensity during sports activities. Practical application. Sports Med 1988; 5: 303-311.
31. May AM, Van Weert E, Korstjens I, Hoekstra-Weebers JE, Van Der Schans CP, Zonderland ML, Mesters I, Borne BV, Ros, WJ. Improved physical fitness of cancer survivors: A randomised controlled trial comparing physical training with physical and cognitive-behavioural training. Acta Oncol 2007: 1-10.
32. Shephard RJ, Cox MH, Simper K. An analysis of "Par-Q" responses in an office population. Can J Public Health 1981; 72: 37-40.
33. Thomas S, Reading J, Shephard RJ. Revision of the Physical Activity Readiness Questionnaire (PAR-Q). Can J Sport Sci 1992; 17: 338-345.
34. 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.
35. 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: 420-427.
36. Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Development and reliability of a system to classify gross motor function in children with cerebral palsy. Dev Med Child Neurol 1997; 39: 214-223.
37. Wood E, Rosenbaum P. The gross motor function classification system for cerebral palsy: a study of reliability and stability over time. Dev Med Child Neurol 2000; 42: 292-296.
38. Washburn RA, Zhu W, McAuley E, Frogley M, Figoni SF. The physical activity scale for individuals with physical disabilities: development and evaluation. Arch Phys Med Rehabil 2002; 83: 193-200.

39. Van der Ploeg HP, Streppel KR, van der Beek AJ, van der Woude LH, Vollenbroek-Hutten M, van Mechelen W. The Physical Activity Scale for Individuals with Physical Disabilities: test-retest reliability and comparison with an accelerometer. J Phys Act Health 2007; 4: 96-100.
40. Scott J, Huskisson EC. Vertical or horizontal visual analogue scales. Ann Rheum Dis 1979; 38: 560.
41. Guyatt GH, Sullivan MJ, Thompson PJ, Fallen EL, Pugsley SO, Taylor DW, et al. The 6-minute walk: a new measure of exercise capacity in patients with chronic heart failure. Can Med Assoc J 1985; 132: 919-923.
42. Bhambhani YN, Holland LJ, Steadward RD. Maximal aerobic power in cerebral palsied wheelchair athletes: validity and reliability. Arch Phys Med Rehabil 1992; 73: 246-252.
43. Borg GA. Psychophysical bases of perceived exertion. Med Sci Sports Exerc 1982; 14: 377-381.
44. Mahler DA, Rosiello RA, Harver A, Lentine T, McGovern JF, Daubenspeck JA. Comparison of clinical dyspnea ratings and psychophysical measurements of respiratory sensation in obstructive airway disease. Am Rev Respir Dis 1987; 135: 1229-1233.
45. Janssen TW, Dallmeijer AJ, Veeger DJ, van der Woude LH. Normative values and determinants of physical capacity in individuals with spinal cord injury. J Rehabil Res Dev 2002; 39: 29-39.
46. Vos JA. Ergometrie en Trainingsbegeleiding. 4 ed. Amersfoort: Nederlands Paramedisch Instituut 2001.
47. Sallis JF, Saelens BE. Assessment of physical activity by self-report: status, limitations, and future directions. Res Q Exerc Sport 2000; 71: S1-14.
48. Melanson EL, Jr., Freedson PS. Validity of the Computer Science and Applications, Inc. (CSA) activity monitor. Med Sci Sports Exerc 1995; 27: 934-940.
49. Freedson PS, Melanson E, Sirard J. Calibration of the Computer Science and Applications, Inc. accelerometer. Med Sci Sports Exerc 1998 ;30: 777-781.
50. Janz KF. Validation of the CSA accelerometer for assessing children's physical activity. Med Sci Sports Exerc 1994; 26: 369-375.
51. Trost SG, Ward DS, Moorehead SM, Watson PD, Riner W, Burke JR. Validity of the computer science and applications (CSA) activity monitor in children. Med Sci Sports Exerc 1998; 30: 629-633.
52. De Vries SI, Bakker I, Hopman-Rock M, Hirasing RA, van Mechelen W. Clinimetric review of motion sensors in children and adolescents. J Clin Epidemiol 2006; 59: 670-680.
53. Washburn RA, Copay AG. Assessing physical activity during wheelchair pushing: validity of a portable accelerometer. Adapt Phys Activ Q 1999; 16: 290-299.
54. Warms CA, Belza BL. Actigraphy as a measure of physical activity for wheelchair users with spinal cord injury. Nurs Res 2004; 53: 136-143.
55. Silva AC, Neder JA, Chiurciu MV, Pasqualin DC, da Silva RC, Fernandez AC, Lauro FA, de Mello MT, Tufik S. Effect of aerobic training on ventilatory muscle endurance of spinal cord injured men. Spinal Cord 1998; 36: 240-245.
56. Le Foll-de Moro D, Tordi N, Lonsdorfer E, Lonsdorfer J. Ventilation efficiency and pulmonary function after a wheelchair interval-training program in subjects with recent spinal cord injury. Arch Phys Med Rehabil 2005; 86: 1582-1586.

Chapter 10

General Discussion

The aim of this thesis was to obtain insight in levels of daily physical activity and fitness of adolescents and young adults with myelomeningocele (MMC) and to study the associations with personal and environmental factors. Furthermore, we studied whether levels of daily physical activity and physical fitness were associated with each other and with participation and health-related quality of life, and the risk of developing cardiovascular disease later in life.

This chapter will start with the main findings that have been presented in the previous chapters. Furthermore methodological issues related to the study will be discussed, as well as clinical implications of the study and suggestions for future research.

Main findings

Physical activity, fitness and associated factors

A large proportion of adolescents and young adults with myelomeningocele (MMC) were physically inactive, had low aerobic fitness and high levels of body fat (Chapter 2). Non-ambulators generally had lower levels of physical activity, lower aerobic fitness and higher body fat compared to ambulators. Females had lower aerobic fitness and more body fat than males, but we found no gender differences in levels of physical activity.

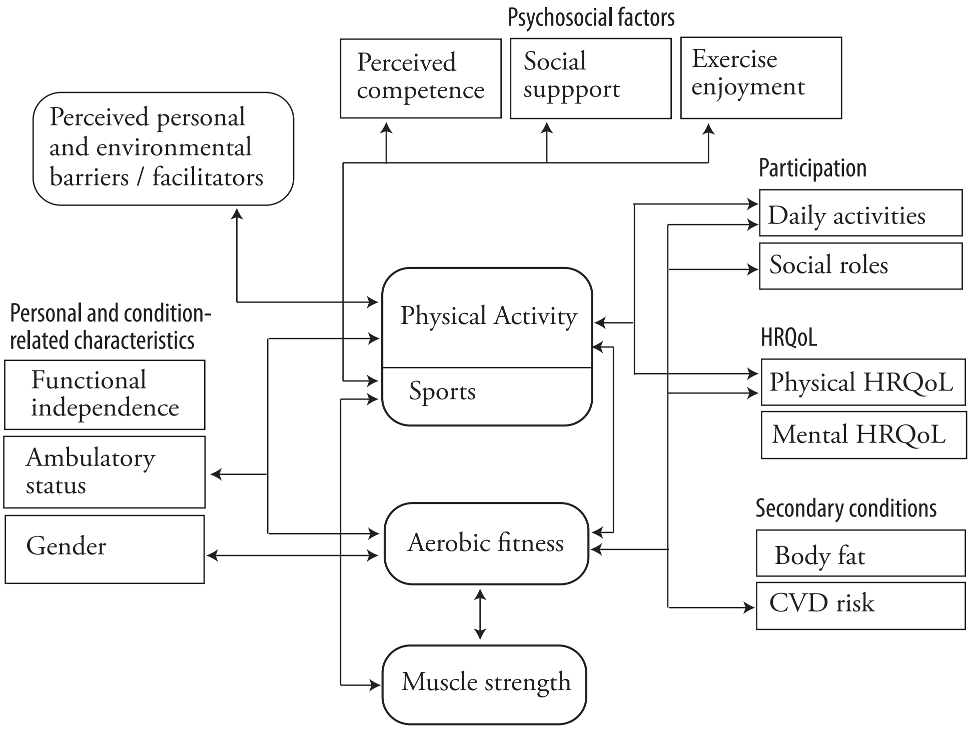
Factors associated with physical activity and fitness are summarized in Figure 1. Chapter 2 showed that levels of physical activity and aerobic fitness were not associated with body fat, suggesting that other strategies, such as reducing energy intake, may be more important in reducing body fat than increasing levels of physical activity and aerobic fitness. An association between levels of physical activity and aerobic fitness was only found in non-ambulatory persons, who generally have very low levels of aerobic fitness. This may suggest that a minimum level of aerobic fitness is needed to be able to be physically active during the day. Other factors are also likely to play a role in physical activity behaviour since > 70% of the variance was left unexplained. Many personal and environmental factors can be perceived as a barrier or facilitator of physical activity (Chapter 8), and it is likely that parameters such as self-efficacy, social support, lack of motivation, not knowing where to exercise and lack of facilities play a role (Chapter 5 and 8).

