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Increasing physical activity in adult women with Prader–Willi syndrome: A transferability study

Alice Bellicha[1,2] | Muriel Coupaye[1,3] | Léonore Hocquaux[4] | Fanny Speter[4] | Jean-Michel Oppert[1,3] | Christine Poitou[1,3,5]

[1]Faculty of Medicine, Institute of Cardiometabolism and Nutrition, Sorbonne Université, Paris, France

[2]Laboratory Bioengineering, Tissues and Neuroplasticity – EA7377, University Paris-Est Créteil, Créteil, France

[3]Department of Nutrition, Faculty of Medicine, Assistance Publique-Hôpitaux de Paris, Pitié-Salpêtrière Hospital, Reference Center for Prader-Willi Syndrome, Sorbonne Université, Paris, France

[4]Siel Bleu, Paris, France

[5]INSERM, UMRS NutriOmics Team, Paris, France

Correspondence
Christine Poitou, Department of Nutrition, Pitie-Salpetriere Hospital, 47-83 boulevard de l'Hôpital, Paris 75013, France.
Email: christine.poitou-bernert@aphp.fr

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Abstract

Background: The present authors aimed (a) to objectively quantify spontaneous physical activity (PA) in adult patients with Prader–Willi syndrome (PWS) and (b) to evaluate the transferability of a home-based exercise training programme in these patients.

Method: Physical activity was compared between 10 adult women with PWS (PWS group) and 20 adult women with non-syndromic obesity (CON group, for cross-sectional comparison). In the PWS group, PA, body composition, walking capacity, quality of life and eating behaviour were then compared before and after a 16-week supervised exercise programme.

Results: The PWS group displayed lower PA and higher sedentary time compared to the CON group. Median attendance to exercise sessions reached 100% (Q1-Q3: 97%–100%) sessions. Moderate-to-vigorous PA and walking capacity increased after the programme without significant effect on body composition.

Conclusion: Supervised home-based exercise sessions are an effective strategy to improve PA in women with PWS who are less active than women matched for adiposity.

KEYWORDS
accelerometers, exercise training, obesity, physical activity, Prader–Willi syndrome

1 | INTRODUCTION

Prader–Willi syndrome (PWS) is the most frequent genetic syndrome causing marked obesity (Driscoll, Miller, Schwartz, & Cassidy, 1993) with a prevalence between one in 20,000 and one in 30,000 births (Bar et al., 2017; Lionti, Reid, White, & Rowell, 2015). PWS is a complex genetic neurodevelopmental disorder caused by an absence of expression of paternal-origin imprinted alleles on chromosome 15 (Driscoll et al., 1993). PWS is characterized by hyperphagia, hypotonia and early fat mass gain as well as decreased motor competencies and physical fitness (Capodaglio et al., 2009; Gross et al., 2017; Lloret-Linares et al., 2013). Subsequently, patients with PWS are prone to develop obesity with severe complications, such as cardiac or respiratory failure as well as physical disabilities (Cassidy, Schwartz, Miller, & Driscoll, 2012). Behavioural approaches for the management of PWS include the promotion of physical activity (PA) (Goldstone, Holland, Hauffa, Hokken-Koelega, & Tauber, 2008); however, day-to-day management of these patients with cognitive, and sometimes psychiatric disorders, is particularly challenging and little is known about PA interventions in this population.

In line with current PA guidelines for the general population (PAGAC, 2018), adult patients with obesity are advised to progressively reach a minimum of 150 min of moderate-intensity PA (MVPA) per week consisting of bouts of at least 10 min (Garvey et al., 2016; Jensen et al., 2014). In a recent study using accelerometers for the objective assessment of PA, only 15% of adult women with PWS

