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Accelerometer-determined physical activity and walking capacity in persons with Down syndrome, Williams syndrome and Prader–Willi syndrome☆

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ABSTRACT

In this study we describe by use of accelerometers the total physical activity (PA), intensity pattern and walking capacity in 87 persons age 16–45 years with Down syndrome (DS), Williams syndrome (WS) and Prader–Willi syndrome (PWS). Participants were recruited from all over Norway, and lived either with their parents or in community residences with support.

On average the participants generated 294 counts per minute (cpm) or 6712 steps per day, with most of the day spent in sedentary activity, 522 min/day, followed by 212 min/day in light PA, 71 min/day in lifestyle activity and 27 min/day in moderate-to-vigorous physical activity (MVPA). Inactivity was prevalent, as only 12% meet the current Nordic recommendations for PA.

When compared, no differences for total physical activity or time in MVPA were observed between the three groups. However, participant with DS spent a mean of 73 min/day less and 43 min/day less in sedentary activities compared to participants with PWS and WS, respectively, ($p = 0.011$, 95% CI: −10.9; −80.1). In addition the DS-group spent a mean of 66 min/day more in light PA than the PWS-group and 41 min/day more than the WS-group, ($p < 0.001$, 95% CI: 29.3; 79.7). Participants with PWS spent on average 30 min/day less in lifestyle activities compared to both participants with DS and WS, ($p < 0.001$, 95% CI: −14.2; −45.4). No association between total PA and BMI were observed. Males were more active than females across all diagnoses. Males accumulated on average 85 counts per minutes more than females, ($p = 0.002$, 95% CI: 33.3; 136.7), 2137 more steps per day, ($p = 0.002$, 95% CI: 778; 3496). The mean walking capacity during six-minutes was 507 m (SD 112 m) for males and 466 m (SD 88 m) for females. Distance walked during testing decreased with 33.6 m when comparing normal or underweight participants to overweight participants, and 78.1 m when comparing overweight to obese participants ($p < 0.001$ 95% CI: −40.4; −85.8). When adjusted for BMI no differences in walking capacity between the three genetic conditions were observed.

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1. Introduction

Regular physical activity (PA) and physical fitness improves functional ability, enhances independence and reduces the risk of non-communicable disorders such as cardiovascular disease, diabetes and several cancers (Nordic Council, 2005; WHO, 2010). The effectiveness of PA in relation to health depends on frequency, duration and intensity of activity, and is inherently difficult to assess due to its complex nature (Westerterp, 2009). Objective assessment of activity using activity monitors overcome many of the challenges related to self-reported measures of PA, such as social desirability bias and recall bias. Thus, accelerometers provide valid and reliable estimates of the degree, nature, and pattern of physical activity (Westerterp, 2009).

Walking capacity is a measurement for everyday physical capacity and cardiovascular fitness and is related to both levels of independence (Cowley et al., 2010) and long term health outcomes (Rasekaba, Lee, Naughton, Williams, & Holland, 2009). Walking is the most common form of exercise and PA in groups with intellectual disability (Draheim, Williams, & McCubbin, 2002). The six-minute walk test (6MWT) is a feasible and objective submaximal exercise test that assesses the distance a person can walk in six minutes (ATS Statement, 2002). In a variety of patient groups a distance less than 350 m walked in 6MWT is associated with increased mortality (Rasekaba et al., 2009).

Down syndrome (DS), Williams syndrome (WS) and Prader–Willis syndrome (PWS) are genetic conditions associated with mild or moderate intellectual disability. DS is caused by the presence of an extra copy or major portion of chromosome 21 (Hattori et al., 2000), WS by a deletion of the elastin gene on chromosome 7q11.23 (Morris, 2010) and PWS by the absence of paternally expressed genes in the 15q11-q13 chromosome region due to deletion, maternal disomy 15 (UPD) or an imprinting defect (Cassidy, Schwartz, Miller, & Driscoll, 2012). Even though molecular diagnosis is available today for all three conditions, diagnosis is in some patients based on clinical manifestations alone, especially in the adult patient population. Clinical manifestations of DS include typical physical, cognitive and behavioral characteristics which in most cases are easily recognized (Hunter, 2010). WS are recognized by typical facial features, short stature and connective tissue abnormalities in addition to a unique social and cognitive profile (Morris, 2010). Characteristics of PWS includes hypotonia, hypogonadism, a unique behavioral profile and childhood onset of hyperphagia that in absence of energy restriction will lead to obesity (Cassidy et al., 2012). It has previously been reported that individuals with DS and PWS have increased risk of inactivity and reduced physical capacity (Butler, Theodoro, Bittel, & Donnelly, 2007; Phillips & Holland, 2011; Temple & Stanish, 2009a), whereas specific knowledge on PA and everyday physical capacity in relation to WS is sparse.

