J Rehabil Med 2020; 52: jrm00115

ORIGINAL REPORT

DETERMINANTS OF PHYSICAL ACTIVITY IN YOUNG WHEELCHAIR-USERS WITH SPINA BIFIDA

Manon A. T. BLOEMEN, PhD[1,2,3], Tim TAKKEN, PhD[2], Janke F. DE GROOT, PHD[1,2,4], Cas L. J. J. KRUITWAGEN, MSc[5], Rosanne A. ROOK, MSc[1,3], Rita H. J. G. VAN DEN BERG-EMONS, PhD[6] and Frank J. G. BACKX, MD, PhD[7]

From the [1]Research Group Lifestyle and Health, HU University of Applied Sciences Utrecht, [2]Child Development and Exercise Center, Wilhelmina Children's Hospital, University Medical Center Utrecht, [3]Master Program Pediatric Physical Therapy, HU University of Applied Sciences Utrecht, Utrecht, [4]Quality and Organization of Care, Netherlands Institute for Health Services Research, [5]Department of Biostatistics and Research Support, Julius Center, University Medical Center Utrecht, Utrecht, [6]Department of Rehabilitation Medicine, Erasmus MC University Medical Center Rotterdam, Rotterdam, and [7]Department of Rehabilitation, Physical Therapy Science and Sports, University Medical Center Utrecht, The Netherlands

Objective: **To explore associations between physical activity and peak oxygen uptake (VO_{2peak}), age, sex, and Hoffer classification in young wheelchair-users with spina bifida.**

Design: **Exploratory study.**

Subjects: **Fifty-three dutch children (age 5–19 years) with spina bifida who use a manual wheelchair.**

Methods: **For the dependent variable physical activity, data from 2 physical activity monitors were analysed: VitaMove data for 34 participants and Actiheart data for 36 participants. Time sedentary, time physically active, and time in moderate to vigorous physical activity were analysed. The Wheelchair Shuttle Test was used to measure VO_{2peak}. Univariate and multivariate regression analyses were performed. Independent variables were VO_{2peak}, age, sex, and Hoffer classification.**

Results: **Time sedentary and time physically active during a school day were influenced by age (β=0.326/β=–0.320) and Hoffer classification (β=0.409/β=–0.534) and during a weekend day by Hoffer classification (β=0.617/β=–0.428). Time in moderate to vigorous physical activity was influenced by Hoffer classification (β=–0.527) during a school day and by age (β=–0.600) during a weekend day.**

Conclusion: **Older age and the inability to walk negatively influence physical activity. Sex and VO_{2peak} were not associated with physical activity. These results imply that increasing cardiorespiratory fitness alone will not improve physical activity in young wheelchair-users with spina bifida.**

Key words: spinal dysraphism; children, adolescents; wheelchair; physical activity.

Accept Jul 27, 2020; Epub ahead of print Aug 17, 2020

J Rehab Med 2020; 52: jrm00115

Correspondence address: Manon A. T. Bloemen, HU University of Applied Sciences Utrecht, Heidelberglaan 7, 3501 AA, Utrecht, The Netherlands. E-mail: Manon.bloemen@hu.nl

LAY ABSTRACT

Children with spina bifida who are wheelchair-users are less physically active than their typically developing peers. It is important to understand relations between physical activity and other factors, so approriate interventions can be developed. We explored relations between physical activity and fitness, age, sex and severity of disability in children that were 5–19 years of age who were diagnosed with spina bifida and who are wheelchair-users. We found that older age and the inability to walk negatively influence physical activity. We did not find a relation between physical activity and fitness or physical activity and sex.

Papers published in the The Lancet describe physical inactivity as a global pandemic, and report the major negative health effects of physical inactivity on non-communicable diseases, such as coronary heart disease, type 2 diabetes and cancer (1–4). The authors conclude that an increase in physical activity (PA) could substantially improve health (2, 4, 5). While it is difficult for typically developing young people to develop and maintain a physically active lifestyle, it is even more difficult for young wheelchair-users with disabilities, such as spina bifida (SB) (2, 6–8). Young wheelchair-users experience a wide range of barriers, such as lack of support from other people, and lack of suitable play and sport facilities (9, 10). A recent study described low levels of PA in young wheelchair-users with SB (11).