met these recommendations (Nordstrom, Hansen, Paus, & Kolset, 2013). This is in agreement with studies that reported a high prevalence of physical inactivity in women with non-syndromic obesity (Tudor-Locke, Brashear, Johnson, & Katzmarzyk, 2010). However, given that no comparison was made with a control group of subjects without intellectual disability in the study by Nordstrom et al., it is unclear whether PA is lower in women with PWS compared to women with non-syndromic obesity. Studies conducted in children and adolescents with PWS reported 30% to 55% lower levels of objectively measured PA compared to both normal-weight and non-syndromic obese controls (van den Berg-Emons, Festen, Hokken-Koelega, Bussmann, & Stam, 2008; Castner, Tucker, Wilson, & Rubin, 2014; Eiholzer et al., 2003), suggesting that the low level of PA observed in patients with PWS might be related to obesity per se as well as the physical and intellectual disabilities associated with PWS. Additionally, patterns of MVPA and sedentary behaviour (i.e. performed in short or longer bouts), which have received growing attention in recent years (Saint-Maurice, Troiano, Matthews, & Kraus, 2018; Shiroma, Freedson, Trost, & Lee, 2013), are not known in adult patients with PWS. Accelerometers are currently the monitoring method of choice for assessing habitual PA and sedentary (sitting) time, especially considering that PA self-reporting is a complex cognitive task that is particularly challenging in patients with PWS (Shephard, 2003).

Exercise training programmes have been found either to increase (Eiholzer et al., 2003; Schlumpf et al., 2006) or maintain (Rubin, Wilson, Dumont-Driscoll, & Rose, 2019) habitual PA (i.e. spontaneous PA performed independently of exercise training) in children with PWS but this has not been assessed in adults. As recently reviewed, different types of exercise programmes have been conducted in adults with PWS (Morales et al., 2019) and most programmes were supervised and performed in a residential or community setting (Cimolin et al., 2014; Grolla et al., 2011; Kaufman, Overton, Leggott, & Clericuzio, 1995; Shields, Bennell, Radcliffe, & Taylor, 2019; Silverthorne & Hornak, 1993). This literature review found that residential programmes that are organized several times per year and include dietary restriction, a high daily amount of supervised PA and a structured environment substantially reduced body mass over a long-term period (Cimolin et al., 2014; Grolla et al., 2011; Kaufman et al., 1995). A walking programme with two weekly sessions for six months was also effective to reduce body mass and body fat and improve cardiorespiratory fitness (Silverthorne & Hornak, 1993). Finally, a strength training programme including two weekly sessions increased muscle strength (Shields et al., 2019). Other programmes that were conducted at home for 6 months without any supervision led to improvements in gait patterns but not muscle strength or balance control (Capodaglio et al., 2011; Vismara et al., 2010). So far, no study has assessed the effectiveness and transferability of a home-based individually supervised exercise training programme in adult patients with PWS. Due to the specific impairment of executive functions and psychopathological features in this syndrome, integration of an exercise programme as part of daily routine along with individual interaction with health professionals might be especially important (Chevalere et al., 2015). Two case reports suggested that a home-based exercise training programme supervised by a PA instructor was feasible and well-accepted by children with PWS, although mixed findings were reported on changes in body mass and body composition, habitual PA and physical fitness (Amaro et al., 2016). A recent controlled study conducted in youth with PWS reported that a home-based exercise training programme facilitated by parents improved muscle strength and gross motor skill proficiency and decreased body mass in subjects who experienced an increase in objectively measured MVPA (Rubin, Wilson, Castner, & Dumont-Driscoll, 2019; Rubin, Wilson, Dumont-Driscoll, et al., 2019).

Given the lack of data on objective PA in adults with PWS vs. non-syndromic obesity and on the effectiveness of home-based supervised exercise training programme in adults with PWS, the present authors aimed (a) to quantify habitual PA using accelerometers, including an analysis of MVPA and sedentary bouts, in adults with PWS and in a control group of patients with non-syndromic obesity matched on age and adiposity; and (b) to evaluate the transferability and effectiveness of a home-based supervised exercise training programme on habitual PA, physical fitness and body composition.

2 | METHODS

2.1 | Study design and participants

The current observational study was conducted in a standard care setting and included 10 women with PWS (PWS group) and 20 women with non-syndromic obesity (CON group) to serve as a cross-sectional comparison. Only the women from the PWS group took part in the intervention.