In Norway all institutions for persons with intellectual disabilities closed, and individuals are offered supported living in community settings when moving from their parental homes (Beadle-Brown, Mansell, & Kozma, 2007). Independent living of individuals with intellectual disability in community settings has previously been associated with inactivity, low physical fitness and obesity (Doody & Doody, 2012; Draheim et al., 2002; Emerson, 2005; Hove, 2004; Martinez-Leal et al., 2011; Robertson et al., 2000). However, this knowledge is based on studies from countries with mixed types of living arrangements, where several confounding factors associated with persons living in institutions and persons living independently in communities, respectively, may occur. The complete deinstitutionalization in Norway opens an unique opportunity for studies of PA and physical capacity in a setting with increased focus on autonomy for all individuals, and for description of similarities and differences between subgroups associated with intellectual disability.

The aim of this study was (1) to describe levels of accelerometer-determined overall PA and sedentary behavior among persons with DS, WS and PWS; and (2) to investigate PA and walking capacity in relation to body mass index (BMI).

2. Methods

2.1. Ethical approval and recruitment procedures

Ethical approval for the study was granted by the Regional Committees for Medical and Health Research Ethics, South-East region.

Participants were recruited through existing information channels in relevant national-wide patient organizations, such as websites, membership bulletins etc. In addition a study-specific website was developed where general information, formal letter-to-participate and consent to participate-scheme was posted and available to download. In order to be eligible for inclusion, the individuals had to be between 16 and 45 years of age, and diagnosed with either DS, WS or PWS verified by standardized clinical methods (Holm et al., 1993; Preus, 1984) or by laboratory genetic testing. We used a convenient sampling frame. All participants who returned written informed consent to participate-scheme, signed by both the participant and legal guardian/parent were invited to participate.

2.2. Participants

A total of 96 participants, 40 with DS, 28 with WS and 28 with PWS from all over Norway participated in the study. All participants with clinical diagnosis were offered voluntary genetic testing for verification of their condition. Nine participants were eliminated from final analysis due to negative result from laboratory genetic testing and not for filling clinical criteria. In total 27 participants with DS have genetically verified trisomy 21 and 13 have clinically diagnosis. Of Participants with WS 18 have a genetically verified diagnosis whereas 7 have a clinically diagnosis (Preus, 1984). In the

PWS-group 21 have a genetically verified diagnosis (15 with deletion, 5 with UPD and 1 with unknown subtype) and one have clinically verified diagnosis (Holm et al., 1993). A total of 83 participants were included in PA assessment analysis and 87 in analysis of walking capacity.

2.3. Anthropometrics

The participant's weight, only wearing underwear, was measured twice on an impedance scale (Tanita BC-418MA, Arligton Heights, IL, USA) and recorded to the nearest 0.1 kg. Height was measured twice in upright position with heels placed into the wall and with head fixed in Frankfurt plane by use of a wall mounted standiometer (Seca 222, Birmingham, UK) and recorded to the nearest 0.1 cm. BMI was categorized by use of standard definitions of underweight, normal weight, overweight and obese. IsoBMI was used to define cut-offs for participants from 16 to 18 years of age. Due to small sample size, underweight participants ($n = 3$) were combined with normal weight in to a common normal or underweight category.

2.4. PA assessment

PA was assessed using the ActiGraph GT3X+ activity monitor (ActiGraph, Pensacola, FL, USA). The GTX3+ is lightweight and small and contains a micro-electro-mechanical system accelerometer (MEMS) that sampled the acceleration in three planes, by a 12-bit analog to digital converter, at 30 Hertz (user determined). The participants were instructed to wear the device in an elastic belt secured to the right hip in their home environment during all waking hours, except during swimming or bathing, for seven consecutive days. All participants were given two reminder-posters and encouraged to hang them up in visible places in their own homes. The accelerometers were returned by mail together with a registration of activities during the data collection period involving swimming, bicycling, resistance training or skiing activities. A total of 83 participants provided valid accelerometer recordings and were included in analysis. Drop-out was due to refusal to participate by two participants, medical safety concerns for one participant and lack of valid registration of activity in one of the returned accelerometers.