Half of the variance in aerobic fitness was explained by gender and ambulatory status (Chapter 3). In addition, we found a small but significant association between aerobic capacity and muscle strength, suggesting that adding strength training to aerobic training may be of additional value in increasing aerobic capacity.

Energy cost per unit time of locomotion and household activities in ambulatory adolescents and young adults with MMC was comparable with able-bodied persons of the same age; energy cost per unit time during wheelchair driving was lower (Chapter 4). However, because of low aerobic capacity, persons with MMC performed these activities at a high percentage of their aerobic capacity, indicating high physical strain. This suggests that increasing aerobic capacity may reduce physical strain of daily activities.

Two third of the participants engaged in sports (Chapter 5). Sports participation was not associated with ambulatory status, the presence of hydrocephalus or functional independence, which supports the opinion that sports participation is due to personal preferences rather than physical ability. Social support from family and friends, perceived competences (athletic competence, physical appearance and global self worth) and exercise enjoyment showed positive associations with sports participation and may serve as important facilitators to target in interventions. Participating in sports is a way to increase physical activity behaviour of adolescents and young adults with MMC; it contributed to 16% of total physical activity behaviour. Sports participants may have higher levels of physical fitness than non-participants, but conclusive evidence is needed.

Figure 1 Factors associated with physical activity and fitness in adolescents and young adults with myelomeningocele.



Benefits of physical activity and aerobic fitness

Chapter 6 and 7 provided some evidence that health care should incorporate improving levels of physical activity and aerobic fitness because it may benefit participation and health-related quality of life. Furthermore, a higher level of aerobic fitness was associated with better cardiovascular health. Of adolescents and young adults with MMC, 63% were restricted in daily activities and 59% in social participation (Chapter 6). They had poorer health-related quality of life than the Dutch reference population regarding physical functions, but not regarding mental functions. Results in Chapter 6 showed that higher levels of physical activity and aerobic fitness were associated with a better participation and a better physical health-related quality of life.

Average lipid and lipoprotein profiles of adolescents and young adults with MMC did not differ from those found in the general male and female Dutch population of similar ages (Chapter 7). Nevertheless, elevated levels of total cholesterol, low-density lipoprotein cholesterol and triglycerides were found in 29%, 38% and 3% of MMC participants respectively. High-density lipoprotein profile was reduced in 19%. Hypertension was found in 20% and 19% were current cigarette smoker. Multiple risk factors were present in a large proportion (42%) of adolescents and young adults with MMC, leaving them at risk of developing cardiovascular disease (CVD). Non-ambulatory persons with MMC were more likely to be at CVD risk than ambulatory persons with MMC. Individuals with lower aerobic fitness tended to be more likely to be at CVD risk. No associations were found between CVD risk and levels of physical activity and body fat. This suggests that increasing aerobic fitness may benefit cardiovascular health.

Methodological issues

Response rate and generalizability

This was a multi-centre study in which participants aged 16 to 30 years were recruited from 4 university hospitals from the Netherlands (Rotterdam, Leiden, Utrecht and Amsterdam), and all rehabilitation centres from the southwest of the Netherlands (Rijndam Rehabilitation Centre Rotterdam, Sophia Rehabilitation The Hague and Delft, Rijnlands Rehabilitation Centre Leiden, Rehabilitation Centre 'de Waarden' Dordrecht and Foundation for Rehabilitation Medicine Zeeland Goes). These regions covered the most densely populated part of the country. However, the generalization of the study is hampered by the low response rate; of the 171 people who were invited, only 51 participated in the study, which is a response rate of 30%.

We checked whether responders and non-responders differed regarding several personal and condition-related characteristics (see Chapter 2, Table 1). Age, gender, level of lesion and the presence of hydrocephalus did not differ between responders and non-responders which support generalization. In addition, high prevalence of middle-level (lumbosacral) and high-level (lumbar or thoracolumbar) lesions was in line with participants from the national Adolescents with SPina Bifida in the Netherlands (ASPINE) study in which 119 people with spina bifida aperta and hydrocephalus participated.[1,2] The ASPINE study addressed a fairly representative sample of adolescents and young adults with spina bifida in the Netherlands; patients were recruited from 11 of the 12 multidisciplinary spina bifida teams from the Netherlands as well as from the Association of Physically Disabled Persons and their parents (BOSK). Considering the similarity of personal and disease-related characteristics between responders and non-responders and between our study sample and the sample from the ASPINE study, we assumed the study sample to be sufficiently representative.

Nevertheless, there may have been a selection bias that the more active and fitter people had higher interest in participating in this study. Of the participants, 69% participated in sports (Chapter 5). This percentage is higher than 53% found in people with various physical disabilities one year after rehabilitation.[3] Furthermore, adolescents and young adults who participated in this study perceived better physical functioning and less pain compared to people with spina bifida aperta and hydrocephalus who participated in the national ASPINE study (Chapter 6). This selection bias may have resulted in an overestimation of levels of physical activity and fitness. However, main reasons for non-participation in the study were no interest in the study, lack of time, duration and extensiveness of the measurements, or feeling uncomfortable with the 48-hour measurement with the activity monitor (AM), rather than physical functioning.

Study design and statistical power

The cross-sectional design of the study limits the ability of being conclusive about benefits of improving levels of physical activity and aerobic fitness on functioning and health. This applies to the findings that individuals with higher levels of physical activity and/or aerobic fitness perceived less participation restrictions and a better health-related quality of life (Chapter 6) and were more likely to have no cardiovascular disease risk (Chapter 7). In addition, the findings that physical activity and aerobic fitness were not associated with body fat (Chapter 2) suggest that it is unlikely that increasing levels of activity and aerobic fitness would reduce body fat.