Bouchard et al. have described a model in which relationships between PA, health-related fitness and health are presumed, with genetics and environmental variables as moderators (12). Cardiorespiratory fitness is part of health-related fitness, with peak oxygen uptake (VO_{2peak}) as the gold standard outcome measure. Studies in typically developing young people have shown weak to moderate associations (r=0.10 –r=0.45) between objectively measured PA and VO_{2peak} (13). Evidence in young people with physical disabilities is scarce, and shows conflicting findings for different clinical populations. Takken et al. (14) analysed the association between objectively measured PA and VO_{2peak} in ambulatory young people with juvenile idiopathic arthritis, and found a significant moderate correlation (r=0.3) between PA and VO_{2peak}. In contrast, Schoenmakers et al. found no correlation between self-reported PA and VO_{2peak} in ambulatory children and adolescents with SB (15).

Foundation of Rehabilitation Information

doi: 10.2340/16501977-2724

Age and sex seem to be important personal factors that influence PA in typically developing young people (4–18 years), with older age related to more time sedentary and less time physically active, and boys being more physically active than girls (5, 16, 17). A similar pattern is reported in ambulatory young people with cerebral palsy (CP), although the effect of sex is not always found (18–20).

In young people with CP, there is evidence of an association between PA and the severity of the disability, as classified by the Gross Motor Function Classification System (GMFCS) ranging from GMFCS level I (walking with minor disability) to GMFCS level V (transported in a wheelchair) (21, 22). Evidence suggests that a higher GMFCS level is associated with more time sedentary and lower PA (18, 20, 23). For children with SB, the modified Hoffer classification is used to classify the severity of the disability, ranging from normal ambulatory (Hoffer 1) to non-ambulatory (Hoffer 5) (24).

It is important to explore and understand the presumed relationships between PA and its determinants, in order to develop specific interventions for this population. To our knowledge, no studies have evaluated the relationship between PA and VO_{2peak}, age, sex and Hoffer classification in young wheelchair-users with SB. The study hypotheses are that PA would be positively associated with VO_{2peak} and negatively associated with age, that boys would perform better than girls, and that ambulatory participants would perform better than wheelchair-using participants.

The aim of this study was to analyse the associations between PA and VO_{2peak}, age, sex and Hoffer classification in young wheelchair-users with SB.

METHODS

Participants

This study is part of the *"Let's Ride…study"*, analysing physical fitness and physical behaviour in young wheelchair-users with SB (11, 25–27). Participants were recruited in the Netherlands and included if they were diagnosed with SB, were between 5–18 years of age at enrollment, used a manual wheelchair in their daily life or for long distances or sports participation, and were able to follow test instructions. Participants were excluded if they had any events (such as medical events or change of wheelchair), that might intervene with the outcomes of the testing (11, 25–27). Participants aged 12 years and over and all parents provided signed informed consent. The medical ethics committee of the University Medical Center Utrecht approved the study procedures (number 11-557).

Demographic and morphological parameters

Participants age, sex, type of SB, lesion level, sport activities, use of wheelchair, type of wheelchair and Hoffer classification (24) were registered through a standard questionnaire. The modified Hoffer classification categorized the ambulation level of people with SB as normal ambulatory (Hoffer 1), community ambulatory (Hoffer 2), household ambulatory (Hoffer 3), therapeutic ambulatory (Hoffer 4) and non-ambulatory (Hoffer 5) (24).

Body mass was measured using an electronic wheelchair scale (Kern MWS-300K100M, Kern & Sohn GmbH, Balingen, Germany) and height was measured using non-stretchable tape while seated using the arm-span length (middle finger-tip to middle finger-tip) as recommended in wheelchair-using children, due to the presence of contractures when lying supine. Body mass index (BMI) was adjusted by $\times 0.95$ for mid-lumbar lesions and $\times 0.90$ for high lumbar/thoracic lesions (28).