The acceleration data were stored in a raw and unfiltered format in units of gravity (Gs). Post-processing of the data included extraction of vertical axis data in 60-s epochs and derivation of the following variables: counts per minutes (cpm), steps taken per day, and minutes of intensity specific PA (sedentary behavior, light PA, and moderate-to-vigorous PA). The cut-points for intensity-specific PA have previously been used in large population studies on adults and older people (Hagstromer, Troiano, Sjostrom, & Berrigan, 2010; Hansen, Kolle, Dyrstad, Holme, & Anderssen, 2012). In order for a participant to be included in the analysis, a minimum of 4 days of at least 10 h per day of valid accelerometer recordings had to be achieved. Accelerometer non-wear time, defined as any consecutive strings of minutes with zero counts of at least 60 min with allowance for one interruption, was excluded from the analysis.

2.5. Assessment of walking capacity

Prior to the six-minute walk test (6MWT) all participants were familiarized with the test situation in two steps. First, the test was explained in general by use of pictures by researcher and participants were instructed to wear appropriate clothing and shoes. Second, a test technician demonstrated how to perform the 6MWT prior to actual testing. The participants performed two 6MWTs during the same day with a break in-between. All 6MWTs were conducted in a hallway using 30 m (m) laps. The rounding points of the lap were marked with cones and in addition each 3 m of the lap was marked on the floor. Testing was performed in accordance with standardized guidelines (ATS Statement, 2002) with exception of more frequent use of the encouragement component. Pulse oximetry (Creative Medical PC-60, Shenzhen, China) was used prior and right after testing. Two participants only completed one of the two tests; one due to medical safety concerns and the other due to lack of motivation for a second test.

2.6. Electronic questionnaire

Demographic data, information on place and type of residence and level of support were self-reported by use of an electronic feedback management tool (QuestBack, Oslo, Norway). The questionnaire was completed by the participants together with a parent or assistant. Level of support was categorized into four categories; 0–30 h of support per week, 31–60 h of support per week, more than 60 h of support per week and level of support unknown.

2.7. Statistical analysis

Descriptive statistics were calculated for all variables and assessed for normality and homogeneity of variance, and presented as mean with standard deviation (SD) or percent of population (%). To identify potential confounders that could interfere in the comparison between groups, initial analyses were performed by use of independent *t*-tests and analysis of variance (ANOVA) with use of turkey post hoc test where appropriate. Linear regressions adjusted for age and BMI where appropriate, were used to evaluate the PA and results of 6MWT in the different groups. In all analysis comparing the different genetic conditions PWS were used as reference group, with exception of analysis of sedentary PA and Light PA where DS were

used as reference group. Standard residuals and multicollinearity were investigated in the regression models. In the analysis including 6MWT only the results of the second walk test were used. For participants with only one completed walk-test, the results of this test were included. A p-value of less than 0.05 was regarded as statistical significant. All statistical analyses were performed using SPSS 19 (SPSS Inc., Chicago, IL, USA).

3. Results

3.1. Sample characteristics

Characteristics of the study population are presented in Table 1. More females ($n = 54$) than males ($n = 33$) participated; however we found no significant differences in age, BMI or type of residence between the genders. Participants living with parents were overall 10.2 years younger than the participants living in supported community settings, ($p < 0.001$, 95% CI −7.2; −13.2). Of the participants living in supported community residences; 67% lived in group-homes, 20% in their own apartments geographically close to other similar apartments, and 12% in independent housing facilities. In addition, 48% received 0–30 h per week of support, 18% from 31 to 60 h per week of support, 18% more than 60 h support per week, whereas the level of support were unknown in 15% of the participants. We found no differences in level of received support based on type of community residence. However, persons with PWS received more support than both DS ($p = 0.023$) and WS ($p = 0.01$).

A total of 78% of the study population was either overweight or obese. The DS-group had the highest average BMI followed by the PWS-group and WS-group. The difference in BMI by diagnosis adjusted for age was found to be 2.85 ($p = 0.004$, 95% CI 1.19; 4.51).