Participants in the PWS group were recruited from June 2016 to January 2018 among patients with PWS and referred to a reference university clinic for patients with PWS. Inclusion criteria were genetically confirmed PWS, age between 18 and 60 years, BMI ≥ 30 kg/m^2 or body fat ≥ 40%, no treatment modifications in the previous two months and enrolment in a social security programme (patient or beneficiary). Considering the low prevalence of PWS and the low number of patients meeting inclusion criteria, all available patients willing to participate were included in the study. No published data allowed sample size calculations relevant to our hypotheses. Approval was obtained from the Ethics Committee of our institution, and the study was registered in the ClinicalTrials.gov website (Identifier: NCT03673813). Informed written consent was obtained from all subjects and caregivers prior to study inclusion. Participants in the CON group were recruited among bariatric surgery patients with non-syndromic obesity and had been included in a previous clinical trial (ClinicalTrials.gov website: NCT01113996). Data were collected from this group pre-surgery, and details of this study, which was entirely performed at our university medical centre, can be found elsewhere (Oppert et al., 2018). Each participant of the PWS group was individually matched with two control participants for gender, age and per cent body fat. Participants from the control

group did not participate in the exercise programme, and their data were used only for cross-sectional comparison of a given set of variables (PA, body composition, handgrip strength).

To address aim 1, the present authors performed a cross-sectional comparison between PWS and CON groups for habitual PA, body composition and handgrip strength, which were measured using similar procedures in participants from the PWS and CON groups. To address aim 2, the present authors compared pre- and post-interventional habitual PA, walking capacity, postural stability, health-related quality of life and eating behaviour in the PWS group. The primary outcome was changes in accelerometry-assessed habitual PA after the intervention. Secondary outcomes included the changes in body composition, physical function, health-related quality of life and eating behaviour following the intervention.

2.2 | Habitual physical activity measurement

PA and sedentary time were measured using a tri-axial GT3x Actigraph accelerometer (Manufacturing Technology, Inc.), as previously described (Oppert et al., 2018). Participants were asked to wear the accelerometers at the hip using an elastic belt for seven consecutive days during all waking hours except during water-based activities. To assess habitual PA, accelerometers were worn before and after the end of the intervention, outside of the exercise training programme. Therefore, exercise sessions were not included in the measurement of habitual PA. Data were considered valid when the accelerometer was worn for at least 3 days for at least eight hours each day. Freedson cutpoints were used to quantify sedentary behaviour (<100 counts/min), light-intensity PA (100 to 1951 counts/min) and moderate-to-vigorous PA (MVPA, ≥1952 counts/min) (Freedson, Melanson, & Sirard, 1998). Because wear time differed between PWS and CON groups, the time spent in each intensity category was expressed as the percentage of total wear time. Thus, the sum of sedentary time, light-intensity PA and MVPA equal to 100% of total wear time.

MVPA and sedentary bouts were defined as follows: MVPA bouts of one to four min ($MVPA_{1-4\ min}$), five to nine min ($MVPA_{5-9\ min}$) and at least 10 min ($MVPA_{\geq 10\ min}$); and sedentary bouts of one to nine min ($SED_{1-9\ min}$), 10 to 29 min ($SED_{10-29\ min}$) and of at least 30 min ($SED_{\geq 30\ min}$). $MVPA_{\geq 10\ min}$ bouts were defined as a minimum of 10 consecutive min above 1952 counts/min with allowance of two min below this threshold (Saint-Maurice et al., 2018; Tudor-Locke, Camhi, & Troiano, 2012). $SED_{\geq 30\ min}$ bouts were defined as a minimum of 30 consecutive min below 100 counts/min (Tremblay et al., 2017). Participants reaching 150 min per week of MVPA in ≥10-min bouts were considered compliant with PA guidelines (PAGAC, 2018).

2.3 | Anthropometry and body composition measurement

Body composition was estimated by whole-body fan-beam DXA scanning (Hologic QDR 2000, Hologic), as previously described (Oppert et al., 2018). Fat mass index and fat-free mass index were calculated as fat mass and lean body mass, respectively, divided by height squared (kg/m^2) (Schutz, Kyle, & Pichard, 2002).

2.4 | Physical function measurement

Walking capacity was assessed with the six-min walk test. The test was performed according to the American Thoracic Society recommendations (American Thoracic Society, 2003), with the exception of more frequent encouragements, as previously described in patients with PWS (Nordstrom et al., 2013). Participants were asked to walk for six min and to cover as much ground as possible between two cones positioned 30 m apart. Handgrip strength was measured with a handgrip dynamometer (Jamar, Sammons Preston Rolyan, Bolingbrook). Participants sat in a chair with arms close to their sides and elbow flexed at 90° and were asked to provide five maximal attempts with each hand (Roberts et al., 2011). The highest value obtained was kept for analyses. Static balance control was assessed using an unipedal stance test. Participants were asked to stand barefoot on one leg for as long as possible, with the eyes open and the other limb raised (Springer, Marin, Cyhan, Roberts, & Gill, 2007). Participants performed three trials on each leg, and the best trial was recorded. For consistency, all tests were undertaken by the same investigator (AB) for participants in the PWS group.

