Relationship between flat foot condition and gait pattern alterations in children with Down syndrome

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Introduction

Down syndrome (DS) is the most common noninherited cause of mental impairment and occurs in 10 out of 10 000 live births (Lai *et al.* 2002; Weijerman & de Winter 2010) as a result of the presence of all or a portion of an extra copy of chromosome 21. There are a number of medical problems that are associated with the syndrome, including obesity, muscular hypotonia, ligament laxity and orthopaedic problems (Shumway-Cook & Woollacott 1985; Aruin *et al.* 1996), which lead to experiencing a wide range of other complications: scoliosis, joint dislocation, hip and knee cap instability, weak

Correspondence: Dr Veronica Cimolin, Bioengineering Department, Politecnico di Milano, p.zza Leonardo Da Vinci 32, 20133 Milano, Italy (e-mail: veronica.cimolin@polimi.it). ankles and feet problems. These abnormalities are responsible for postural and gait alterations widely documented by the literature (Mahan *et al.* 1983; Caselli *et al.* 1991; Caird *et al.* 2006; Concolino *et al.* 2006; Galli *et al.* 2008; Mik *et al.* 2008; Agiovlasitis *et al.* 2009; Cimolin *et al.* 2010, 2011; Weijerman & de Winter 2010; Steingass *et al.* 2011).

Among the feet problems, one of the most common abnormalities is flat foot which is present in 60% of the individuals with DS (Concolino *et al.* 2006; Pau *et al.* 2012). Flat foot is a condition in which the medial longitudinal arch of the foot collapses during weight bearing and restores after removal of body weight (Bordelon 1983). In DS the flat foot condition is generally due to hypotonia and ligamentous laxity, which are typical features of this syndrome.

In this perspective it is of particular clinical interest to acquire a deeper understanding of the alterations originated by DS in both structure and functionality of the foot, given the importance of this body part in maintaining upright stance, allowing gait to develop, carrying the weight of the body, shock absorbing and adjusting the body to uneven surfaces. The various problems associated with flat foot can interfere significantly with normal daily activities. Individuals with flat feet demonstrate several biomechanical inefficiencies in the foot and ankle, as well as a variety of gait abnormalities. A need for careful surveillance of foot development during childhood and adolescence has been recognised in DS, to help reduce the risk of mobility impairment in adulthood and minimise possible consequences originating from such issues (Mahan et al. 1983).

In patients with DS the assessment of walking abnormalities has mainly focused on biomechanical limitations related to the orthopaedic problems (Caselli *et al.* 1991; Roizen & Patterson 2003; Galli *et al.* 2008; Cimolin *et al.* 2010) without taking in consideration the role of flat foot. In literature only two studies experimentally investigated plantar pressure patterns of DS children and the role of flat foot mainly in maintaining posture. Concolino *et al.* (2006) found a significantly altered foot function, with consequences on postural stability, gait cycle and rearfoot–forefoot surface ratio values; recently, Pau *et al.* (2012) demonstrated that DS children exhibited larger midfoot and reduced forefoot contact areas with respect to healthy participants during upright quiet stance maintenance and increased average contact pressures in the midfoot and rearfoot.

From these considerations and from clinical need the aim of this study was to quantitatively assess the relationship between the flat foot and the gait alterations in DS children. Our hypothesis is that children with DS and flat foot exhibit non-physiological gait patterns whose alterations could be dependent on the severity of flat foot.

Materials and methods

Participants

Twenty-nine patients with DS were enrolled in this study (age: 9.8 ± 2.3 years; height: 131.8 \pm 12.9 cm; BMI = 12.2 \pm 6.7 kg/m²) for a total of evaluated 58 lower limbs. The distribution of chromosomal anomalies is pure trisomy 21 in all of the patients. The patients with DS were all admitted to the Rehabilitation Unit of the IRCCS 'San Raffaele Pisana', Tosinvest Sanità, Roma, Italy, for multidisciplinary rehabilitation. Inclusion criteria for the individuals with DS were low to medium intelligence quotient (IQ), no clinical sign of dementia, no previous surgery or other significant orthopaedic treatments. All subjects were able to understand and complete the test and to walk independently without the assistance or use of crutches, walkers or braces.