Because some dependent variables were dichotomous or in some cases were dichotomized because of non-normally distributed data, we used logistic regression analyses (Chapters 5, 6 and 7) to determine relationships among variables. However, logistic regression has limited statistical power in small sample sizes. Although a sample size of 51 is relatively large considering the prevalence of the condition, the absolute number is quite small. Some studies were based on a sub-group of this sample (Chapter 4 and 7) and thus included even smaller sample sizes. Therefore, regarding some relationships, the sample size was barely sufficient to include a number of independent variables, and some conclusions will have to be interpreted with caution, as indicated by large confidence intervals.

Outcome measures

Physical activity

Because we measured physical activity objectively with the AM, we were restricted to 2 days of monitoring. Therefore, we were not able to characterize and individual's actual activity pattern which is assumed to require at least 7 days of activity monitoring.[4] However, the strength of the AM measurement is that it provides detailed objective information on mobility-related physical activities in both ambulatory and non-ambulatory persons. Some evidence has been published that a 24-hour measurement with the AM is of adequate duration to assess activity levels.[5] Furthermore, analyses of variance of the 48-hour measurements showed that the variance between the two measurement days was small enough allowing to distinguish active and inactive people (intraclass correlation coefficient= 0.89). Therefore, we assumed that a sample of 2 random days during the week was sufficient to adequately assess an individual's physical activity level.

Aerobic capacity

We used different exercise modes to assess aerobic capacity for ambulatory and non-ambulatory persons. Measurement in people with cerebral palsy showed that the main mode of ambulation elicits the highest oxygen uptake.[6] However, due to large differences in active muscle mass, arm and leg exercise have different physiological responses. Nevertheless, we pooled the data from arm and leg exercise, because we assumed that we measure a person's highest measurable oxygen uptake (peakVO_2). In the analyses we corrected for differences in exercise mode by using ambulatory status as proxy for type of ergometer. There was a high collinearity between type of ergometer and ambulatory status (r= 0.92; p< 0.001) and we found ambulatory status to be more clinically meaningful. Peripheral local fatigue may have caused exercise cessation before reaching maximum oxygen uptake. However, based on objective (heart rate and respiratory exchange ratio) and subjective criteria of maximal exercise (rate of perceived exertion), we consider the peakVO_2 a reasonable estimate of a person's peak performance (Chapter 3).

To be able to compare individuals, it is common practice to normalize values of peakVO_2 for body mass or fat free mass. However, throughout this thesis, we used absolute values of peakVO_2 because many persons with MMC have excessive body fat, particularly non-ambulatory persons, and therefore normalizing for body mass seemed inappropriate. We have considered normalizing peakVO_2 for fat-free mass; however, estimation of fat-free mass would be based on skin fold measurements using algorithms developed from an able-bodied reference population, which may not be applicable to the MMC population. In persons with cerebral palsy, these algorithms were be invalid.[7] In the analyses we used absolute values of peakVO_2 corrected for gender and ambulatory status which covers part of the differences in fat-free mass: ambulatory persons typically being taller than non-ambulatory persons and males typically having more fat-free mass than females (Chapter 2).

To limit the duration of all assessments, we did not measure ASIA motor levels of lesions. Instead, we obtained information on lesion level from medical records. From the ASPINE study it is known that this information might be unreliable because lesion levels were determined at different ages, sometimes using motor, sometimes using sensory levels, and sometimes, no description of the method was given.[8] Although ambulatory status is a more global measure, it is significantly correlated to lesion level (r= 0.58; p< 0.001) and therefore, ambulatory status, according to the classification of Hoffer,[9] was used as relevant clinical characteristic of people with MMC. Since ASIA data are lacking, we were however, unable to distinguish which part of low fitness in ambulatory persons was due to deconditioning and which part to reduced active muscle mass caused by paresis of lower extremity muscles.

Clinical implications

Results showed that many adolescents and young adults with MMC had low levels of physical activity and low fitness. In addition, higher levels of physical activity and aerobic fitness were associated with better participation and health-related quality of life. Higher aerobic fitness was also associated with a better cardiovascular health. With increasing life-expectancy of people with chronic conditions such as MMC, improving levels of physical activity and fitness deserves attention in health care to prevent secondary conditions and to ensure a healthy adult life.

Rehabilitation medicine has the potential to play an important role in promoting physical activity in people with chronic conditions. It can inform patients on important health benefits and on where and how to exercise, it can facilitate in equipment and programmes, and supervise and assist during exercise. Whereas a general practitioners could play an important role in improving physical activity behaviour in the general population,[10] a rehabilitation physician can fulfil this position in people with a childhood onset physical disability. This is recognized by the Netherlands Society of Rehabilitation and Physical Medicine (VRA), who recently started a project group 'Bewegen en Sport' (Physical activity and Sports) to emphasize the importance of physical activity and fitness for health, and to raise the focus on this topic in Rehabilitation medicine.

Interventions aiming to increase physical activity behaviour and fitness are warranted. Chapter 9 describes the design of the intervention: 'Active Lifestyle and Sports Participation', an intervention aiming to improve levels of physical activity and fitness in adolescents and young adults with childhood-onset physical disabilities. Together with a 'personal coach,' each participant conducts a personalised programme tailored to his or her needs. Previously, the study of van der Ploeg and co-workers showed that the combination of both personalised tailored physical activity counselling and tailored sports advice increased sport participation and daily physical activity behaviour after rehabilitation in people with physical disabilities.[3,11] Therefore, the integration of both approaches formed the base of the intervention. Furthermore, other results of this thesis have contributed to the design of the intervention. Several personal and environmental barriers and facilitators of physical activity and sports participation have been identified in Chapter 5 and Chapter 8. The intervention focuses at overcoming the barriers to become more physically active.

Furthermore, we included fitness training as a component of the intervention. Chapter 7 shows that low aerobic fitness was associated with higher risk of cardiovascular disease, which is in line with previous studies in people with spinal cord injuries.[12-14] Low aerobic fitness was also associated with higher physical strain during the performance of daily activities, and lower participation and health-related quality of life. Increasing aerobic fitness may break through the vicious cycle of inactivity leading to low fitness exacerbating inactivity. Furthermore, in line with previous studies,[15-18] the results from Chapter 3 support the enclosure of strength exercises in addition to aerobic training as it may contribute to improving aerobic fitness.

In addition to adolescents and young adults, health promotion efforts should also focus on children with a childhood onset physical disability. Several initiatives to improve fitness have been shown in children with cerebral palsy[19,20] and ambulatory children with spina bifida,[21] and should be further encouraged.

Suggestions for future research

Physiological responses to exercise

Maximal exercise tests showed low aerobic fitness in many adolescents and young adults with MMC (Chapter 2 and 3), but little information is available on to what extent this is the result of reduced active muscle mass caused by paralyses of lower extremity muscles or to deconditioning. To determine whether a person has a low aerobic fitness regarding his or her condition, normative values including adjustments for lesion level and testing method (arm or cycle ergometer) are useful. Furthermore, training studies may provide insight in the aerobic fitness gain that is achievable by training.

From studies in persons with spinal cord injury it is known that absence of sympathetic vasoconstriction below the lesion and absence of the leg muscle pump results in disturbed distribution of blood during exercise and venous blood pooling. As a consequence there is a diminished increase in venous preload and in stroke volume during exercise.[22] To compensate for the lower stroke volume, heart rate increases in order to maintain a stable cardiac output. People with lesions below the 6th thoracic vertebrae are able to compensate completely for a lower stroke volume by an increase in heart rate.[23] Thus in most people with MMC, it is unlikely that oxygen transport capacity during exercise is limited by a limited cardiac output. Nevertheless, due to limited information in people with MMC, future studies should provide insight in physiological responses to exercise in adolescents and young adults with MMC with various lesion levels.