3.2. Total PA and time spent in different PA intensities

Participants achieved a mean 6.2 days of valid activity recordings and a mean daily accelerometer wear time of 13.9 h ± 1.4 h. PA by diagnosis and gender is presented in Table 2. Regardless of diagnosis most of the day was spent in sedentary activity 522 min/day (63%), followed by 212 min/day of light activity (25%), 71 min/day in lifestyle activity (9%) and 27 min/day of MVPA (3%). The total PA measured by cpm or steps per day did not differ significantly between the three groups. However a tendency that participants with PWS were less active than DS and WS was detected. For overall activity the PWS-group accumulated in mean 57 and 65 cpm less compared to participants with DS and WS, respectively, ($p = 0.055$, 95% CI: −119.2; 1.4). A large variation in PA was observed, especially in the WS-group and PWS-group, with an activity range of 103–788 cpm in the WS-group and range of 55–515 cpm in the PWS-group. In the DS-group the range was found to be 178–546 cpm. Time in different PA intensities is also presented in Table 2. Participant with DS spent a mean of 73 min/day less and 43 min/day less in sedentary activities compared to participants with PWS and WS, respectively, ($p = 0.011$, 95% CI: −10.9; −80.1). In addition the DS-group spent a mean of 66 min/day more than the PWS-group and 41 min/day more than the WS-group in light activities ($p < 0.001$, 95% CI: 29.3; 79.7). For lifestyle activities the PWS-group spent a mean of 30 min/day less compared to both DS and WS, ($p < 0.001$, 95% CI: −14.2; −45.4). No differences were found for time spent in MVPA. In all groups less than half of the time in MVPA was spent in bouts of 10 min or more.

Males were more active than females across all diagnoses. Males accumulated on average 85 cpm more than females ($p = 0.002$, 95% CI: 33.3; 136.7), 2137 more steps per day ($p = 0.002$, 95% CI: 778; 3496). Males also spent 23.6 more minutes a day in lifestyle activities ($p = 0.001$, 95% CI: 9.4; 37.9) and 13.8 more minutes a day in MVPA ($p = 0.003$, 95% CI: 4.7; 22.9). The largest gender difference was found among participants with WS with a mean of 147 cpm higher for males, whereas the mean gender differences were 63 cpm in DS-group and 56 cpm in PWS-group. No association with BMI-categories and total PA were observed. However some differences in time accumulated in sedentary activities and bouts of MVPA were found. Results for the BMI-categories are presented in Table 3. Under or normal weight participants spent a mean of 75 min/day and 62 min/day more in sedentary activities compared to overweight and obese, respectively ($p = 0.003$, 95% CI: 23.1; 105.2). On average obese subjects spent about 5 min/day less in both MVPA and bouts of MVPA compared to both normal weight and overweight subjects.

When participants living in supported community settings were compared to participants living with parents, no difference in overall PA or activity intensities was detected. Level of received support was not associated with the participants PA.

Table 1
Characteristics of study population by diagnosis.

	All ($n = 87$)	Down Syndrome ($n = 40$)	Williams syndrome ($n = 25$)	Prader–Willi syndrome ($n = 22$)
Females (%)	62.1	62.5	64.0	59.1
Community residence (%)	74.7*	60.0	84.0	90.9
Age (years)	28.5 (7.5)*	26.8 (7.5)	31.5 (6.2)	28.1 (7.5)
BMI (kg/m^2)	30.0 (6.7)*	31.8 (6.5)	26.6 (6.5)	30.7 (6.2)

Data presented as present of population or mean with (standard deviation).
* $p < 0.05$, when diagnosis were compared.

Table 2
Physical activity (PA) by diagnosis and gender.