A group of 15 similarly aged subjects with typical development made up the control group (CG) (age: 9.2 ± 5.7 years; height: 130.3 ± 7.1 cm; weight: 33.5 ± 9.4 kg) and they were tested at the IRCCS 'San Raffaele Pisana', Tosinvest Sanità, Roma, Italy. Exclusion criteria for the control group included prior history of cardiovascular, neurological or musculoskeletal disorders. They showed normal flexibility and muscle strength and no obvious gait abnormalities. Approximately 20% of them exhibited flat foot, and such incidence was found in agreement with previous studies (Prasher et al. 1995; Bordin et al. 2001; Concolino et al. 2006; Chen et al. 2011). For healthy children the IQ was not measured, but it was considered in the normal range since all the subjects were recruited among school students. The study was approved by the Ethics

Committees of the Institute and written informed consent was obtained by the parents of the children recruited for the study.

Experimental set-up

Patients were assessed at the Movement Analysis Lab of the IRCCS 'San Raffaele Pisana', Tosinvest Sanità, Roma, Italy, using a 12-camera optoelectronic system (ELITE2002, BTS, Milan, Italy) with a sampling rate of 100 Hz, two force platforms (Kistler, CH) and two TV camera video system (BTS, Italy) synchronised with the system and the platforms for video recording. Plantar pressure measurements were obtained by means of a pressure-sensitive mat (Tekscan Inc., South Boston, MA, USA), composed of 2016 sensing elements arranged in a 42×48 matrix and connected via USB interface to a personal computer.

To identify foot type, the participants were placed on the mat with the help of an assistant who asked them to stand as still as possible for 5 s trials. A total of 40 temporal frames (sampled at 8 Hz) were acquired for each trial, and text matrices containing the foot–ground contact pressure value for each element of the sensitive grid were exported for further processing (Pau *et al.* 2012).

To evaluate the kinematics of each body segment, passive markers were positioned on the participants' body, as described by Davis et al. (1991). After the collection of some anthropometric measures (height, weight, tibial length, distance between the femoral condyles or diameter of the knee, distance between the malleoli or diameter of the ankle, distance between the anterior iliac spines and thickness of the pelvis), passive markers were placed at special points of reference, directly on the subject's skin, and in particular at C7, sacrum and bilaterally at the anterior superior iliac spine, greater trochanter, femoral epicondyle, femoral wand, tibial head, tibial wand, lateral malleolus, lateral aspect of the foot at the fifth metatarsal head and at the heel (only for static offset measurements). All acquisitions were acquired by the same operator to assure reproducibility of the acquisition technique and to avoid the introduction of errors due to different operators. After placement of the markers participants completed two or more practice trials across the plate walkway to ensure that the children were comfortable with the experimental procedure. After familiarisation, a trial was considered as valid when the following criteria were met: (1) a natural walk with self-preferred walk speed and (2) a whole foot is measured to the plate. Average values of three valid trials from each side foot were analysed. For each trial, the participant was measured with a right or a left foot strike on the plate. Six repeated trials (three for each foot) were conducted in order to guarantee reproducibility of the results (kinematics, kinetics and plantar pressures). All trials were undertaken while barefoot. Participants were asked to walk from a 'start line' to a 'finish line' at their normal or comfortable speed. The start line was approximately 3 m in front of the force platforms, and the stop line was 3 m behind the plates. The participants were allowed to rest if they feel tired between trials.

Data analysis

All graphs obtained from 3D Gait Analysis were normalised as % of gait cycle and kinetic data were normalised for individual body weight.