Measurements of physical activity

Self-reported physical activity assessments are prone to overestimation of physical activity levels,[24] and may lack sensitivity to low physical activity levels. Therefore, motion sensors are more appropriate for quantifying physical activity behaviours.[25] The AM we used in the studies provides detailed information on which activity is performed for how long. However, the AM has several practical concerns. Until now, measurements are restricted to 48-hour measurements and the device we used was rather large, heavy to wear, and people can not take a shower or bath during activity monitoring. Currently, projects focus on remodelling this sophisticated device to make it more user-friendly and to allow more days of monitoring. For larger study samples, objective physical activity monitoring with a simpler device and less detailed information may be an alternative. For example, the ActiGraph activity monitors are small and can be worn on the hip, ankle or wrist. They integrate information about motion frequency, duration, they are sensitive to light activity, small in size, comfortable to wear, able to collect and save data over long duration, and showed good validity in ambulatory people.[26-28] In contrast to the detection of physical activity in the ambulatory population, little information is available regarding the detection of wheelchair driving. Laboratory measurements showed that an ActiGraph, an uniaxial accelerometer, was a valid indicator of energy expenditure for wheelchair users.[29] One study reported on its use in free-living conditions and showed concurrent validity with a self-report measure of activity intensity and frequency in wheelchair users.[30] Future studies on the use of simpler devices to detect physical activity are warranted.

Determinants of physical activity

Few studies are available on factors associated with physical activity levels in people with physical disabilities (Chapter 8) and in particular in adolescents and young adults with childhood-onset physical disabilities. Chapter 5 reported some correlates of sports participation, which is a sub-category of physical activity that is planned and structured.[31] Chapter 8 presented perceived barriers and facilitators as measured using a qualitative study. Future quantitative studies on barriers and facilitators of physical activity using objective physical activity monitoring are warranted, as well as longitudinal studies on this topic to identify mediators of physical activity improvements. Furthermore, because of scarcity of information, these studies should include environmental factors, including both the physical and social environment, in changing physical activity behaviour.

In addition, many young adults with MMC are impaired in executive functioning, including divided attention, in memory and verbal learning and in reaction speed.[32,33] Results of a small sample of adolescents and young adults suggested that impairments in executive functioning were related to levels of physical activity.[33] It is worthwhile to study the influence of cognitive functioning on levels of physical activity in larger sample sizes.

Intervention 'Active Lifestyle and Sports Participation'

Chapter 9 presents the design of the intervention 'Active Lifestyle and Sports Participation.' The intervention offers a personalised programme tailored to the needs of each participant, and aims to increase levels of physical activity and fitness. It seems to be a promising approach to offer the intervention in the outpatient Rehabilitation department for young adults. However, only 2 case studies are presented and future evaluation is warranted to determine whether the intervention effectively increases levels of physical activity and fitness in adolescents and young adults with a childhood onset physical disability, both on the short and long-term. In addition, cost-effectiveness of the intervention should be evaluated.

A possible future direction of the intervention 'Active Lifestyle and Sports Participation' lies in computer tailoring. Personal counselling, which forms the base of the intervention, may be too time-consuming and too expensive to apply for every individual with a childhood onset disability. Computer tailoring makes personalization of education applicable to large groups of people at relatively low costs.[34] Furthermore, it enables long-distance personal counselling[34] which is a major advantage for people with transportation problems and for those living far away from the rehabilitation centre.

Reducing overweight and obesity

Many adolescents and young adults with MMC are overweight and obese. Excessive body fat is the result of a positive energy balance, i.e. the energy intake is higher than the energy expenditure. Physical activity, which results in energy expenditure, showed no relationship with body fat (Chapter 2). Besides the duration of physical activity, as measured in this thesis, also the intensity of physical activity influences the amount of expended energy. Future studies focusing on energy expenditure and energy intake (dietary intake) may obtain insight in the energy balance in adolescents and young adults with MMC. Furthermore, hormonal analysis of metabolism and medication are warranted to study underlying mechanisms of excessive body fat, as is may originate from the condition itself.[35] In addition to increasing physical activity, future behaviour-change interventions should also focus on improving dietary behaviour in adolescents and young adults with a childhood onset physical disability, such as MMC.

References

1. Verhoef M. Spina Bifida: implications for functioning and health in young adults. Utrecht: University Medical Centre Utrecht; 2005. PhD thesis: ISBN 9039340455.
2. Barf H. Spina Bifida: implications for cognitive functioning, disability and health in young adults. Utrecht: University Medical Centre Utrecht; 2008. PhD Thesis: ISBN 978-90-71382-24-6.
3. Van der Ploeg HP, Streppel KR, van der Beek AJ, van der Woude LH, Vollenbroek-Hutten MM, van Harten WH et al. Successfully improving physical activity behavior after rehabilitation. Am J Health Promot 2007; 21: 153-159.
4. Trost SG, Pate RR, Freedson PS, Sallis JF, Taylor WC. Using objective physical activity measures with youth: how many days of monitoring are needed? Med Sci Sports Exerc 2000; 32: 426-431.
5. White DK, Wagenaar RC, Del Olmo ME, Ellis TD. Test-retest reliability of 24 hours of activity monitoring in individuals with Parkinson's disease in home and community. Neurorehabil Neural Repair 2007; 21: 327-340.
6. Bhambhani YN, Holland LJ, Steadward RD. Maximal aerobic power in cerebral palsied wheelchair athletes: validity and reliability. Arch Phys Med Rehabil 1992; 73: 246-252.
7. Van den Berg-Emons RJ, van Baak MA, Westerterp KR. Are skinfold measurements suitable to compare body fat between children with spastic cerebral palsy and healthy controls? Dev Med Child Neurol 1998; 40: 335-339.
8. 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: 420-427.
9. 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.