	All ($n = 83$)	Downs syndrome ($n = 38$)	Williams syndrome ($n = 24$)	Prader–Willi syndrome ($n = 21$)
Total PA (counts/min)				
Male	347 (147.7)*	345 (93.4)	406 (204.7)	284 (142.7)
Female	262 (89.5)	282 (83.5)	259 (92.7)	228 (92.6)
All	294 (121.1)	306 (91.4)	314 (158.4)	249 (114.1)
Steps (steps/day)				
Male	8051 (3808)*	7584 (2440)	9954 (4985)	6725 (3948)
Female	5914 (2421)	6578 (2783)	5469 (1577)	5199 (2341)
All	6712 (3167)	6949 (2673)	7151 (3883)	5781 (3053)
Sedentary (min/day)				
Male	511 (86.9)	474 (91.0)†	524 (70.5)	559 (75.6)
Female	528 (76.3)	500 (63.0)	566 (91.2)	540 (63.9)
All	522 (80.3)	491 (74.4)†	534 (60.3)	564 (66.9)
Light PA (min/day)				
Male	227 (66.8)	255 (71.5)†	228 (50.4)	177 (48.0)
Female	203 (56.5)	232 (52.6)†	182 (48.5)	173 (49.2)
All	212 (61.4)	240 (60.3)†	199 (53.4)	174 (47.5)
Lifestyle PA (min/day)				
Male	86.0 (39.6)*	92.4 (29.6)	100.4 (53.5)	58.5 (24.6)††
Female	62.3 (25.6)	70.4 (14.8)	66.3 (35.5)	43.0 (18.2)††
All	71.2 (33.4)	78.5 (23.7)	79.1 (45.2)	48.9 (21.7)††
MVPA (min/day)[a]				
Male	35.8 (26.2)*	30.7 (17.7)	47.2 (34.4)	31.8 (27.7)
Female	22.0 (15.5)	22.5 (17.3)	20.5 (13.1)	22.7 (15.5)
All	27.1 (21.1)	25.5 (17.7)	30.5 (26.3)	26.2 (20.8)
Bouts MVPA (min/day)[b]				
Male	13.8 (15.8)	8.9 (10.5)	18.1 (17.3)	17.7 (20.9)
Female	9.5 (12.2)	9.6 (12.4)	6.9 (11.7)	12.1 (12.4)
All	11.1 (13.7)	9.4 (11.6)	11.1 (15.1)	14.2 (15.1)

All data are presented as mean with (standard deviation).
[a] Moderate-to-vigorous physical activity.
[b] All MVPA that occurred in bouts of 10 min or more with allowance for interruptions of 1–2 min.
* $p < 0.05$, when all males were compared to all females.
† $p < 0.05$, when diagnosis were compared by use of DS as reference group, adjusted for age and BMI.
†† $p < 0.05$, when diagnosis were compared by use of PWS as reference group, adjusted for age and BMI.

3.3. Adherence to physical activity recommendations

In the study population 12% meet the Nordic recommendations of physical activity (Nordic Council, 2005). More males than females met the recommendations, 14% and 10%, respectively. Diagnosis and gender specific prevalence are listed in Table 4. The highest proportions of participants meeting the PA recommendation were among participants with PWS (19%) followed by the participants with WS (13%) and participants with DS (8%).

Table 3
Physical activity (PA) by BMI category.

	Normal or underweight ($n = 18$)	Overweight ($n = 25$)	Obesity ($n = 38$)
Total PA (counts/min)	269.5 (111.9)	334 (136.4)	278 (110.3)
Steps (steps/day)	6907 (3104)	7629 (3683)	6010 (2704)
Sedentary (min/day)	575 (54.3)*	500 (83.2)	512 (79.3)
Light PA (min/day)	182 (36.9)	228 (67.9)	215 (62.3)
Lifestyle PA (min/day)	55.8 (22.2)	80.8 (40.3)	71.8 (30.6)
MVPA (min/day)[a]	29.2 (19.6)	33.2 (24.6)	22.1 (18.4)*
Bouts MVPA (min/day)[b]	13.4 (14.1)	13.7 (14.9)	8.3 (12.5)*

All data are presented as mean with (standard deviation).
* $p < 0.05$, when BMI categories were compared.
[a] Moderate-to-vigorous physical activity.
[b] All MVPA that occurred in bouts of 10 min or more with allowance for interruptions of 1–2 min.

Table 4
Prevalence of the population meeting current Nordic physical activity recommendation[a].

	≥30 min of daily MVPA[a]
All (%)	12.0
Down syndrome	
Male (%)	7.1
Female (%)	8.3
Williams syndrome	
Male (%)	22.2
Female (%)	6.7
Prader–Willi syndrome	
Male (%)	25.0
Female (%)	15.4

[a] Thirty minutes or more of daily moderate-to-vigorous physical activity in bouts of 8–10 min.

3.4. Walking capacity

We found no significant difference in walking capacity between participants living with parents compared to participants living in supported community residence. However, as shown in Table 5, we found an association in walking capacity with gender and BMI. Males walked on average 41.0 m longer ($p = 0.014$, 95% CI: 9.9; 86.3) compared with females. The mean distance walked during testing decreased with 33.6 m when comparing under or normal weight to overweight, and 78.1 m when comparing overweight to obese, ($p < 0.001$ 95% CI: −40.4; −85.8). Participants with DS walked a mean of 54.7 m shorter than participants with WS and a mean of 49.6 m shorter than participants with PWS. However when this was adjusted for differences in BMI, this association was no longer significant.