Using specific software (BTS EliteClinic, version 3.4.109) data were exported in .txt and .xls files. From these data format we identified and calculated some parameters related to ankle kinetics (ankle moment and power) (Table I; Fig. I). As the anatomic structure of one transverse, one medial longitudinal and one lateral longitudinal foot arch can perform the functions of buffering, amortising, stabilising and generating propulsion in human activities (Chan & Rudins 1994; Saltzman & Nawoczenski 1995), the attention was focused on ankle kinetics (ankle moment and power) which are directly related to propulsion ability during gait.

To identify foot types, we calculated the arch index (AI) from the plantar pressure according to Cavanagh & Rodgers (1987). First we divided the foot–ground contact area into three regions, i.e. forefoot, midfoot and rearfoot. Three relative contact areas were estimated and then used to calculate AI according to the following equation [eq. I]:

 $AI = \frac{Midfoot area}{Rearfoot area + Midfoot area + forefoot area}$

[eq. 1]

Gait parameter: ankle kinetics	Description
AMMax (N·m/kg)	The maximum value of ankle plantarflexion moment during terminal stance
APMax (W/kg)	The maximum value of generated ankle power during terminal stance (maximum value of positive ankle power during terminal stance) (APMax index), representing the push-off ability of the foot during walking
APmin (W/kg)	Minimum value of absorbed ankle power in early stance and midstance, when muscle is contracting eccentrically and absorbing energy (minimum value of negative ankle power)
AI (J/kg)	Absorbed work in early and midstance (total negative work of ankle power plot) according to the formula $A_{l} = \int_{\pi}^{T_{2}} P dt (Fig. 1)$
A2 (J/kg)	Generated work at push-off during terminal stance (positive work of ankle power plot) according to the
	formula $A_2 = \int_{T_3}^{T_4} P dt$ (Fig. 1)

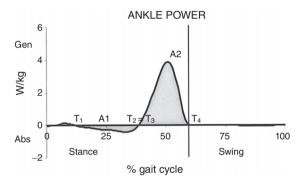


Figure I Ankle power pattern for CG showing AI and A2 indices and TI, T2, T3 and T4 (CG, control group; AI, absorbed work in early and midstance; A2, generated work at push-off during terminal stance; TI and T2, frames in which the ankle joint power pattern goes through the zero in the first phase of stance; T3 and T4, frames in which the ankle joint power pattern goes through the zero at push-off).

To derive these three areas, a foot axis line is drawn from the middle of metarsals 2 and 3 to the middle of the heel. Perpendicular to this foot axis, the foot excluding the toes is divided into three equal parts. Thus, AI was essentially a ratio of midfoot area to total foot contact area without the toes (Cavanagh & Rodgers 1987). Based on the AI, plantar arches were classified as follows: AI < 0.21, high arch; 0.22 < AI < 0.26, normal arch; AI \ge 0.26, low arch (Cavanagh & Rodgers 1987). This procedure was performed by the same operator to ensure data reproducibility; the whole process was carried out by means of a custom Matlab routine that processes the matrices exported by the Tekscan system.

Statistical analysis

According to the classification of plantar arches described above (Cavanagh & Rodgers 1987) the DS group was divided into two subgroups: the group with 'high/normal arch' (AI < 0.26; 17 lower limbs) and the group with 'low arch' (AI > 0.26; 41 lower limbs).

All the previously defined parameters were computed for each participant and then the mean values and standard deviations of all indexes were calculated for each subgroup and for CG. Kolmogorov– Smirnov tests were used to verify if the parameters were normally distributed; the parameters were not normally distributed; the parameters were not normally distributed, so we used non-parametric analysis. Data of the two subgroups and CG were compared using Kruskal–Wallis followed by *post hoc* comparison, in order to detect significant differences. Spearman correlation was calculated between the AI and parameters obtained by the Gait Analysis in the two pathological subgroups. Null hypotheses were rejected when probabilities were below 0.05.

Results

According to the classification of the plantar arches, in Table 2, the clinical characteristics of the two subgroups are reported, showing that no statistical differences were found for age, height and BMI, with the exception of AI.