2.5 | Quality of life, eating behaviour assessment in patients with PWS

Health-related quality of life was assessed with the 12-item short-form (SF-12) questionnaire and summarized in a physical and mental component score ranging from zero (poor) to 100 (good) (Gandek et al., 1998). Eating behaviour was assessed with the Hyperphagia questionnaire, a tool specifically designed for assessing eating behaviour in patients with PWS (Dykens, Maxwell, Pantino, Kossler, & Roof, 2007).

2.6 | Exercise training programme in patients with PWS

Patients with PWS participated in a 16-week exercise training programme. The programme was conceived and implemented in partnership with Siel Bleu, a non-profit organization specialized in delivering PA intervention to patients with chronic diseases and older adults (El-Khoury et al., 2015). The programme included two exercise sessions per week, conducted at home on an individual basis and supervised by a specifically trained PA instructor. Patients were supervised by the same instructor throughout the programme. As recommended in PA guidelines for adults with disabilities available at the time the study was designed, exercise sessions were based on a combination of endurance-based and muscle-strengthening activities of moderate intensity (Department of Health & Human Services, 2008). Each training session lasted one hour and included 10 min of a light endurance warm-up, 30 min of moderate-intensity endurance training, 15 min of strength training and five min of stretching and cooling down. To make training

sessions more enjoyable and improve adherence, endurance activities varied along the programme and included walking, Nordic walking, orienteering, running, step or stair climbing, ball games (football, basketball, volleyball) and racket games (badminton, tennis table). To supervise intensity, PA instructors regularly asked patients to rate their level of effort and verified that patients were able to talk while performing the activity (PAGAC, 2018). Strength exercises targeting major muscle groups were performed with training equipment such as free weights or elastic bands (sit-to-stand transitions, squats, biceps and triceps curls with free weights, or rowing with an elastic band). Exercises were performed either indoors or outdoors close to home, depending on weather conditions. In addition to the training programme, patients and their families were encouraged to increase habitual PA. The programme was provided free of charge to the participants.

2.7 | Transferability of the exercise training programme

Transferability was assessed using the RE-AIM framework (Glasgow, Vogt, & Boles, 1999), as used previously by our research group in a French setting (Bellicha et al., 2016). The following dimensions were described: reach (participation to exercise sessions), effectiveness (patient and family satisfaction assessed with a five-point scale and potential adverse consequences, e.g. injuries, temper outbursts, behavioural changes), adoption (perception of PA instructors involved in the study), implementation (the extent to which the programme was delivered as intended) and maintenance (the number of participants who maintained the exercise sessions after the end of the study).

2.8 | Procedures

For the women from the PWS group, all the measures were implemented prior to the intervention and within 2 weeks following the intervention. For the control group, data were collected at a single time point, during the pre-operative visit at our university clinic, as part of standard care.

2.9 | Statistical analysis

Given the limited sample size and the non-normality of most variables, data are presented as median (25th–75th percentile) and non-parametric tests were used. To address aim 1, characteristics of participants in the PWS and CON groups were compared with Mann–Whitney test for continuous variables and with chi-square

TABLE 1 Body composition, quality of life and eating behaviour in patients with Prader–Will syndrome before and after the exercise programme and in controls