10. Smith BJ, van der Ploeg HP, Buffart LM, Bauman AE. Encouraging physical activity--five steps for GPs. Aust Fam Physician 2008; 37: 24-28.
11. Van der Ploeg HP, Streppel KR, van der Beek AJ, van der Woude LH, Vollenbroek-Hutten MM, van Harten WH et al. Counselling increases physical activity behaviour nine weeks after rehabilitation. Br J Sports Med 2006; 40: 223-229.
12. Janssen TW, van Oers CA, van Kamp GJ, TenVoorde BJ, van der Woude LH, Hollander AP. Coronary heart disease risk indicators, aerobic power, and physical activity in men with spinal cord injuries. Arch Phys Med Rehabil 1997; 78: 697-705.
13. Dallmeijer AJ, Hopman MT, van der Woude LH. Lipid, lipoprotein, and apolipoprotein profiles in active and sedentary men with tetraplegia. Arch Phys Med Rehabil 1997; 78: 1173-1176.
14. Dallmeijer AJ, van der Woude LH, van Kamp GJ, Hollander AP. Changes in lipid, lipoprotein and apolipoprotein profiles in persons with spinal cord injuries during the first 2 years post-injury. Spinal Cord 1999; 37: 96-102.
15. Cooney MM, Walker JB. Hydraulic resistance exercise benefits cardiovascular fitness of spinal cord injured. Med Sci Sports Exerc 1986; 18: 522-525.
16. Janssen TW, van Oers CA, Hollander AP, Veeger HE, van der Woude LH. Isometric strength, sprint power, and aerobic power in individuals with a spinal cord injury. Med Sci Sports Exerc 1993; 25: 863-870.
17. Zoeller RF, Jr., Riechman SE, Dabayebeh IM, Goss FL, Robertson RJ, Jacobs PL. Relation between muscular strength and cardiorespiratory fitness in people with thoracic-level paraplegia. Arch Phys Med Rehabil 2005; 86: 1441-1446.
18. Haisma JA, Bussmann JB, Stam HJ, Sluis TA, Bergen MP, Dallmeijer AJ et al. Changes in physical capacity during and after inpatient rehabilitation in subjects with a spinal cord injury. Arch Phys Med Rehabil 2006; 87: 741-748.
19. Van den Berg-Emons R. Physical training of school children with spastic cerebral palsy. Maastricht: University of Maastricht; 1996. PhD thesis: ISBN 90-9009532-2.
20. Verschuren O. Physical fitness in children and adolescents with cerebral palsy. Utrecht: University of Utrecht; 2007. PhD thesis: ISBN 978-90-393-4665-5.
21. De Groot J. Utrecht Spina Bifida And Graded Exercise (USAGE) Studie: Fysieke activiteit, fysieke fitheid en de ontwikkeling van een oefenprogramma ter verbetering van dagelijkse activiteiten, participatie en gezondheidsgerelateerde kwaliteit van leven bij kinderen met Spina Bifida.
22. Hopman MT, Oeseburg B, Binkhorst RA. Cardiovascular responses in paraplegic subjects during arm exercise. Eur J Appl Physiol Occup Physiol 1992; 65: 73-78.
23. Hopman MT, Pistorius M, Kamerbeek IC, Binkhorst RA. Cardiac output in paraplegic subjects at high exercise intensities. Eur J Appl Physiol Occup Physiol 1993; 66: 531-535.
24. Sallis JF, Saelens BE. Assessment of physical activity by self-report: status, limitations, and future directions. Res Q Exerc Sport 2000; 71: S1-14.
25. Tudor-Locke CE, Myers AM. Challenges and opportunities for measuring physical activity in sedentary adults. Sports Med 2001; 31: 91-100.
26. Melanson EL, Jr., Freedson PS. Validity of the Computer Science and Applications, Inc. (CSA) activity monitor. Med Sci Sports Exerc 1995; 27: 934-940.
27. Trost SG, Ward DS, Moorehead SM, Watson PD, Riner W, Burke JR. Validity of the computer science and applications (CSA) activity monitor in children. Med Sci Sports Exerc 1998; 30: 629-633.

28. De Vries SI, Bakker I, Hopman-Rock M, Hirasing RA, van Mechelen W. Clinimetric review of motion sensors in children and adolescents. J Clin Epidemiol 2006; 59: 670-680.
29. Washburn RA, Copay AG. Assessing physical activity during wheelchair pushing: validity of a portable accelerometer. Adapt Phys Activ Q 1999; 16: 290-299.
30. Warms CA, Belza BL. Actigraphy as a measure of physical activity for wheelchair users with spinal cord injury. Nurs Res 2004; 53: 136-143.
31. Caspersen CJ, Powell KE, Christenson GM. Physical activity, exercise, and physical fitness: definitions and distinctions for health-related research. Public Health Rep 1985; 100: 126-131.
32. Barf HA, Verhoef M, Jennekens-Schinkel A, Post MW, Gooskens RH, Prevo AJ. Cognitive status of young adults with spina bifida. Dev Med Child Neurol 2003; 45: 813-820.
33. Roebroeck ME, Hempenius L, van Baalen B, Hendriksen JG, van den Berg-Emons HJ, Stam HJ. Cognitive functioning of adolescents and young adults with meningomyelocele and level of everyday physical activity. Disabil Rehabil 2006; 28: 1237-1242.
34. Brug J, Oenema A, Campbell M. Past, present, and future of computer-tailored nutrition education. Am J Clin Nutr 2003; 77: 1028S-1034S.
35. 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: 305-311.

Summary

The introductory **Chapter 1** describes the various types of spina bifida, including the most severe myelomeningocele (MMC), and its associated symptoms. Furthermore, it introduces new challenges that rise in medical care with the increased life expectancy of children who are born with this condition. Similarly to persons with other chronic conditions, there is a shift from disability prevention towards prevention of secondary conditions. Sufficient levels of physical activity and fitness are important for disease prevention, maintenance of functional independence, participation and quality of life. Studies on this topic in people with MMC are scarce. Therefore, this thesis aimed to obtain insight in the levels of daily physical activity and fitness of adolescents and young adults with MMC and to study the associations with personal and environmental factors, participation, health-related quality of life and cardiovascular disease (CVD) risk.

Chapter 2 shows that many adolescents and young adults with MMC were inactive, as measured with an accelerometry-based activity monitor; they had low aerobic fitness and high body fat compared to normative values. Differences existed between subgroups regarding gender and ambulatory status. Non-ambulators generally had lower levels of physical activity and aerobic fitness, and higher body fat than ambulators. Females had lower aerobic fitness and more body fat than males, but the level of physical activity was comparable. In non-ambulators, who were most inactive, we found an association between physical activity and aerobic fitness, suggesting that a minimum level of aerobic fitness might be needed to have an active lifestyle. Physical activity and aerobic fitness were not associated with body fat suggesting that increasing levels of activity and fitness may not be sufficient to reduce body fat.

Aerobic fitness, or aerobic capacity, is one component of health-related physical fitness. The other components are muscle strength and endurance, flexibility and body composition. **Chapter 3** describes health-related physical fitness components of adolescents and young adults with MMC and studies relationships between aerobic capacity and the other components. Results showed that both ambulatory and non-ambulatory adolescents and young adults with MMC had low health-related physical fitness. In addition to reduced aerobic capacity and excessive body fat, the majority had subnormal muscle strength and mobility restrictions. A large part of the variance of aerobic capacity was explained by gender and ambulatory status. In addition, we found a small, but significant relationship between aerobic capacity and muscle strength, suggesting that adding strength training to aerobic training may have additional value in increasing aerobic capacity.

Chapter 4 studies the energy cost and physical strain of daily activities of ambulatory and non-ambulatory adolescents and young adults with MMC, and age-and gender matched comparison people without physical disabilities in a laboratory setting. During an activity protocol consisting of locomotion-related (walking, wheelchair driving, cycling) and housekeeping-related activities, oxygen uptake was measured to assess the energy cost. Physical strain was calculated by dividing the energy cost per unit time by a person's aerobic capacity. Results showed no differences in oxygen costs of all activities between ambulatory persons with MMC and comparison people. Non-ambulators had lower energy cost during wheelchair driving than the comparison people during walking. Furthermore, results showed that people with MMC perform daily activities at a higher percentage of their aerobic capacity than comparison people, indicating a higher physical strain. This may increase fatigue and discomfort and subsequently contribute to an inactive lifestyle. The high physical strain was mainly caused by a reduced aerobic capacity, suggesting that improving aerobic capacity may therefore be an important treatment goal for this population.

Sports and exercise can be seen as a sub-category of physical activity which is planned, structured and repetitive. Sports-related physical activities can easily be provided through rehabilitation services and could increase levels physical activity and fitness. **Chapter 5** showed that two-third of the participating adolescents and young adults with MMC reported to be engaged in sports. Ambulatory status, the presence of hydrocephalus and functional independence were not related to sports participation, which supports the view that engaging in sports is due to personal preferences rather than physical ability. Sports participation was associated with social support from family, perceived athletic competence and physical appearance and tended to be associated with global self-worth, which makes them interesting targets for interventions that aim to improve physical activity behaviour. Furthermore, persons who perceive higher enjoyment during sports tended to be more likely to participate in sports. Therefore, interventions aiming to increase physical activity levels should be tailored individually ensuring that each person finds the type of activity they enjoy. Results showed that sports participation has the potential to increase physical activity and fitness. Sports participants reported higher levels of self-reported physical activity than non-participants. Sports contributed to 16% of total physical activity. Non-participants did not seem to compensate with other physical activities such as household and occupations. Although the present results indicated that sports participants may have higher aerobic capacity and higher muscle strength than non-participants, conclusive evidence is warranted.