4. Discussion

This study provides objectively measured PA and walking capacity measures in three genetic conditions associated with intellectual disability recruited from all over Norway. The use of objective methods and standard protocols facilitate the opportunity for comparison with previous research in the general adult population in Norway. This study is to the best of our knowledge, the first to report on PA and physical capacity for persons with WS.

Total PA and time in different PA intensities was in general for males in our study population in accordance to findings from the general Norwegian adult population 20–64 years of age (Hansen et al., 2012). However, male participants with DS used less time in sedentary PA and more time in lifestyle activities compared to both other groups in our study and the adult Norwegian population. In addition we observed a tendency of male participants with WS to be more active and males with

Table 5
Results of six-minute walk test by gender, diagnosis, place of residence and body mass index category.

	6-Minute walk test
M (SD)	
All ($n = 87$)	481.1 (99.1)
Male ($n = 33$)	506.6 (112.3)
Female ($n = 54$)	465.6 (87.5)
Diagnosis	
Down syndrome ($n = 40$)	452.9 (91.6)
Williams syndrome ($n = 25$)	507.6 (81.9)
Prader–Willi syndrome ($n = 22$)	502.5 (118.9)
Place of residence	
With parents ($n = 22$)	498.9 (107.1)
Supported living in communities ($n = 65$)	475.1 (96.3)
Body mass index category	
Under or normal weight ($n = 19$)	545.0 (89.1)*
Overweight ($n = 26$)	511.4 (107.6)
Obesity ($n = 40$)	433.5 (72.5)

All data are presented as mean meter walked during test with (standard deviation).
* $p < 0.05$, when BMI categories were compared.

PWS, with exception for time in MVPA, to be less active. In contrast females, in our study generate 262 cpm compared to 345 cpm among females in the adult Norwegian population (Hansen et al., 2012). Again the participants with PWS were found to be least active and to spend more time in sedentary activities and less in light PA and lifestyle PA, whereas no difference for time in MVPA was observed.

The clear gender difference with respect to total PA was in accordance with some previous research of PA in individuals with intellectual disability (Bodde, Seo, Frey, Van Puymbroeck, & Lohrmann, 2013; Emerson, 2005; Phillips & Holland, 2011) but in contrast to others (Robertson et al., 2000; Stanish, Temple, & Frey, 2006; Temple & Stanish, 2009a) with no difference between genders. This was also in contrast to what is found in the general adult Norwegian population, with no difference in overall PA between genders (Hansen et al., 2012).

Compared to other more similar study populations, total PA and activity in MVPA was lower for all participants as well as participants with DS, than in a mixed population of individuals with intellectual disability (Phillips & Holland, 2011). Some of the difference seen in overall PA may be explained by the fact that we used a less strict definition of non-wear time. Time in MVPA, accumulated in bouts of 10 min was in accordance with other reports (Bodde et al., 2013). Our study groups spent more time in MVPA, 11.1 min/day versus 7.7 min/day, but this may be due to a lower mean BMI in our study population. Overall activity level in PWS-group was higher than what was reported by Butler et al. (2013), and can mainly be explained by more time in MVPA, which indicate more frequent participation in exercise activities by our participants with PWS. Likewise, for the DS-group, we report higher total PA than Temple (2009b). However caution should be used when comparing results from different countries due to differences in political, cultural or environmental factors.

Physical activity has several positive effects in these groups (Bartlo & Klein, 2011) and needs to be emphasized and facilitated, as only 12% meets the current recommendations. This was lower than reports by use of objective methods from the general adult Norwegian population where 22% females and 20% males met the Nordic recommendation (Hansen et al., 2012), and also lower than reports from adult populations of mild intellectual disability of mixed age and etiology, ranging from 14–33% (Hilgenkamp, Reis, van Wijck, & Evenhuis, 2012; Peterson, Janz, & Lowe, 2008; Temple, Frey, & Stanish, 2006). However this was higher than what was reported for mixed groups of persons with intellectual disability from UK (Phillips & Holland, 2011).