	CON group (*N* = 20)	PWS group Pre-intervention (*N* = 10)	*p*-value*	PWS group Post-intervention (*N* = 10)	*p*-value**
Age, years	30 (26;36)	28.8 (24.2;33.0)	.25	–	–
Height, cm	165 (158;168)	151 (143;156)	**<.001**	–	–
Body weight, kg	121.2 (106.6;133.9)	82.3 (74.3;107.8)	**.002**	84.5 (73.3;99.0)	.15
BMI, kg/m^2	43.4 (40.9;48.3)	37.2 (34.3;45.8)	**.05**	37.1 (34.5;42.0)	.16
Waist circumference, cm	–	106.0 (94.3;116.4)	–	106 (89;106)	.47
Body fat, %	52.4 (50.1;53.7)	51.9 (49.2;54.7)	.42	52.8 (49.4;54.2)	.42
Fat mass, kg	62.9 (56.0;70.4)	40.2 (39.3;58.6)	**.004**	41.3 (36.8;55.2)	.15
Lean body mass, kg	57.8 (51.2;62.2)	50.1 (34.1;46.8)	**<.001**	41.1 (33.2;42.8)	.50
Fat mass index, kg/m^2	22.2 (21.3;25.0)	19.8 (16.8;25.0)	.08	19.2 (16.9;23.4)	.13
Lean body mass index, kg/m^2	21.2 (18.3;22.7)	17.0 (16.4;18.7)	**.006**	17.2 (16.3;18.2)	.46
Quality of life					
Physical score	–	49.6 (48.5;53.5)	–	54.1 (51.5;56.3)	.06
Mental score	–	51.6 (41.8;58.2)	–	55.3 (42.2;60.4)	.50
Dykens questionnaire[a]					
Hyperphagic behaviour	–	8.5 (6.0;10.8)	–	7.0 (6.0;7.75)	.08
Hyperphagic drive	–	7.5 (6.0;11.8)	–	9.0 (6.8;11.0)	.39
Hyperphagic severity	–	3.0 (3.0;4.0)	–	3.0 (2.0;5.0)	.50

Note: Data are median (25th; 75th percentile).
Bold values indicate significance with $p < .05$.
Abbreviations: CON group, control group; PWS group, patients with Prader–Willi syndrome.
[a]Dykens questionnaires were obtained before and after the programme in 8 participants.
**p*-value from Mann–Whitney test for cross-sectional comparison of characteristics between the PWS group and the control group.
***p*-value from Wilcoxon test for comparison of the PWS group before vs. after the PA programme.

TABLE 2 Objectively measured physical activity and physical function in patients with Prader–Will syndrome before and after the exercise programme and in controls

	CON group (*N* = 20)	PWS group Pre-intervention (*N* = 10)	*p*-value*	PWS group Post-intervention (*N* = 10)	*p*-value**
Physical function					
6-min walk test distance, m	–	398 (366;484)	–	450 (378;509)	**.02**
Handgrip strength, kgF	33 (28;38)	15 (13;18)	**<.001**	15 (14;18)	.48
Stance test, eyes open, s	–	8 (3;28)	–	9 (5;33)	**.02**
Physical activity[a]					
Activity counts, counts/min	334 (292;384)	211 (141;333)	**.02**	311 (188;361)	.11
Sedentary time, % wear time	58 (53;64)	72 (69;75)	**<.001**	68 (67;75)	.41
Light-intensity PA, % wear time	38 (32;45)	24 (23;28)	**<.001**	26 (21;29)	.34
MVPA, % wear time	3.2 (2.6;4.5)	1.7 (0.8;5.0)	.11	4.0 (1.1;6.1)	**.02**
MVPA in 10-min bouts, min/week	20 (0;68)	5 (0;101)	.38	90 (21;238)	**.03**

Note: Data are median (25th; 75th percentile).
Bold values indicate significance with $p < .05$.
Abbreviations: CON group, control group; MVPA, moderate-to-vigorous physical activity; PWS group, patients with Prader–Willi syndrome.
[a]Physical activity data were obtained in seven participants in the PWS group after the exercise programme.
*p-value from Mann–Whitney test for cross-sectional comparison of characteristics between the PWS group and the control group.
**p-value from Wilcoxon test for comparison of the PWS group before vs. after the PA programme.

test for categorical variables. To address aim 2, characteristics of participants in the PWS group before and after the exercise training programme were compared with Wilcoxon test (continuous variables) and with McNemar test (categorical variables). Pre- to post-intervention changes were expressed as mean (*SD*). The null hypothesis was rejected at $p < .05$. Analyses were performed using R version 3.4.3 (R Core Team, 2017).

3 | RESULTS

3.1 | Aim 1: Comparing clinical characteristics, physical activity and sedentary behaviour between women with PWS and women with non-syndromic obesity

As expected, patients with PWS were comparable to patients from the CON group in terms of age and percentage body fat but were shorter and had lower BMI, lean body mass and handgrip strength (all $p < .05$) (Table 1). Nine participants were given replacement therapy for gonadotropin deficiency and 3 participants for thyrotropin deficiency. Four participants had received growth hormone (GH) treatment during childhood; however, none had received GH treatment in adulthood.

