

ORIGINAL REPORT

TRIAD OF PHYSICAL ACTIVITY, AEROBIC FITNESS AND OBESITY IN ADOLESCENTS AND YOUNG ADULTS WITH MYELOMENINGOCELE

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**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 accelerometer-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 ( $\beta = 0.541$ ), aerobic fitness ( $\beta = 0.397$ ) and body fat ( $\beta = -0.243$ ). Gender was related to aerobic fitness ( $\beta = -0.529$ ) and body fat ( $\beta = 0.610$ ). Physical activity was related to aerobic fitness in non-ambulatory persons with myelomeningocele ( $\beta = 0.398$ ), 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.

**Key words:** spina bifida, aerobic fitness, daily physical activity, body fat.

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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 I). 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  $\chi^2$  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 Höffer 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.

*Measurements*

**Daily physical activity.** Daily physical activity was determined using an AM (Temece 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 pos-

tures 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 minutes/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 (K4b2, 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 (peak $\dot{V}O_2$ ). 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

Table I. Characteristics of participants and non-participants

	Participants ( $n = 51$ )	Non-participants ( $n = 108$ )	<i>p</i> -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	
Thoraco-lumbar	14	7	
Lumbar	29	35	
Lumbosacral	41	42	
Sacral	14	7	
Hydrocephalus (%)	82	79	0.90

SD: standard deviation.

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  $\geq 30$  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), peak  $\text{VO}_2$  (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 ( $\beta$ ) 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 II.

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  $\geq 30$ ) 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 III). Persons with a higher level of ambulatory status (community ambulators vs household ambulators vs non(functional) ambulators) were more physically active during the day ( $\beta = 0.541$ ;  $p < 0.001$ ), had higher aerobic fitness ( $\beta = 0.397$ ;  $p < 0.001$ ) and less body fat ( $\beta = -0.243$ ;  $p = 0.03$ ) compared with less ambulatory persons. Males had higher values of aerobic fitness ( $\beta = -0.529$ ;  $p < 0.001$ ) and less body fat ( $\beta = 0.610$ ;  $p < 0.001$ ) 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 IV). Aerobic fitness was related to physical activity in non-ambulatory persons ( $\beta = 0.398$ ;  $p = 0.04$ ), 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 III. Regression models for daily physical activity, aerobic fitness and body fat for total group ( $n = 50^*$ )

Dependent variable	Independent variable	$\beta$	$p$ -value	$R^2$
Daily physical activity (min/day)	Ambulatory status	-0.541	< 0.001	0.29
Aerobic fitness (peak $\text{VO}_2$ , litres/min)	Gender	-0.529	< 0.001	0.50
Body fat (sum of 4 skin-folds in mm)	Ambulatory status	-0.397	< 0.001	0.47
	Gender	0.610	< 0.001	
	Ambulatory status	0.243	0.03	

$\beta$ : regression coefficient;  $R^2$ : explained variance. \*One missing due to ambulatory problems.

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

	Total group ( $n = 51$ ) Mean (SD)	Community ambulator ( $n = 15$ ) Mean (SD)	Household ambulator ( $n = 8$ ) Mean (SD)	Non-ambulator ( $n = 28$ ) Mean (SD)
Height (cm)	157.7 (38.5)	165.5 (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) <sup>b</sup>	127 (80)	93 (52) <sup>a</sup>	52 (32) <sup>a</sup>
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) <sup>a</sup>	29.0 (7.7)	22.3 (6.6) <sup>a</sup>	19.2 (6.8)
Sum of 4 skin-folds (mm)	74.4 (38.5) <sup>b</sup>	59.1 (29.2)	65.5 (32.3)	86.0 (42.0) <sup>b</sup>
BMI (kg/m <sup>2</sup> )	27.5 (6.6)	23.3 (3.6)	24.2 (5.2)	30.4 (6.7)

<sup>a</sup> $n = 1$ .

<sup>b</sup> $n = 2$ .

BMI: body mass index; SD: standard deviation.

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

Dependent variable	Ambulatory ( $n = 22$ )				Non-ambulatory ( $n = 28$ )			
	Independent variable	$\beta$	$p$ -value	$R^2$	Independent variable	$\beta$	$p$ -value	$R^2$
Physical activity (min/day)	–				Peak $\text{VO}_2$ (l/min)	0.398	0.04	0.16
Aerobic fitness (peak $\text{VO}_2$ in l/min)	Gender	–0.673	< 0.001	0.45	Gender	–0.512	0.003	0.42
Body fat (sum of 4 skin-folds in mm)	Gender	0.513	0.02	0.26	Physical activity (min/day)	0.365	0.03	
					Gender	0.725	< 0.001	0.53

$\beta$ : 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 peak $\text{VO}_2$  in non-ambulatory males with MMC (1.57 l/min) during arm exercise was 21% lower than the average peak $\text{VO}_2$  of males with paraplegic spinal cord injuries with lesions below T10 (1.98 l/min) (29). The low level of aerobic fitness must, however, be interpreted with caution because peak $\text{VO}_2$  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 peak  $\text{VO}_2$  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 peak  $\text{VO}_2$  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 thoracolumbar) 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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