This thesis found some evidence that improving levels of physical activity and fitness should be incorporated in health care of adolescents and young adults with MMC, since higher levels of physical activity and fitness were associated with fewer participation restrictions, better health-related quality of life (HRQoL), and reduced CVD risk.

Chapter 6 focused on participation and HRQoL. Results showed that many adolescents and young adults perceived difficulties in the performance of daily activities (63%) and social roles (59%), particularly regarding self care, housing, mobility, recreation and employment. Furthermore, adolescents and young adults with MMC perceived lower physical HRQoL compared to a general Dutch reference population, particularly regarding mobility. In general, they perceived good mental HRQoL, but perceived somewhat lower vitality and social functioning than the reference population. People with higher levels of physical activity and higher aerobic fitness were less likely to perceive difficulties during the performance of daily activities and perceived a higher physical HRQoL. Also, people with higher aerobic fitness tended to perceive less difficulty in social roles. Body fat was not related with participation and HRQoL.

Chapter 7 describes biological and lifestyle-related CVD risk factors including lipid and lipoprotein profiles, blood pressure, aerobic fitness, body fat, daily physical activity and smoking behaviour. From literature, it is known that the severity of atherosclerosis, causing CVD, increases as the number of CVD risk factors increases, and that this clustering of risk factors is a better measure for cardiovascular health. Therefore, participants were identified as being at increased CVD risk when 2 or more of the risk factors from the Framingham Risk Assessment (total cholesterol, systolic blood pressure, high-density lipoprotein cholesterol, and smoking) were present. Based on this criterion, 42% of the participants were at increased CVD risk. Non-ambulators were more likely to be at risk than ambulators. Results showed that people with higher aerobic fitness tended to be more likely to have no CVD risk. Levels of physical activity and body fat were not associated with CVD risk.

In order to develop programs to promote physical activity in people with a childhood onset physical disability, such as MMC, it is important to understand factors that determine physical activity behaviour. **Chapter 8** provides an overview of previous studies on personal and environmental barriers and facilitators of physical activity in young adults with a physical disability. Furthermore in presents results of focus groups that we held in adolescents and young adults with a childhood onset physical disability to obtain additional insight in their perceived barriers and facilitators of physical activity. The literature review and focus groups showed that lack of energy, existing injuries of complications or fear for developing them, lack of motivation, facility limitations, or lack of information and knowledge appeared to be important barriers to physical activity. Fun and social contacts were most frequently mentioned as facilitators of physical activity. This indicates that tailored physical activity counselling is important to ensure that persons have fun during physical activity.

Chapter 9 presents the design of the intervention 'Active Lifestyle and Sports Participation' which is offered in the outpatient Rehabilitation Department for young adults of Erasmus MC and Rijndam Rehabilitation Centre. The main approach of the intervention consists of tailored physical activity counselling session with a physical activity counsellor, who serves a person's "personal coach." Together with the personal coach, participants discuss their involvement in other components of the intervention, which are sports participation by means of tailored sport advice and sport specific training, and fitness training. In this way, participants will conduct a personalised programme tailored to their needs. Two case studies are described and future directions of the intervention are presented.

Finally, **Chapter 10** discusses the main findings of the studies and presents a schematic model of factors associated with physical activity and fitness in adolescents and young adults with MMC. Furthermore, it discusses some methodological issues related to the studies including the low response rate and the generalizability of the results, study design and statistical power and the choice of outcome measures. In addition, this chapter describes how important findings of the studies are integrated in the design of the intervention 'Active Lifestyle and Sports Participation'. In the final part of this chapter we provide some suggestions for future studies. More insight in physiological responses to exercise is warranted, as well as studies on the use of simpler devices to objectively detect physical activity and studies on determinants of physical activity. Furthermore, future studies should focus on evaluation and refinement of the intervention 'Active Lifestyle and Sports Participation'.

Samenvatting

Het inleidende **Hoofdstuk 1** beschrijft de verschillende vormen van spina bifida, inclusief de meest ernstige vorm meningomyelocele (MMC), en de symptomen die daarmee samenhangen. Het introduceert nieuwe uitdagingen in de medische zorg die zijn ontstaan door de toegenomen levensverwachting van kinderen die met deze aandoening worden geboren. Net zoals bij personen met andere chronische aandoeningen vindt er een verschuiving plaats van behandeling van de primaire aandoening naar de preventie van secundaire aandoeningen. Voldoende lichaamsbeweging en een goede fitheid zijn belangrijk voor de preventie van ziektes, het behoud van de functionele onafhankelijkheid, de participatie en de kwaliteit van leven. Bij mensen met een MMC zijn hier nog maar weinig studies naar verricht. Daarom heeft dit proefschrift als doel om inzicht te krijgen in de hoeveelheid dagelijkse lichamelijke activiteit en in de fitheid van jongeren en jongvolwassenen met een MMC. Tevens bestuderen we de relaties met persoonlijke en omgevingsfactoren, de participatie, de gezondheidsgerelateerde kwaliteit van leven en het risico op het ontstaan van hart- en vaatziekten (HVZ).

Hoofdstuk 2 laat zien dat veel jongeren en jongvolwassenen met een MMC inactief waren, zoals gemeten met een activiteiten monitor die gebruik maakt van versnellingssensoren. Bovendien hadden ze een lage aërobe fitheid en overtollig lichaamsvet in vergelijking met norm waarden. We vonden verschillen tussen subgroepen; niet-ambulante jongeren en jongvolwassenen met een MMC waren lichamelijk inactiever en hadden een lagere aërobe fitheid en meer lichaamsvet dan ambulante jongeren en jongvolwassenen. Vrouwen hadden een lagere aërobe fitheid en meer lichaamsvet dan mannen, maar kregen evenveel lichaamsbeweging. In de personen die het minst actief waren, de niet-ambulante jongeren, vonden we een relatie tussen de hoeveelheid lichaamsbeweging en de aërobe fitheid. Dit suggereert dat er mogelijk een minimum niveau van aërobe fitheid nodig is om een actieve leefstijl te hebben. Lichamelijke activiteit en aërobe fitheid waren niet gerelateerd aan de hoeveelheid lichaamsvet, wat suggereert dat het bevorderen van het beweeggedrag en de fitheid mogelijk niet voldoende is om de hoeveelheid lichaamsvet te verlagen.

Aërobe fitheid, ofwel aërobe capaciteit, is een van de componenten van gezondheidsgerelateerde fysieke fitheid. De andere componenten zijn spierkracht en spieruithoudingsvermogen, lenigheid, en lichaamssamenstelling. **Hoofdstuk 3** beschrijft de componenten van gezondheidsgerelateerde fysieke fitheid van jongeren en jongvolwassenen met een MMC, en bestudeert de relaties tussen aërobe capaciteit en de andere componenten. Uit deze studie bleek dat zowel ambulante als niet-ambulante jongeren en jongvolwassenen met een MMC een lage gezondheidsgerelateerde fysieke fitheid hadden. Naast een lage aërobe capaciteit en overtollig lichaamsvet had de meerderheid ook een subnormale spierkracht en bewegingsbeperkingen. Een groot deel van de variantie in de aërobe capaciteit werd verklaard door het geslacht en het ambulantieniveau. Daarnaast vonden we een kleine, maar significante relatie tussen de aërobe capaciteit en de spierkracht. Dit suggereert dat spierkrachttraining als aanvulling op aërobe training van toevoegende waarde kan zijn om de aërobe capaciteit te verbeteren.