The accelerometer was worn during seven days in both groups for a median duration of 12.1 hr/day in the PWS group and 13.5 hr/day in the CON group ($p < .001$) (Table 2). Median activity counts were 37% lower in the PWS group compared with the CON group ($p < .05$). Sedentary time was higher in the PWS group, while light-intensity PA was lower in patients with PWS (both $p < .001$). Duration of MVPA was not significantly different between groups, although it tended to be lower in the PWS group ($p = .11$). $MVPA_{\geq 10\ min}$ was low in both groups, with no significant difference between groups. Only two participants in the PWS group and one participant in the CON group were compliant with PA guidelines ($p = .52$).

MVPA patterns did not differ between the PWS and CON groups (Figure 1). In the PWS group, $MVPA_{1-4\ min}$ represented on average 61% of total MVPA, $MVPA_{5-9\ min}$ represented 21% of total MVPA and $MVPA_{\geq 10\ min}$ represented 18% of total MVPA (vs. 60%, 27% and 14% in the CON group, respectively). Sedentary patterns differed between groups (Figure 2): $SED_{1-9\ min}$ bouts were lower in the PWS group (31% of total sedentary time vs. 44% in the CON group, $p < .001$) and $SED_{\geq 30\ min}$ were higher (41% of total sedentary time vs. 18% in the CON group, $p < .001$).

3.2 | Aim 2: Assessing the effectiveness and transferability of the exercise training programme in women with PWS

After the exercise training programme, walking capacity increased by mean (*SD*) 29 (37) m ($p = .02$) and stance time increased by 4 (6) s ($p = .02$) (Table 2) although no effect was found on body weight and composition (Table 1). Total MVPA increased by 11 (13) min/day ($p < .05$) and $MVPA_{\geq 10\ min}$ by 86 (97) min.week ($p = .03$). After the programme, three participants (43%) were compliant with PA guidelines. The percentage of MVPA accumulated in $MVPA_{\geq 10\ min}$ increased to 36% of total MVPA ($p = .03$) while MVPA accumulated in $MVPA_{5-9\ min}$ tended to decrease ($p = .05$) (Figure 1). The physical component of quality of life tended to increase after the programme but no effect was found on, sedentary patterns or eating behaviour.

Patient participation in exercise sessions was very good, with a median (25th–75th percentile) number of exercise sessions attended of 32 (31–32) sessions. Participants and their families were very satisfied with the programme as participants scored their satisfaction

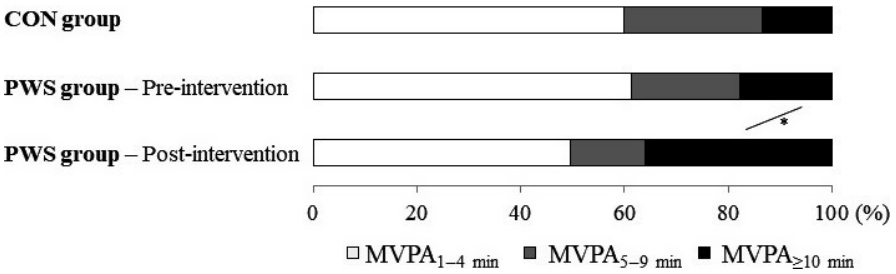

FIGURE 1 Distribution of MVPA in very short, short and prolonged bouts. *In the PWS group, significantly different after the programme compared to pre-intervention values ($p < .05$, p-value from Wilcoxon test). Abbreviations: CON group, control group; MVPA, moderate-to-vigorous physical activity; PWS group, patients with Prader–Willi syndrome

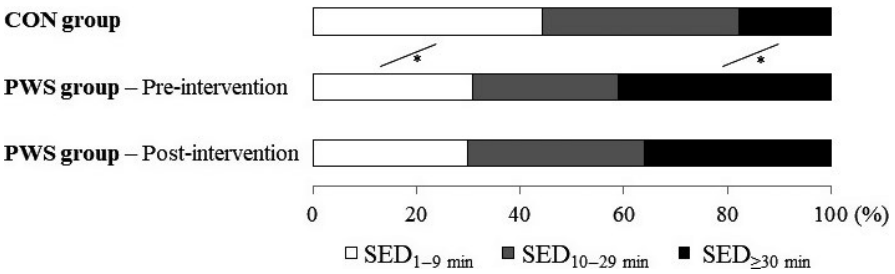

FIGURE 2 Distribution of sedentary time in very short, short and prolonged bouts. * Significantly different from the CON group ($p < .001$, p-value from Mann–Whitney test). Abbreviations: CON group, control group; PWS group, patients with Prader–Willi syndrome; SED, sedentary time

