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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.

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Introduction

During the last decades, life expectancy of persons with myelomeningocele (MMC) has increased and many will nowadays survive into adulthood (Bowman et al. 2001). 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 (Lamb et al. 1988; Twisk et al. 2002) and it may be important for the performance of functional activities and quality of life (Noreau and Shephard 1995; Stewart et al. 1994). Low physical fitness may result in high physical strain during the performance of activities (Bruinings et al. 2007). 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 (Caspersen et al. 1985).

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 (Buffart

et al. in press). Several other studies have also reported low aerobic capacity (Agre et al. 1987; Sherman et al. 1997; van den Berg-Emons et al. 2003). Furthermore, children with MMC, and particularly non-ambulatory children, were found to have reduced strength of lower extremity muscles (Agre et al. 1987; McDonald et al. 1991; Schoenmakers et al. 2004). Hip and knee contractures have been reported in respectively 18% and 36% of adolescents and young adults with MMC and hydrocephalus (Verhoef et al. 2004). Finally, high levels of body fat have been found in persons with MMC (Bandini et al. 1991; Mita et al. 1993; Shepherd et al. 1991); previously we reported that 35% of adolescents and young adults with MMC were obese (Buffart et al. in press). 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.
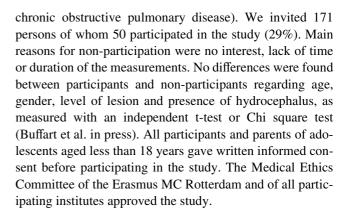
Due to the scarcity of studies in persons with MMC, the first aim of this study was to describe health-related physical fitness i.e. aerobic capacity, muscle strength, joint flexibility and body composition in a relatively large group of adolescents and young adults with MMC, allowing descriptions of 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 (Haisma et al. 2006; Janssen et al. 1993; Zoeller et al. 2005). 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 (Buffart et al. in press). 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 (Hoffer et al. 1973): (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) (Donkervoort et al. 2007).

Aerobic capacity

Aerobic capacity was measured in a progressive maximal exercise test, based on the McMaster All-Out Progressive Continuous Cycling and Arm test (Bar-Or 1983), 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 (Bhambhani et al. 1992). 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

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 (%)[a]	
Community ambulator	30
Household ambulator	14
Non (functional) ambulator	56
Educational level (%)	
Low	37
Medium	39
High	24

[a] No gender differences as tested with a Chi square test

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 (K4b2, 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 (Vos 2001). 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 (Reinhard et al. 1979; Wassermann et al. 1999). 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) (Borg 1982; Mahler et al. 1987).

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) 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 (van der Ploeg et al. 1991). 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 (Phillips et al. 2000) and for knee extensors, we used reference values of Bohannon and co-workers (Bohannon 1997). 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 (Vos 2001).

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 coefficient (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 ($r = 0.58$; $P < 80.001$), 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 (Verhoef et al. 2004).

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 ($\beta = -0.61$; $P < 0.001$), higher in community ambulatory persons than in household and non-ambulatory persons ($\beta = -0.48$; $P < 0.001$) and higher in persons with a lower level of lesion ($\beta = -0.43$; $P = 0.002$). 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$_2$ ($\beta = 0.22$; $P = 0.04$) 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$_2$ ($\beta = 0.25$; $P = 0.08$).

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

	All ($n = 50$)	Gender		Ambulatory status		
		Male ($n = 25$)	Female ($n = 25$)	Community ($n = 15$)	Household ($n = 7$)	Non(functional) ($n = 28$)
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[a]	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 (W)	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 four 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

[a] Only for those measured on the cycle ergometer, $n = 17$ (ten males, seven females)

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 (Lundberg 1978). However, peakVO$_2$ is influenced by the amount of active muscle mass (Davies and Sargeant 1974, 1975; Lewis et al. 1983), 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 (Bar-Or and Zwiren 1975). 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 (Janssen et al. 2002), 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 (van den Berg-Emons et al. 2001), particularly in non-ambulatory persons with MMC (Buffart et al. in press).

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$_2$max during leg exercise (Davis et al. 1997), and values of 40–60% have been found in the able-bodied population during arm

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 four 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

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)

[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

exercise (Flandrois et al. 1986; Lin et al. 1993; Schneider et al. 1999). 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 (Coutts and McKenzie 1995).

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 in 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 (Agre et al. 1987; McDonald et al. 1991; Schoenmakers et al. 2004) and might be related to disturbed innervations 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 (Verhoef et al. 2004). 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 (Agre et al. 1987).

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 (Bandini et al. 1991; Mita et al. 1993; Shepherd et al. 1991).

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 (Haisma et al. 2006; Janssen et al. 1993; Zoeller et al. 2005). 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$_2$ (Janssen et al. 1993). In persons with spinal cord injury, strength training resulted in increased peakVO$_2$ (Cooney and Walker 1986). 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 (Unnithan et al. 2006).

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 (Agre et al. 1987).

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$_2$ (Bhambhani et al. 1992), 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 (Lundberg 1978). 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 hand-held dynamometry. This method is known to be cheap, quickly applicable (van der Ploeg et al. 1984) and reliable (Bohannon 1997). However, for an average examiner, values above 250 N are considered too high to apply sufficient resistance (van der Ploeg et al. 1991), 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 (Verhoef et al. 2004). 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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