Physical activity equipment

PA was measured using 2 objective monitors: the VitaMove (2M Engineering, Veldhoven, The Netherlands) and the Actiheart (CamNtech Ltd, Papworth Everard, UK), as described in detail elsewhere (11). Participants wore both devices for 2 school days and 1 weekend day, from the time they got dressed until they went to bed, except during bathing and swimming. To avoid measurement bias, it was explained that the researchers wanted to know if these monitors were able to detect posture: it was not explained that the researchers were measuring PA. Subjects were instructed to continue their usual daily life.

VitaMove was used to measure the type of activities in this study. This is a wireless monitoring system with body-fixed accelerometers (Freescale MMA7260Q, Denver, CO, USA) and is valid for measuring mobility-related activities in young wheelchair-users (29). A detailed description of the configuration and analysis has been given elsewhere (11, 29–31).

The Actiheart was used to measure the intensity of PA. It was attached to the subject's chest by electrocardiogram electrodes (H99SG, Kendall, Covidien, Ireland) and is a valid device for measuring heart rate (HR) (32). A detailed description of the analysis has been given elsewhere (11)

Physical activity outcome measures

A minimum duration of one normal day with no peculiarities, such as illness, and a minimum wear time of 8 h per day were required to include the data in this study (33). To correct for differences in wear time, all PA data were calculated as a percentage of wear time.

Regarding the type of activities, sitting and lying were combined as time sedentary. The activities walking, running, wheeling, (hand)biking and non-cyclic moving were combined as time physically active.

As a measure of intensity, the following formula was used to determine the percentage of heart rate reserve (HRR):

$HRR = (HR_{measured\ by\ Actiheart} - HR_{rest})/(HR_{peak} - HR_{rest}) \times 100\%$ (34).

HR_{peak} was recorded during the Wheelchair Shuttle Test (WST) and the Graded Wheelchair Propulsion Test, which are both highly valid and highly reliable maximal exercise tests for young wheelchair-users with SB (25, 27). If a higher HR_{peak} was recorded in daily life by the Actiheart, this HR_{peak} was used (35). Before maximal exercise testing, HR_{rest} was measured while participants had to sit still for 10 min. If a lower HR_{rest} was measured in daily life by the Actiheart, this HR_{rest} was used (35). The time in moderate to vigorous physical activity (MVPA; >40% HRR, according to the American College of Sports Medicine), was used in the analyses (36).

Peak oxygen uptake

VO_{2peak} was measured during the WST (27). Cardiorespiratory responses were measured by a calibrated mobile gas analysis system (Cortex Metamax B3, Cortex Medical GmbH, Leipzig, Germany) and a HR monitor (miniCardio, Hosand Technologies Srl, Verbania, Italy). Absolute VO_{2peak} was calculated as the mean value over the highest 30 s and HRpeak was defined as the highest value during the WST. Data from the WST were included in the analysis if the subjective criteria for maximal exercise testing (signs of intense effort, such as sweating, facial flushing, clear unwillingness to continue despite encouragement) were met (37).

Statistical analyses

Data were initially checked for normality using histograms, Q-Q plots and Shapiro–Wilk tests. The Wilcoxon signed rank test showed no significant differences between the first and second school day, thus data were averaged (11, 33). If data for only 1 school day were obtained, this single school day was used. Due to significant differences between the school and weekend days, determinants of PA were analysed separately for school and weekend days (11).

The linearity of relationships between the dependent variables of PA (time sedentary, time physically active, and time in MVPA) and the independent variables age and VO_{2peak} were assessed using scatterplots. Thereafter, Spearman's rank correlations were used to test associations between PA and age and VO_{2peak} separately. Mann–Whitney U tests were used to test for significant differences in PA between boys and girls and between participants with Hoffer 1–3 and Hoffer 4–5 (38).