4.4 (4.0–5.0) on a five-point scale and their relatives scored theirs 4.8 (4.1–5.0). No adverse events occurred during the programme. All of the five PA instructors who participated in the programme had at least two years of experience with the management of patients with chronic diseases and physical and/or intellectual disability, and all received specific training on PWS before the programme. The main difficulties reported by instructors were related to psychological disorders associated with PWS (emotional lability and immaturity, mental rigidity), physical disorders (increased fatigability, balance disorders and reduced motor skills) and a lack of motivation. One-to-one sessions were seen as an important aspect to personalize exercises and general advice about PA and to improve the patient–care provider relationship. All instructors declared that they sometimes had to adapt the exercise sessions according to the patient's motivation, mood or fatigue. Mainly, the type of endurance-based activities performed was chosen by the instructor in agreement with the patient to make the sessions more enjoyable and to increase motivation. The most frequently performed activities were walking (66% sessions), Nordic walking (19% sessions), ball games (12% sessions), racket games (6% sessions), step or stair climbing (4%), orienteering (3% sessions), cycling (3% sessions) and running (2% sessions). Finally, three participants continued exercise sessions (two to two weekly sessions) at their own expense with the same organization after the end of the study, that is from four months after the end of the programme.

4 | DISCUSSION

This study provides a detailed analysis of habitual PA and sedentary patterns in adult women with PWS. Compared to control women matched for age and adiposity, total PA assessed with accelerometers was lower and sedentary time was higher in patients with PWS. Our data also demonstrate the effectiveness and feasibility of an exercise programme including one-on-one sessions of endurance and strength exercises for patients with PWS, when conducted at the participant's home or community setting and supervised by the same PA instructor throughout the programme.

In our sample of participants with PWS, the amount of PA assessed as accelerometer counts per min was found to be 37% lower in patients with PWS compared to the CON group. Sedentary time accounted for almost two-quarters of accelerometer wear time, which was comparable to values previously reported in adult women with PWS (Nordstrom et al., 2013). This was, however, higher than values obtained in our CON group or previously reported in subjects with severe obesity (King et al., 2015). Participants in the PWS group were also characterized by more prolonged, uninterrupted sedentary bouts. Prolonged bouts of 30 min or longer accounted for more than 40% of total sedentary time, more than twofold the values obtained in the CON group. The present authors found no other study examining PA in PWS to compare these data to. The increase in sedentary time in the PWS group was mainly explained by a decrease in light-intensity PA, and to a lesser extent, by a decrease in MVPA. Previous studies have reported a lower duration of light-intensity PA in youth with PWS and a trend for a lower duration of MVPA (Castner et al., 2014; Rubin, Duran, Haqq, Gertz, & Dumont-Driscoll, 2018).

After the exercise training programme, total MVPA increased by more than twofold and MVPA spent in bouts of ≥10 min increased from five to 90 min per week. Despite this substantial increase in MVPA, more than half of the participants remained insufficiently active. Sedentary time and patterns, as well as light-intensity PA, did not change after the programme. Growing evidence suggests

that breaking prolonged sedentary bouts with short bouts of light-intensity PA elicits cardiometabolic benefits (Duvivier et al., 2018). Interestingly, the cardiometabolic risks associated with an increase in sedentary time might be higher in subjects with lower levels of MVPA (Biswas et al., 2015). Improvements in cardiometabolic factors have been reported after a PA intervention in patients with PWS (Kaufman et al., 1995; Rubin et al., 2018). It should be noted, however, that the study by Kaufman et al. (1995) also included a nutritional intervention, and the study by Rubin et al. (2018) reported higher metabolic benefits in patients who experienced no gain or a decrease in body fat. Thus, the cardiometabolic benefits described cannot be attributed solely to the PA programme. More studies are needed to better understand the contribution of PA to the cardiometabolic health of patients with PWS.

