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Gait strategy in genetically obese patients: A 7-year follow up

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ABSTRACT

The aim of this study was to quantitatively evaluate the change in gait and body weight in the long term in patients with Prader–Willi Syndrome (PWS). Eight adults with PWS were evaluated at baseline and after 7 years. During this period patient participated an in- and out-patient rehabilitation programs including nutritional and adapted physical activity interventions. Two different control groups were included: the first group included 14 non-genetically obese patients (OCG: obese control group) and the second group included 10 age-matched healthy individuals (HCG: healthy control group). All groups were quantitatively assessed during walking with 3D-GA. The results at the 7-year follow-up revealed significant weight loss in the PWS group and spatial–temporal changes in gait parameters (velocity, step length and cadence). With regard to the hip joint, there were significant changes in terms of hip position, which is less flexed. Knee flexion–extension showed a reduction of flexion in swing phase and of its excursion. No changes of the ankle position were evident. As for ankle kinetics, we observed in the second session higher values for the peak of ankle power in terminal stance in comparison to the first session. No changes were found in terms of ankle kinetics. The findings demonstrated improvements associated to long-term weight loss, especially in terms of spatial–temporal parameters and at hip level. Our results back the call for early weight loss interventions during childhood, which would allow the development of motor patterns under normal body weight conditions.

1. Background

Prader–Willi syndrome (PWS) is a rare condition, representing the most common form of genetic obesity. PWS affects multiple body systems and its most consistent characteristics include muscular hypotonia, hyperphagia, leading most subjects to develop morbid obesity from early childhood, dysmorphogenetic abnormalities, behavioral disorders and cognitive impairment, hypogonadism and growth failure (Cassidy, Schwartz, Miller, & Driscoll, 2012). General health status is usually preserved in individuals with PWS. Obesity is a common feature in PWS and it is often massive; many individuals with PWS exceed by more than 200% their ideal body weight. Motor problems are most prominent in infancy, but continue to be of clinical importance in adulthood, due to hypotonia, decreased muscle mass and the excessive amount of fat, which

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influences the biomechanics of activities of daily living, causing and increasing functional limitations over time (Capodaglio et al., 2009; Menegoni et al., 2009; Vismara et al., 2007). Within the wide spectrum of physical and intellectual disabilities associated with PWS, musculoskeletal diseases represent a serious issue, as they affect a significant proportion of individuals with PWS. Therefore, rehabilitation, physical activity interventions and surveillance play a key role in achieving the highest possible quality of life, especially in the light that PWS adults with lower body mass indexes (BMI) show a near-to-normal life expectancy.

Some studies have already focused on the quantification of motor performance in individuals with PWS, evidencing an abnormal gait pattern and postural ability and a delay in performance maturation when compared to normal weight subjects or BMI-matched non-genetically obese subjects (Butler et al., 2002; Capodaglio et al., 2009; Cimolin et al., 2010; Cimolin, Galli, Rigoldi, Grugni, Vismara, & Mainardi, 2011; Cimolin, Galli, Vismara, Grugni, Camerota, & Celletti, 2011; Cimolin, Galli, Vismara, Grugni, Priano, & Capodaglio, 2011; Galli et al., 2011; Kroonen, Herman, Pizzutillo, & MacEwen, 2006; Reusa et al., 2011; Vismara et al., 2007). The functional effects of weight loss after bariatric surgery (Aaboe, Bliddal, Messier, Alkjær, & Henriksen, 2011; Hortobagyi, Herring, Pories, Rider, & DeVita, 2011; Messier, Gutekunst, Davis, & DeVita, 2005; Vartiainen et al., 2012) or after short-term cyclical training (no later than 1 year) in obese subjects (Capodaglio et al., 2011; Reus, van Vlimmeren, Staal, Otten, & Nijhuis-van der Sanden, 2012; Vismara et al., 2010) have been also investigated. However, the effect of weight loss on gait is still controversial (Aaboe et al., 2011; Cimolin et al., 2013; Fontana et al., 2009; Messier et al., 2005; Vartiainen et al., 2012). We should bear in mind that in other genetically obese conditions, like in Down Syndrome, adults show precocious age-related changes in physiologic function and motor performance as compared to their counterparts: a decreased ability to perform activities of daily living and an earlier onset of age-related medical problems, such as osteoarthritis, hearing loss, and dementia, have been demonstrated in Down syndrome (Lott & Head, 2001). We can speculate that also in PWS patients, who share common features like muscular hypotonus, ligament laxity and obesity with Down syndrome, a similar decreased performance capacity may well be present. However, there is a lack of evidence in the literature and no longitudinal studies have been reported in PWS individuals. In particular, in this study we focused the attention on gait, as walking is a fundamental task for everyday life and the most common modality of prescribed physical activity. Eventual gait differences at long-term follow-up in PWS could be important for evaluating the effects of rehabilitative and weight loss programs on function. Therefore, our goal was to describe long term changes in gait biomechanics using 3D-gait analysis as well as in body mass in PWS subjects.

2. Materials and methods

2.1. Participants

Eight adult patients with a diagnosis of PWS were enrolled in this study (Table 1). All patients showed the typical PWS clinical phenotype (Cassidy et al., 2012). Cytogenetic analysis was performed in all participants. Seven patients had interstitial deletion of the proximal long arm of chromosome 15 (del15q11-q13), while uniparental maternal disomy for chromosome 15 (UPD15) was found in the remaining subject. Incomplete development of secondary sex characteristics was present in both genders. In males, testes were palpable with a volume of less than 6 ml. Primary amenorrhea was present in 3 subjects, while the remaining female suffered from irregular menses. Three females were undergoing sex steroid replacement treatment. Two hypertensive patients were on treatment with angiotensin-converting enzyme inhibitor plus Ca-antagonist and loop diuretics, respectively. One of the hypertensive patients had type 2 diabetes and was treated with insulin. No patient was undergoing weight-reducing medical therapy. Four patients were receiving treatments with neuroleptics. Six PWS were treated with growth hormone (GH). Mean duration of GH treatment was 9.7 years (range 8.1–10.1 years).

All PWS subjects showed mild mental retardation. In this respect, one of the requirements for participating in the study was a score over the cut-off value of 24 in the Mini Mental State Examination (MMSE) Italian version (Neri, Andermacher, Spanó, Salvioli, & Cipolli, 1992). Scores over the MMSE cut-off are recognized as suggesting the absence of widespread acquired cognitive disorders in adult people. In this light, our PWS patients were all able to understand and complete testing.

Table 1
Clinical characteristics of the study groups.

	PWS patients		HCG	OCG
	T0	T1		
Participants (M/F)	8 (4/4)		10 (5/5)	14 (5/9)
Age (years)	28.7 ± 4.6	36.4 ± 5.1	33.4 ± 9.6	29.4 ± 7.9
Weight (kg)	102.7 ± 21.7[+]	87.2 ± 24.5[*,+]	66.9 ± 8.5[§]	101.2 ± 12.9
BMI (Kg/m^2)	44.2 ± 6.4[+]	37.4 ± 5.9[*,+]	22.8 ± 3.2[§]	40.2 ± 3.3
[*]All values are mean ± SD				

[*] $p < 0.05$, PRE versus POST.
[+] $p < 0.05$, if compared to HCG.
[§] $p < 0.05$, if compared to OCG.