Hoofdstuk 4 bestudeert de energiekosten en de fysieke belasting tijdens dagelijkse activiteiten bij ambulante en niet-ambulante jongeren en jongvolwassenen met een MMC, in vergelijking tot een referentie populatie bestaande uit een groep personen zonder lichamelijke beperking van dezelfde leeftijd en geslacht. De metingen werden uitgevoerd in een bewegingslaboratorium. Om de energiekosten te bepalen werd de zuurstofopname gemeten tijdens het uitvoeren van een activiteiten protocol, bestaande uit activiteiten die gerelateerd zijn aan de voortbeweging (lopen, rolstoel rijden, fietsen) en uit huishoudelijke activiteiten. De fysieke belasting werd berekend door de energiekosten per tijdseenheid te delen door de aërobe capaciteit van de persoon. De zuurstofkosten van alle activiteiten verschilden niet tussen ambulante personen met een MMC en de referentiegroep. Niet-ambulante jongeren hadden lagere energiekosten tijdens rolstoel rijden dan de referentiegroep tijdens lopen. Bovendien bleek dat mensen met een MMC de dagelijkse activiteiten op een hoger percentage van hun aërobe capaciteit uitvoerden dan de referentie groep, wat betekent dat ze een hogere fysieke belasting hadden. Een hogere fysieke belasting kan leiden tot vermoeidheid en vervolgens weer bijdragen aan een inactieve leefstijl. Aangezien de hogere fysieke belasting voornamelijk werd veroorzaakt door een verlaagde aërobe capaciteit, lijkt verbetering van de aërobe capaciteit een belangrijk behandelingsdoel te zijn voor deze populatie.

Sport en inspanning is een subcategorie van lichamelijke activiteit, welke gepland en gestructureerd is en herhaald wordt. Sportgerelateerde lichamelijke activiteiten kunnen gemakkelijk in de revalidatie worden aangeboden, en kunnen het beweeggedrag bevorderen en de fitheid verbeteren. **Hoofdstuk 5** laat zien dat tweederde van de deelnemende jongeren en jongvolwassenen met een MMC aan sport deed. Het ambulantieniveau, het wel of niet hebben van een hydrocephalus en de functionele onafhankelijkheid waren niet gerelateerd aan sportdeelname. Het lijkt er daarom op dat deelname aan sport bepaald wordt door persoonlijke voorkeuren in plaats van fysieke mogelijkheden. Sportdeelname was gerelateerd aan de sociale steun van familie, en aan de competentiebeleving ten aanzien van sportieve vaardigheden en de fysieke verschijning. We vonden een trend met het algemene gevoel voor eigenwaarde. Deze factoren zijn daarom interessante doelwitten voor interventies die erop gericht zijn om het beweeggedrag te bevorderen. Tevens vonden we dat personen die meer plezier beleefden aan sport, eerder geneigd waren aan sport deel te nemen. Daarom is het belangrijk dat interventies ter bevordering van het beweeggedrag afgestemd zijn op het individu, om ervoor te zorgen dat elk persoon het type activiteit vindt waar hij of zij plezier in heeft. Uit de resultaten bleek dat sportdeelname de potentie heeft om het beweeggedrag te bevorderen en de fitheid te verbeteren. Sporters gaven aan lichamelijk actiever te zijn dan niet-sporters. De bijdrage van sport aan het totale lichamelijke activiteiten patroon was 16%. Niet-sporters leken dit niet te compenseren met andere lichamelijke activiteiten zoals huishoudelijke of werk activiteiten. Hoewel de resultaten in de richting wezen dat sporters een betere aërobe fitheid en een hogere spierkracht hadden dan niet-sporters is dit nog niet voldoende aangetoond.

Dit proefschrift levert enig bewijs dat bevordering van het beweeggedrag en de verbetering van de fitheid aandacht moeten krijgen in de zorg voor jongeren en jongvolwassenen met een MMC. Een lichamelijk actievere leefstijl en een betere fitheid hangen namelijk samen met minder beperkingen in de participatie, een betere gezondheidsgerelateerde kwaliteit van leven en een verminderd risico op HVZ.

Hoofdstuk 6 richt zich op de participatie en de gezondheidsgerelateerde kwaliteit van leven. Uit de studie bleek dat veel jongeren en jongvolwassenen moeite ervaarden bij het uitvoeren van dagelijkse activiteiten (63%) en sociale rollen (59%). Bovendien ervaarden jongeren en jongvolwassenen met een MMC een lagere fysieke gezondheidsgerelateerde kwaliteit van leven vergeleken met een Nederlandse referentie populatie; het betrof hier voornamelijk de mobiliteit. Over het algemeen ervaarden zij een goede mentale gezondheidsgerelateerde kwaliteit van leven, maar hun scores op vitaliteit en het sociaal functioneren waren lager dan de referentie waarden. Mensen die lichamelijk actiever waren en een hogere aërobe fitheid hadden, ervaarden minder moeite tijdens het uitvoeren van dagelijkse activiteiten en hadden een betere fysieke gezondheidsgerelateerde kwaliteit van leven. Bovendien leken mensen met een betere aërobe fitheid minder moeite te ervaren met sociale rollen. We vonden geen relatie tussen de hoeveelheid lichaamsvet en de participatie en de gezondheidsgerelateerde kwaliteit van leven.

Hoofdstuk 7 beschrijft biologische en leefstijlgerelateerde risicofactoren van HVZ, zoals het lipide en lipoproteïne profiel, de bloeddruk, de aërobe fitheid, de hoeveelheid lichaamsvet, de dagelijkse lichamelijke activiteit en het rookgedrag. Vanuit de literatuur weten we dat de ernst van arteriosclerose, dat HVZ veroorzaakt, toeneemt naarmate het aantal risicofactoren dat aanwezig is stijgt. Het clusteren van de risicofactoren is een betere maat voor de cardiovasculaire gezondheid. We vonden dat de deelnemers bij wie 2 of meer risicofactoren van de Framingham Risk Assessment (totaal cholesterol, systolische bloeddruk, high-density lipoprotein cholesterol, en roken) aanwezig waren, een verhoogd risico hadden op HVZ. Op basis van dit criterium had 42% van de deelnemers een verhoogd risico. De kans op een verhoogd risico op HVZ was hoger bij niet-ambulante deelnemers dan bij ambulante deelnemers. Bovendien bleek dat mensen met een betere aërobe fitheid een lager risico hadden op het ontstaan van HVZ. De hoeveelheid dagelijkse lichamelijke activiteit en de hoeveelheid lichaamsvet vertoonden geen samenhang met het risico op HVZ.