Another important objective of this study was to assess the transferability of a four-month exercise training programme in participants with PWS. Participation in exercise sessions was excellent, as well as the satisfaction from both participants and their families. Studies that implemented home-based exercise training programmes, either supervised or non-supervised, usually reported high participation in exercise sessions compared to programmes based outside the home (Amaro et al., 2016; Capodaglio et al., 2011; Rubin, Wilson, Dumont-Driscoll, et al., 2019). Instructors involved in this programme reported they had to adapt to the participants' psychological and cognitive disorders and physical limitations associated with PWS, and sometimes to patients' fatigue and lack of motivation. Individual interaction was found to be very important to build-up confidence, improve the patient–health professional relationship and promote self-esteem in these very sensitive patients. This study showed the potential of transferability of a home-based supervised exercise training programme in patients with PWS, even though the sample size was rather small. The data also suggest the need to involve experienced PA instructors with a well-developed capacity to adapt exercise programmes and make the sessions enjoyable with these patients.

The programme was effective to improve physical function although no change in body weight or body composition was found. Previous studies that reported significant weight loss after a PA intervention in adult patients with PWS also included a dietary intervention (Cimolin et al., 2014; Grolla et al., 2011). Given the major importance of restricting food intake for obtaining negative energy balance, the expected benefit of PA alone should not be to lose weight but rather to focus on improving physical function or metabolic outcomes. A significant 30-m improvement in walking distance was observed after the programme, which is consistent with studies that previously examined a three- to four-month exercise training programmes in subjects with severe obesity (Baillot et al., 2014; Marcon et al., 2017). This suggests that exercise training could be equally effective to increase walking capacity in patients with PWS or with common obesity. To increase the generalizability of such findings, future research should investigate whether this type of home-based supervised exercise training programme is effective in adult patients with other forms of genetic (e.g. Bardet-Biedl) or hypothalamic lesional (e.g. craniopharyngioma) obesity, who represent up to five per cent of obesity cases (Huvenne, Dubern, Clement, & Poitou, 2016), and more broadly in individuals with intellectual disabilities, who often present with overweight or obesity (Flygare, Ljunggren, Carlsson, Pettersson, & Wandell, 2018).

4.1 | Study strengths and limitations

The strengths of this study include the objective monitoring of habitual PA by accelerometers over a relatively long (more than six days) period in a group of adult women with PWS and in a control group of subjects with non-syndromic obesity carefully matched on gender, age and adiposity. Accelerometer data processing was performed using the same procedures, increasing the comparability of data between groups. The min-by-min analysis of accelerometer data provided new insights about MVPA and sedentary patterns in patients with PWS, highlighting the added value of objective measure of PA for improving our understanding of PA patterns and guiding patient management. However, the cutpoints used to define sedentary time and light-, moderate- and vigorous-intensity PA were initially validated in young and non-obese individuals (Freedson et al., 1998), and have not been validated in patients with PWS. According to previous reports (Agiovlasitis et al., 2011), the relation between counts per min and oxygen consumption differs in patients with intellectual disabilities, suggesting that traditional cutpoints might not be adapted to these subjects. More research is needed to validate specific intensity cutpoints in patients with PWS, and more broadly in patients with intellectual disabilities (Leung, Siebert, & Yun, 2017). In addition, the sample size was limited and only women were included in the analyses, which prevent us from extrapolating results to all patients with PWS. Finally, the control group did not receive the intervention and was not used as a control group to compare the effectiveness of the exercise programme in PWS vs. non-syndromic obesity. Hence, although unlikely, the present authors cannot exclude that the improvements observed in the PWS group after the intervention might have been due to other factors.

5 | CONCLUSION

This study shows substantially lower objectively measured total PA and higher sedentary time, especially in uninterrupted bouts of 30 min or longer, in women with PWS compared to control subjects with common obesity. Participation in the home-based supervised exercise training programme was very high, showing the feasibility and transferability potential of such programmes in patients with PWS. The 16-week programme was found to be effective in increasing habitual MVPA and improving walking capacity, although body weight and body composition were not changed. This increase in PA is likely to have beneficial health effects over time, showing the adjunct value of including a specifically designed PA

programme in the clinical management and rehabilitation of patients with PWS.

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CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

ORCID

Alice Bellicha https://orcid.org/0000-0002-5572-487X

Jean-Michel Oppert https://orcid.org/0000-0003-0324-4820

Christine Poitou https://orcid.org/0000-0001-7769-6331

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