After univariate analyses, multiple linear regressions were performed, with PA as the dependent variable; a separate analysis was performed for every outcome measure of PA (time sedentary, time physically active and time in MVPA). The independent variables age, VO_{2peak}, sex, and Hoffer classification (Hoffer 1–3 vs Hoffer 4, 5) were entered in the regression if the p-value was ≤ 0.20 during univariate analysis (18). Because no *a priori* hypotheses was formulated about the order in which to include the independent variables, backward linear regressions were performed. Variables with a p-value > 0.1 were excluded and multicollinearity was checked for by assuring a tolerance of >0.1. Normality and homoscedasticity of the residuals was visually checked using histograms, QQ-plots and residual plots.

RESULTS

VitaMove data for 34 participants were available for time sedentary and time physically active, and Actiheart data for 36 participants were available for time in MVPA. A recent descriptive study gives a detailed overview of the reasons (technical dysfunctioning of the device, wear time < 8 h, skin irritation, holiday or illness) for missing data (11). Of the 36 participants with VitaMove data and 34 participants with Actiheart data, 30 met the subjective criteria for maximal exercise testing during the WST, so only these data were included in the analyses regarding the associations between PA and VO_{2peak}. For the VitaMove, a mean of 2.4 days were measured and for the Actiheart, a mean of 2.7 days were measured. Fig. 1 gives an overview of the included participants. Table I shows the participants' characteristics.

Univariate analyses are presented in Table II. Correlations with a p-value ≤ 0.20 were found between all outcome measures of PA and age, except for the time sedentary during a weekend day ($p = 0.251$). Correlations between PA and VO_{2peak} all showed p-values > 0.20. Differences between boys and girls and between Hoffer 1–3 and Hoffer 4–5 are shown in Table III. Sex differences with a p-value ≤ 0.20 were found for MVPA during a school day. Hoffer 1–3 vs Hoffer 4–5 showed p-values ≤ 0.20 for all outcome measures of PA.

The independent variables with a p-value ≤ 0.20 were used in the multiple regression analyses (Table IV). Time sedentary and time physically active during a school day was influenced by both age and Hoffer classification. During a weekend day, time sedentary and time physically active was influenced by Hoffer classification alone. MVPA was influenced by Hoffer classification during a school day and by age during a weekend day. Overall, young people with SB with Hoffer 4–5 were performing worse than Hoffer 1–3, and older participants were performing worse than younger participants.

DISCUSSION

This study investigated associations between PA and VO_{2peak}, age, sex and Hoffer classification in young wheelchair-users with SB. By measuring time seden-

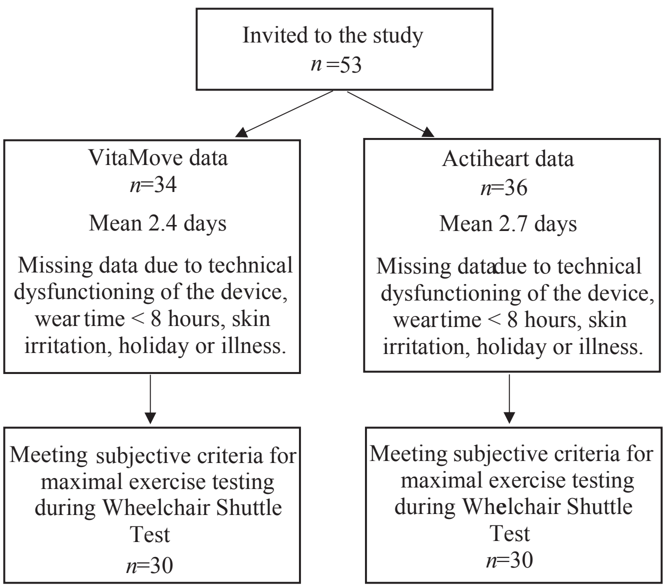

Fig. 1. Overview of included participants.