The research for differences between the two subgroups as for the Gait Analysis parameters (Table 3) showed statistical differences only for the peak of ankle plantarflexion moment (AMMax index) and the maximum ankle power during terminal stance (APMax index). The values of both parameters were significantly lower for the subgroup with low arch; no other differences were found between the two subgroups, which displayed all the analysed parameters different from CG from a statistical point of view (P < 0.05).

The correlations between the AI values and the other measures are presented in Table 4 for the two subgroups. We found that while in the group with high/normal arch no significant correlations were present, in the group with low arch the AI index displayed a moderate correlation (r varies between 0.31 and 0.48; P < 0.05) with all kinetic parameters with the exception of AI parameter. Our data showed that in presence of low arch, the higher the AI value, the lower the maximum of ankle moment (AMMax index) and of the generated ankle power

	Group with high/normal arch	Group with low arch
# limbs (%)	17/58 (29%)	41/58 (71%)
AI	0.19 (0.04)*	0.34 (0.04)
Age (years)	9.03 (3.26)	9.96 (1.48)
Height (cm)	132.22 (11.99)	130.81 (13.48)
BMI (kg/m ²)	12.29 (7.16)	12.08 (6.69)

Data are expressed as mean (standard deviation).

* P < 0.05, group with high/normal arch vs. group with low arch. AI, arch index.

(APMax index) (Fig. 2) during terminal stance and the minimum of absorbed ankle power (APmin index).

Discussion

The purpose of the present study was to explore the relationships between the AI and parameters obtained from 3D Gait Analysis in children with DS. Measurements of AI have been used to infer dynamic foot function and guide clinical intervention (Jonely et al. 2011), but its relationships with walking parameters are not well understood, especially in syndrome like DS.

To our knowledge no experimental assessment on relationship between biomechanical abnormalities of ankle and foot-ground interactions (i.e. analysis of contact area and pressure distribution) is present in

Table 4 Correlation between arch index values and the gait parameters for the two subgroups

Gait Analysis parameters	Group with high/normal arch	Group with low arch
AMMax (N·m/kg)	0.14	-0.31*
APMax (W/kg)	-0.27	-0.48*
APmin (W/kg)	-0.04	0.31*
AI (J/kg)	-0.33	0.13
A2 (J/kg)	-0.28	-0.42*

* *P* < 0.05.

AMMax, ankle moment maximum; APMax, ankle power maximum; APmin, ankle power minimum; A1, absorbed work; A2, generated work.

Gait Analysis	Group with	Group with	
parameters	high/normal arch	low arch	CG
AMMax (N·m/kg)	0.99 (0.17)* [†]	0.86 (0.27) [†]	1.29 (0.23)
APMax (W/kg)	1.60 (0.65) ^{*†}	1.20 (0.52) [†]	3.06 (1.16)
APmin (W/kg)	-0.49 (0.19) [†]	-0.44 (0.23) [†]	-0.22 (0.19)
AI (J/kg)	-7.79 (3.31) [†]	-7.71 (4.02) [†]	-3.89 (0.27)
A2 (J/kg)	15.64 (6.76) [†]	12.38 (6.23)†	37.08 (0.45)

Table 3 Comparison of selected ankle kinetic parameters [mean (SD)] in the two analysed groups

* P < 0.05, group with high/normal arch vs. group with low arch; [†] P < 0.05, if compared to CG.

CG, control group; AMMax, ankle moment maximum; APMax, ankle power maximum; APmin, ankle power minimum; A1, absorbed work; A2, generated work.

Table 2 Subject characteristics of the two subgroups

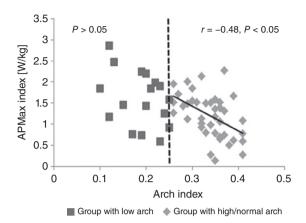


Figure 2 Plot representative of the correlation between maximum ankle power (APMax index) and arch index (AI), expressed in terms of the Spearman correlation for the group with high/normal arch (AI < 0.26) and the group with low arch (AI \ge 0.26, r = -0.48, P < 0.05).

pathological conditions. The studies on this topic have been conducted on healthy individuals during walking (Lin *et al.* 2001; Levinger *et al.* 2010; Fan *et al.* 2011; Shih *et al.* 2012). Lin *et al.* (2001) and Shih *et al.* (2012) demonstrated that movement patterns at main lower limb joints during gait were in general similar between children with and without flat foot. Levinger *et al.* (2010) observed that flatarched feet altered motion associated with greater pronation during gait in adult individuals. Fan *et al.* (2011) compared the non-pathological flat foot vs. high-arched foot of adults during gait; they found that there was a significant difference in the distributions of vertical force and in the rate of change of footprint area.