Om voor kinderen met een aangeboren of jong verworven lichamelijke beperking, zoals een MMC, programma's te ontwikkelen ter bevordering van het beweeggedrag is het belangrijk om inzicht te krijgen in de factoren die het beweeggedrag bepalen. **Hoofdstuk 8** presenteert een literatuurstudie naar persoonlijke en omgevingsfactoren die belemmerende of een bevorderende invloed hebben op het beweeggedrag van jongeren en jongvolwassenen met lichamelijke beperking. Bovendien beschrijven we de resultaten van de focusgroepen die we hebben gehouden met jongeren en jongvolwassenen met een aangeboren of jong verworven lichamelijke beperking. Deze focusgroepen hadden als doel inzicht krijgen in factoren die de jongeren en jongvolwassenen als belemmerend en bevorderend ervaren ten aanzien van hun beweeggedrag. Uit het literatuuroverzicht en de focusgroepen kwamen een gebrek aan energie, de aanwezigheid van wonden, blessures en complicaties of de angst deze te ontwikkelen, een gebrek aan motivatie, beperkte mogelijkheden en gebrek aan informatie of kennis naar voren als belangrijke belemmerende factoren. Het plezier en sociale contacten werden het vaakst genoemd als bevorderende factoren voor bewegen. Dit onderstreept het belang van geïndividualiseerde bewegingscounseling om ervoor te zorgen dat personen plezier hebben tijdens lichaamsbeweging.

Hoofdstuk 9 beschrijft de behandelmodule 'Actieve Leefstijl en Sportstimulering'. Deze module wordt aangeboden in de Polikliniek Jongvolwassenen van de afdeling Revalidatiegeneeskunde van het Erasmus MC en Rijndam revalidatiecentrum te Rotterdam. In deze module staat individuele bewegingscounseling met een bewegingscounselor ('personal coach') centraal. Samen met de personal coach bespreken de deelnemers of ze gebruik gaan maken van de andere componenten van de module; dit zijn sportstimulering, door middel van individueel sportadvies en sportspecifieke training, en fitheidstraining. Op deze manier volgen de deelnemers een geïndividualiseerd programma, afgestemd op hun eigen wensen en mogelijkheden. Verder worden er 2 case reports beschreven en worden de ideeën voor de toekomst van de module gegeven.

Tot slot worden in **Hoofdstuk 10** de belangrijkste bevindingen van de studies besproken en wordt een schematisch model van de factoren die samenhangen met de lichamelijke activiteit en de fitheid van jongeren en jongvolwassenen met een MMC gepresenteerd. Bovendien worden methodologische aspecten van de studies besproken, zoals het lage responspercentage en de generaliseerbaarheid van de resultaten, de studieopzet en de analyses, en de keuze van de uitkomstmaten. Verder beschrijft dit hoofdstuk hoe de belangrijke bevindingen van de studies geïntegreerd zijn in het ontwerp van de behandelmodule 'Actieve Leefstijl en Sportstimulering'. In het laatste deel van dit hoofdstuk geven we enkele suggesties voor toekomstig onderzoek. Zo is er bijvoorbeeld meer inzicht nodig in de fysiologische reacties op inspanning. Ook zijn er meer studies nodig naar het gebruik van eenvoudigere meetinstrumenten om lichamelijke activiteit objectief te kunnen meten en naar determinanten van het beweeggedrag. Bovendien moeten toekomstige studies zich richten op de evaluatie en verfijning van de behandelmodule 'Actieve Leefstijl en Sportstimulering'.

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Curriculum Vitae

Laurien Buffart was born in Portsmouth (UK) on 19th of June 1980. She finished her atheneum at RSG Wiringherlant in Wieringerwerf in 1998. In the same year she started studying Human Movement Sciences at the Vrije Universiteit in Amsterdam. She did her research internship at the Rehabilitation Centre Amsterdam under supervision of dr T. Janssen, studying physiological responses to voluntary arm and electrically-induced cycle exercise in people with spinal cord injury. She finished the Master of Science education in 2003 with a major in human movement systems.

After her studies, she started as a junior researcher at the department of Rehabilitation Medicine of Erasmus MC studying arm/hand functioning in children with a congenital reduction deficiency of the upper limb. From 2004 she started with her PhD thesis on physical activity and fitness in adolescents and young adults with myelomeningocele at the same department.

During her PhD, she worked for the Transfergroep Rotterdam several times, to educate physical therapists about 'Evidence Based Practice'. Furthermore, she visited the Centre for Physical Activity and Health at the University of Sydney in Australia for 5 months, working on a project on General Practitioners' perceptions and practices of physical activity and weight counselling.

List of Publications

Buffart LM, van den Berg-Emons RJ, van Meeteren J, Stam HJ, Roebroeck ME. Lifstyle, participation and health-related quality of life in adolescents and young adults with myelomeningocele. *Submitted for publication.*

Buffart LM, van der Ploeg HP, Bauman AE, van Asbeck FW, Stam HJ, Roebroeck ME, van den Berg-Emons RJ. Sports participation in adolescents and young adults with myelomeningocele and its role in physical activity behaviour and fitness. J Rehabil Med *in press*.

Buffart LM, van den Berg-Emons RJ, Burdorf A, Janssen WG, Stam HJ, Roebroeck ME. Cardiovascular disease risk factors and the relations with physical activity, aerobic fitness, and body fat in adolescents and young adults with myelomeningocele. Arch Phys Med Rehabil *in press*.

Buffart LM, Roebroeck ME, Janssen WG, Hoekstra A, Selles RW, Hovius SE, Stam HJ. Hand function and activity performance in children with longitudinal radial deficiencies. J Bone Joint Surg *in press*.

Buffart LM, Allman-Farinelli M, King LA, van der Ploeg HP, Smith BJ, Kurko J, Bauman AE. Are General Practitioners ready and willing to tackle obesity management? Obes Res Clin Pract 2008 *in press*.

Buffart LM, van der Ploeg HP, Smith BJ, Kurko J, King LA, Bauman AE. General practitioners' perceptions and practices of physical activity counselling: changes over the past 10 years. Br J Sports Med 2008; *Jul 15 Epub ahead of print.*

Buffart LM, van den Berg-Emons RJG, 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: 181-188.

Smith BJ, van der Ploeg HP, Buffart LM, Bauman AE. Encouraging physical activity – five steps for GPs. Aust Fam Physician 2008; 37: 24-28.

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.

Bruinings AL, van den Berg-Emons HJ, Buffart LM, van der Heijden-Maessen HC, Roebroeck ME, Stam HJ. Energy cost and physical strain of daily activities in adolescents and young adults with myelomeningocele. Dev Med Child Neurol 2007; 49: 672-677.

Buffart LM, Roebroeck ME, van Heijningen VG, Pesch-Batenburg JM, Stam HJ. Evaluation of arm and prosthetic functioning in children with a congenital transverse reduction deficiency of the upper limb. J Rehabil Med 2007; 39: 379-386.

Buffart LM, Roebroeck ME, Janssen WG, Hoekstra A, Hovius SE, Stam HJ. Comparison of instruments to assess hand function in children with radius deficiencies. J Hand Surg [Am] 2007; 32: 531-540.

Buffart LM, Roebroeck ME, Pesch-Batenburg JM, Janssen WG, Stam HJ. Assessment of arm/hand functioning in children with a congenital transverse or longitudinal reduction deficiency of the upper limb. Disabil Rehabil 2006; 28: 85-95.

Boeschoten K, Hoekstra A, Buffart L, van Heijningen V. Assisting Hand Assessment, een instrument voor het meten van de handfunctie bij kinderen met een unilaterale aandoening aan arm/hand. Nederlands Tijdschrift voor Ergotherapie 2006; 34: 8-12

Hoekstra A, Boeschoten K, Buffart L, van Heijningen V. Assisting Hand Assessment; een nieuw instrument voor het meten van handfunctie bij kinderen met een unilaterale aandoening aan arm/hand. Kinderfysiotherapie 2005; 45: 14-17.