Table I. Participants' characteristics

	VitaMove $n=34$	VitaMove and maximal exercise testing WST $n=30$	Actiheart $n=36$	Actiheart and maximal exercise testing WST $n=30$
Age, years, months, mean (SD)	13.7 (3.2)	14.0 (3.0)	13.5 (3.6)	14.2 (3.1)
Weight, kg, mean (SD)	52.8 (18.1)	53.5 (16.3)	49.7 (19.5)	51.8 (16.9)
Height, cm, mean (SD)	159.1 (19.5)	160.8 (18.0)	155.5 (21.4)	158.6 (18.1)
Body mass index, kg/m^2, mean (SD)	23.9 (6.3)	24.0 (6.4)	23.2 (7.4)	23.8 (7.6)
Mass of wheelchair, kg, mean (SD)	19.6 (6.7)	18.8 (5.8)	19.1 (5.8)	19.4 (6.1)
Physical activity, % of wear time, median (IQR)				
Time sedentary school day	90 (8)	91 (8)	Na	Na
Time sedentary weekend day	96 (10)	96 (6)	Na	Na
Time physically active school day	8.9 (7)	8 (6)	Na	Na
Time physically active weekend day	4 (6)	3 (6)	Na	Na
Time in MVPA school day	Na	Na	9 (9)	8 (8)
Time MVPA weekend day	Na	Na	4 (10)	4 (8)
VO_{2peak}, l/min, mean (SD)	Na	1.20 (0.36) school day 1.28 (0.40) weekend day	Na	1.19 (0.31) school day 1.21 (0.31) weekend day
	n	*n*	*n*	*n*
Sex, boys/girls, *n*	20/14	18/12	21/15	17/13
Sports (No/1×week/2×week/3×week), *n*	7/14/9/4	4/13/9/4	6/17/9/4	3/15/8/4
Type, open/closed, *n*	33/1	30/0	33/3	29/1
Level of lesion, *n*				
Thoracic	5	4	6	4
Lumbar	29	26	29	26
Sacral	0	0	1	0
Hoffer classification, *n*				
Community ambulator (Hoffer 2)	2	1	4	1
Household ambulator (Hoffer 3)	3	2	6	5
Therapeutic ambulator (Hoffer 4)	3	3	2	2
Non-ambulator (Hoffer 5)	26	24	24	22

MVPA: moderate to vigorous physical activity; WST: Wheelchair Shuttle Test; SD: standard deviation; Na: not applicable; IQR: interquartile range.

tary, time physically active and time in MVPA, the study attempted to examine all aspects of PA behaviour and to explore possible associations with the different PA outcome parameters. Only age and Hoffer classification influenced the different PA outcome parameters. Hoffer classification appears to be the most important independent variable associated with PA, with children who use a manual wheelchair during daily life performing worse than children who use a manual wheelchair for sports or long distances; results that are in line with evidence in adolescents and young adults (mean age 21 years) with SB (38). The effect of age and severity of the disability on PA, but not sex, was also found by van Wely et al. in ambulatory young people with CP

Table II. Spearman's rank correlations between physical activity (time sedentary, time physically active, time in moderate to vigorous physical activity) and age and peak oxygen uptake

Spearman's rank correlations	Time sedentary school day	Time sedentary weekend day	Time physically active school day	Time physically active weekend day	Time in MVPA school day	Time in MVPA weekend day
Age, months	0.388 (***p*=0.028**)* $n=32$	0.229 ($p=0.251$) $n=28$	−0.369 (***p*=0.037**)* $n=32$	−0.286 (***p*=0.148**) $n=27$	−0.311 (***p*=0.069**) $n=35$	−0.512 (***p*=0.005**)* $n=29$
VO_{2peak}, (l/min)	−0.042 ($p=0.832$) $n=28$	0.026 ($p=0.872$) $n=23$	−0.013 ($p=0.946$) $n=28$	−0.057 ($p=0.795$) $n=23$	−0.091 ($p=0.633$) $n=30$	−0.259 ($p=0.222$) $n=24$

MVPA: moderate to vigorous physical activity; *n*: number; *$p<0.05$, bold: $p<0.200$

Table III. Differences between boys and girls and Hoffer 1–3 vs 4–5 for physical activity (time sedentary, time physically active, time in moderate to vigorous physical activity)