The mean walking capacity in our study population, 507 m for males and 466 m for females, was lower than the reference values in normal weight healthy adult's age 20–40 years; 800 m for males and 699 m for females (Gibbons, Fruchter, Sloan, & Levy, 2001). In total 7% of the participants walked shorter than 350 m, a result associated with increased risk for health complications (Rasekaba et al., 2009). However, the mean waking capacity was higher than what was reported from a population of DS with and without severe cardiac disease (Vis et al., 2009), in agreement with results from Casey et al. evaluating the use of the 6MWT in a group of persons with DS (Casey, Wang, & Osterling, 2012), and lower than reports from a younger group of persons with intellectual disability (Elmahgoub, Van de Velde, Peersman, Cambier, & Calders, 2012) and in participants attending a special Olympic program (Nasuti, Stuart-Hill, & Temple, 2013). No clear differences in walking capacity were detected for the different genetic conditions. This may indicate that the small differences seen in PA assessment regarding total PA and time in different PA intensities were not a result of different physical capacity related to differences in syndrome specific somatic abnormalities. Based on this, we suggest that differences observed are possibly related to personality and behavioral characteristics related to the genetic conditions. In addition we suggest that the large differences in PA seen within the groups, at least partly, to be explained by differences in structure, focus and opportunities for PA in the support systems (Temple, 2009b). Further studies regarding these issues are warranted to define more accurately determinants for both inactivity and activity in these populations in Norway.

A negative association between BMI and walking capacity was found, whereas no association was found between BMI and total PA. The lack of association with PA was in contrast to findings from populations studies were increased BMI was associated with a reduction in PA (Hansen, Holme, Anderssen, & Kolle, 2013), and further reduction in PA over time (Lakerveld et al., 2011). The association between walking capacity and BMI was in agreement with the suggestion by Doody and Doody (2012), that obesity leads to reduced everyday capacity and possible increased need for assistance. The lack of association with PA and BMI may be due to the fact that most participants were overweight or obese. However, this may also indicate other limiting variables for PA in these groups than in the general population. Others have suggested that unique barriers may be present in these groups and includes; lack of ability to perform physical activity due to reduced environmental availability of physical activity resources (Howie et al., 2012), lack of transportation, lack of guidance and social support (Frey, Buchanan, & Rosser Sandt, 2005; Stanish et al., 2006; Temple, 2009b). In addition to this physical restrictions or limitations related to somatic abnormalities associated with each genetic disorder.

The study has limitations. The convenient sampling frame challenges the generalization in the studied groups. However about one third of the known Norwegian population of both PWS and WS in relevant age category participated in the study.

Accelerometers have been found to be valid and reliable for measuring PA in adults (Westerterp, 2009). However, current algorithms for categorizing activity in intensity levels may underestimate energy expenditure and intensity in the study population due to reduced mean height, reduced coordination of movements, lower maximum oxygen consumption and increased mean weight (Agiovlasitis, Beets, Motl, & Fernhall, 2012; Hinckson & Curtis, 2013). In addition, the measurement precision varies with different type of activities. While good precision is achieved when assessing walking and running, underestimation of activity is prevalent in water activities and activities not involving movement of the hip. The activity diary indicated that there might be a small underestimation in the PA assessment for 16% of the participants due to either swimming activities (6%), bicycling (2%) or resistance training (8%).

The standard reference method to measure cardiovascular fitness is the maximal oxygen consumption test (VO2 max). However, 6MWT provides a global assessment of exercise capacity and may better reflect everyday activity capacity than laboratory tests (ATS Statement, 2002). It is both feasible and reliable for individuals with an intellectual disability (Elmahgoub et al., 2012; Vis et al., 2009) but level of intellectual disability have effects on distance walked (Casey et al., 2012; Vis et al., 2009). In our groups the use of the results as an exact estimate of walking capacity may possibly also be affected by diagnosis related to restrictions such as mental rigidity and fluctuations in motivation.

5. Conclusions

Inactivity was prevalent among individuals in all diagnosis and females were more inactive than males. When diagnoses were compared similar results were obtained for total PA and time in MVPA. However, the participants with DS were found to spend less time in sedentary activities and to spend more time in light PA, whereas participants with PWS spent less time in lifestyle PA. Walking capacity was lower in all groups tested compared to normal population but overall in accordance with investigations in similar groups. A negative association between BMI and physical capacity was detected, whereas no association between BMI and total PA was observed.

An increased focus on promoting PA, especially MVPA and lifestyle PA in these groups is warranted, also in Norway. In addition, Norway is a country with emphasis on autonomy for adults with intellectual disability, and more research investigating how best to facilitate and motivate for PA in individuals with intellectual disabilities are needed.

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