In the present study, the relationship between the degree of the flat foot, represented by the AI, and ankle kinetics during gait in DS children has been examined. A flat foot condition is commonly explained by hypotonia and ligamentous laxity, which are typically observed in individuals with DS, and which are likely to cause a collapse of the medial longitudinal arch. These conditions could be amplified by large BMI which is common in DS children (Weijerman & de Winter 2010). Obese individuals, in fact, are at an increased risk of developing foot discomfort and/or foot pathologies due to increased plantar loads. Furthermore, continual

bearing of excessive mass by children appears to flatten the medial midfoot region during walking (Dowling *et al.* 2004).

First, significant differences were found between children with high/normal arch and those with low arch, which showed a less functional gait pattern in terms of ankle kinetics. In particular in DS children with flat foot the AI resulted to be significantly related to ankle moment and power generation at push-off during walking. It may suggest that the presence of flat foot, together with other factor like obesity, is more likely to cause pain and discomfort in the lower extremities with weaker efficient walking.

The research for correlations displayed significant correlation results in the subgroup with low arch which suggest that there may be meaningful relationships between AI and some parameters related to ankle moment and ankle power generation during terminal stance. Interpretation of these outcomes suggests that increasing flat foot tended to result in lower push-off ability, leading to a less functional walking. According to a previous study (Shih *et al.* 2012) which demonstrated no adaptation during gait in healthy children with flat foot, we can suppose that the anomalies in gait pattern related to the flat foot and its degree obtained in the present study in DS children may be directly connected to the syndrome.

It is demonstrated that the structural difference in the types of foot causes significant difference of ground reaction forces distribution of foot in healthy adults (Fan et al. 2011). The differences in structure and in the forces distribution have an effect on foot muscle tension while walking. This offers important evidence to analyse foot muscle fatigue. The force distribution of foot can well explain why the flat-footed experience pain more readily when they walk for a long time. The smaller rate of the footprint areas brings greater stability to the high-arched. The lack of stability suffered by the flat-footed requires more consumption of energy, and thus may well explain the fatigue felt by the flat-footed on long walks. The foot arch can not only lessen muscle fatigue, it can also reduce energy consumption (Collins & Kuo 2010). These features are exacerbated in individuals with DS, which present not only flat foot, but also other orthopaedic disorders and obesity.

It is not possible to directly compare our results with literature as, to our knowledge, these are the first biomechanical data documenting the effect of flat foot on propulsion capacity in DS children. However, our outcomes may suggest information about the patterns of plantar pressure loading in feet of children with DS that could be clinically meaningful in the diagnosis and management of their feet pathologies.

From a clinical point of view, these results could develop and enhance the rehabilitative options in DS. As one of the primary causes of flat foot in DS is the presence of hypotonia and ligamentous laxity, it appears important to plan, starting from the early stages of childhood, a specific rehabilitative programme designed to avoid the effect of hypotonia, improving muscle strength of the foot muscles and motor control during gait, too. In this way it could be possible to counteract the documented evolution of midfoot contact area and the flat foot condition which is common in DS. In addition they should be encouraged to walk for its positive impact on muscle mass and strength and energy balance, to optimise gait pattern and prevent the onset of compensatory strategies. Evidence-based rehabilitation programmes would contribute to improve daily functioning, quality of life and weight management issues including also a diet programme. In addition, data obtained in this study could represent an important aid in the definition of therapeutic intervention, including the design optimisation of orthotic devices, especially in the most serious situations of flat foot.

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