Mann–Whitney *U* test	Time sedentary school day $n=32$	Time sedentary weekend day $n=27$	Time physically active school day $n=32$	Time physically active weekend day $n=27$	Time in MVPA school day $n=35$	Time in MVPA weekend day $n=29$
Sex, boys/girls (%)	91/88 ($p=0.377$)	96/93 ($p=0.753$)	8/12 ($p=0.301$)	4/6 ($p=0.680$)	10/7 (***p*=0.158**)	4/5 ($p=0.948$)
Hoffer (1–3/4–5) (%)	67/92 (***p*=0.000**)*	86/96 (***p*=0.006**)*	18/8 (***p*=0.000**)*	7/3 (***p*=0.055**)	15/7 (***p*=0.002**)*	6/3 (***p*=0.199**)

MVPA: moderate to vigorous physical activity; n: number; *$p<0.05$, bold: $p<0.200$
Time sedentary, time physically active, and time in MVPA are depicted as % of wear time. Medians are presented for boys/girls and Hoffer 1, 2, 3/Hoffer 4, 5.

Table IV. Backward multiple regression analyses for physical activity (time sedentary, time physically active and time in moderate to vigorous physical activity), for school day and weekend day separately

	B	95% CI	*p*-value	Beta	Adjusted R^2
Time sedentary on a school day (age and Hoffer included)					
Constant	63.997	47.749; 80.246	0.000		
Age, month	0.082	–0.004; 0.167	0.062	0.327	
Hoffer	12.197	1.908; 22.486	0.022	0.409	0.257
Time sedentary on a weekend day (Hoffer included)					
Constant	76.456	66.333; 86.579	0.000		
Hoffer	18.738	7.882; 29.594	0.002	0.617	0.351
Time physically active on a school day (age and Hoffer included)					
Constant	21.982	15.237; 28.726	0.000		
Age, month	–0.037	–0.072; –0.001	0.045	–0.320	
Hoffer	–7.303	–11.574; –3.032	0.002	–0.534	0.391
Time physically active on a weekend day (Hoffer included)					
Constant	12.113	5.344; 18.882	0.000		
Hoffer	–7.576	–14.835; –0.316	0.042	–0.428	0.144
Time in MVPA on a school day (age, sex and Hoffer included)					
Constant	17.979	13.242; 22.716	0.000		
Hoffer	–9.809	–15.414; –4.204	0.001	–0.527	0.256
Time in MVPA on a weekend day (age and Hoffer included)					
Constant	30.720	18.320; 43.120	0.000		
Age, month	–0.138	–0.211; –0.065	0.001	–0.600	0.336

Time sedentary, time physically active and time in MVPA are % of wear time. Hoffer: Hoffer 1–3 is coded as 0 and Hoffer 4–5 as 1.
MVPA: moderate to vigorous physical activity; 95% CI: 95% confidence interval.

(19). However, there are also several studies of young people with CP that reported that boys performed better than girls (18, 20).

No relationship was found between PA and VO_{2peak} in young wheelchair-users with SB. These results differ from evidence in typically developing young people, in whom low-to-moderate relationships were reported between PA and VO_{2peak} (13). Comparing the current results with ambulatory children with SB, Schoenmakers et al. also found no correlations between self-reported PA and VO_{2peak} (15). Buffart et al. analysed associations between PA and VO_{2peak} in adolescents and young adults with SB and found a significant association (beta 0.398) only when separately analysing data for participants classified as Hoffer 4–5 (38). Secondary analyses with the study data, including only Hoffer 4–5 ($n=20$ and $n=25$), still showed no association between PA and VO_{2peak}. The current study hypothesis is that a minimum level of VO_{2peak} is required in order to be physically active. All participants in the current study probably have VO_{2peak} levels higher than this required minimum, as they all participated in normal daily life (going to school, playing with friends, etc.). However, the level of VO_{2peak} required for wheelchair-using children is not known. Another aspect may be the fact that, overall, the participants did not spend much time in MVPA, and that the intensity during activities, such as wheeling and (hand)biking, was quite low (11). In other words, the intensity of PA in wheelchair-using children with SB may be too low to have any effect on VO_{2peak}. On the other hand, the current results could also be due to low variability within our sample; it is possible that some children with SB have higher levels of MVPA and thus greater exercise intensity, leading to associations between PA and VO_{2peak}.

It appears that in young wheelchair-users with SB, variables other than VO_{2peak} or sex are important and influence PA. In addition, the current study found different results for associations with Hoffer classification and age between the different outcome measures of PA and between school days and weekend days. These results imply that different factors are important regarding type of activity and intensity of PA, and during school days compared with weekend days. Furthermore, considering the explained variance (26–39% on a school day and 14–34% on a weekend day), a large percentage of the variance in PA remained unexplained. We believe that there is a wide variety of personal and environmental factors that influence whether a person is physically active and that context and motivation play an important role, as reported in a qualitative study in young people with SB (10). Moreover, several of these factors will be individually based, as the context differs widely between children (9, 10). Therefore, understanding why a child does not participate in PA is complex and appears to be multifaceted. Participation in PA is part of behaviour; consequently this behaviour should be part of our clinical and research thinking when trying to understand PA in young wheelchair-users with SB. The International Classification of Functioning, Disability and Health – Child and Youth version (ICF-CY) provides a framework that helps clinical and research thinking, taking personal and environmental factors into account (39). However, the ICF-CY lacks a psychological model taking into account behavioural change. The Physical Activity for persons with Disability model (PAD-model) combines the ICF with the psychological model of Attitude, Social Influence and Self-Efficacy (40). This results in a model that enlarges the personal and environmental factors that are part of the ICF. These factors either facilitate or hinder the intention to participate in PA, but also participation itself, meaning that it converges with the term "behaviour". Due to the small sample size, the current study was only able to explore associations between PA and 4 independent variables based on previous literature. Future research is needed to further understand this behaviour and to clarify the influence of other personal and environmental variables on PA in young wheelchair-users with SB, helping to develop interventions to improve PA in this population.

Strengths and limitations

This study has several strengths and limitations. The study is, to our knowledge, the first to report on determinants for objectively measured PA in young wheelchair-users. Two highly valid monitors, analysing time sedentary and physically active, and time in MVPA were used. In addition, the study was able to measure VO_{2peak} directly using mobile gas analysis during a highly valid and reliable maximal incremental exercise test (27). A limitation was that not all VO_{2peak} data could be included, as some participants did not reach the subjective criteria for maximal exercise testing. It was not possible to use objective criteria for maximal exercise testing, as it is still unclear if objective criteria used in ambulatory children (HRpeak >180/min, peak respiratory exchange ratio >0.99) are applicable for wheelchair exercise testing in young people (37).

A second limitation was that the study was only able to collect PA data using 3 monitoring days with a minimum wear time of 8 h, because of feasibility reasons in young wheelchair-users with SB. This should be kept in mind when interpreting the results. Furthermore, some PA data were lost due to technical problems, and hence the sample size was relatively small, limiting the number of potentially independent variables. Therefore univariate analysis was used first, to select the independent variables that were used during multiple regression analyses (18). Finally, a cross-sectional design was used, so that no causal relationships could be established.

Conclusion

In conclusion, PA were found to be associated with age and Hoffer classification in young wheelchair-users with SB, with older age and the inability to walk negatively influencing PA. Sex and VO_{2peak} were not associated with PA in young wheelchair-users with SB. A large percentage of the variance in PA remains unexplained, implying that there are other numerous personal or environmental factors that should be explored in order to improve PA. Focusing on a healthy active lifestyle should start as early as possible, so that it becomes routine behaviour, especially in young people with SB who are classified as Hoffer 4 and 5. Clinicians should evaluate which individual personal and environmental factors might be important when aiming to improve PA. Increasing cardiorespiratory fitness alone does not seem to be the optimum intervention to improve PA.

ACKNOWLEDGEMENTS

All the children and adolescents and their parents are thanked for their participation in the *"Lets ride.... study"*.

Funding. This research was funded by Foundation Innovation Alliance – Regional Attention and Action for Knowledge circulation (SIA RAAK PRO), project number SIA RAAK PRO-4-03 and by a personal PhD grant (Manon A.T. Bloemen)from HU University of Applied Sciences Utrecht.

The authors have no conflicts of interest to declare.

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