The effects of low arched feet on foot rotation during gait in children with Down syndrome

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Introduction

Down syndrome (DS) is a chromosomal disorder caused by trisomy of chromosome 21 (Hsa21) associated with a number of signs and symptoms including learning disabilities, heart defects, craniofacial dysmorphia and childhood leukaemia (Wiseman *et al.* 2009). Physical activity patterns of DS are influenced by obesity, ligaments' laxity and reduced muscle strength and tone (American Academy of Pediatrics, Committee on Genetics 2001). These features may contribute to the reduced motor skills observed in this population (Barnhart & Connolly 2007; Bhaumik *et al.* 2008) and are responsible for postural and gait alterations

widely documented by the literature (Caird *et al.* 2006; Galli *et al.* 2008; Mik *et al.* 2008; Agiovlasitis *et al.* 2009; Cimolin *et al.* 2010, 2011; Weijerman & de Winter 2010; Rigoldi *et al.* 2011; Steingass *et al.* 2011). These motor disorders tend to progressively worsen as the clinical picture advances, severely limiting the individuals 'quality of life. Among the wide spectrum of orthopaedic issues encountered in individuals with DS, one of the most common abnormalities is flat foot which is present in 60% of the children with DS (Concolino *et al.* 2006; Pau *et al.* 2012). In DS this condition is generally due to hypotonia and ligamentous laxity, which are typical features of this syndrome.

In previous studies, the assessment of walking abnormalities gait analysis has mainly focused on DS with special reference to their specific associated orthopaedic conditions and biomechanical limitations (Roizen & Patterson 2003; Galli *et al.* 2008; Cimolin *et al.* 2010), without taking in consideration the role of flat foot. Few studies investigated the effects of flat foot in children with DS and they were focused mainly on posture (Concolino *et al.* 2006; Pau *et al.* 2012). To our knowledge the only study assessing quantitatively the effect of flat foot on gait pattern in DS was conducted by Galli *et al.* (2013). The authors demonstrated that children with flat foot displayed a less functional gait pattern in terms of ankle kinetics than children without flat foot, suggesting that the presence of flat foot may lead to a weaker efficient walking. From the studies conducted using gait analysis, the individuals with DS are characterised by an external foot rotation during gait which may be considered a strategy for balance maintenance.

In a review on musculoskeletal issues linked to obesity, Hills *et al.* (2002) observed that in young obese subjects, the excess mass increases the foot-ground contact area and the peak pressures. This would predispose to the development of a pathological foot, as demonstrated by the greater incidence of flat foot in obese children (Must & Strauss 1999). Particularly, in children with DS, abnormalities in foot loading and hypotonia may be responsible for changes in the foot structure and can cause the collapse of the longitudinal arch and a decrease in foot functionality.

From a clinical perspective it is crucial 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, absorbing shocks and adjusting the body to uneven surfaces. As the problems associated with flat foot could interfere significantly with normal daily activities, it is important to investigate the foot development during childhood and adolescence in DS. These evaluations could reduce the risk of mobility impairment in adulthood and minimise possible consequences originating from such issues (Mahan *et al.* 1983). Although age and dementia have long been recognised as major predictors of mortality for individuals with DS, it has been demonstrated that other factors, such as mobility and functional skills also contribute to survival (Coppus *et al.* 2008).

From these considerations and from clinical need the aim of this study was to determine if DS children with flat foot are characterised by an accentuated external foot rotation during walking.

Materials and methods

Participants

Fifty-five children with DS were enrolled in this study [mean age: 9.6 years (SD: 1.7 years)] for a total of evaluated 110 lower limbs. All DS children were characterised by a pure trisomy 21 chromosome abnormality. Inclusion criteria were low to medium intelligence quotient (IQ), no clinical sign of dementia, no previous surgery or other significant orthopaedic treatments. All individuals 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 individuals with typical development were recruited for the control group (CG) [mean age: 9.2 years (SD: 5.7 years)]. 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. 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

All participants were assessed at the Movement Analysis Lab of the Research Institute 'San Raffaele Pisana', Tosinvest Sanità, Roma, Italy, using a 12-camera optoelectronic system (ELITE 2002, BTS, Milan, Italy) with a sampling rate of 100 Hz, two force platforms (Kistler, Winterthur, Switzerland) and two-TV camera Video system (BTS) synchronised with the system and the platforms for videorecording. 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 characterise foot morphology, 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).

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, as described by Davis (Davis *et al.* 1991) to evaluate the kinematics of each body segment. In particular they were placed at C7, sacrum and bilaterally at the ASIS, 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). The Davis marker-set was chosen as the protocol of choice to acquire the movement of lower limbs and trunk based on Ferrari *et al.* (2008). 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, at least six trials were acquired asking the participants to walk at their self-selected velocity and barefoot. Average values of three consistent trials from each side foot were analysed.

Data analysis

All graphs obtained from gait analysis were normalised as % of gait cycle; in the present analysis only kinematic data were considered and in particular the attention was focused on the foot rotation graph, representative of the foot position on the transverse plane (internal–external rotation) during walking (Fig. 1).

From foot rotation data we identified and calculated the following parameters:

- FR IC (Foot Rotation at Initial Contact): the value of the foot position in the transverse plane at initial contact;
- Mean FR St (Mean Foot Rotation in Stance): the mean value of the foot position in the transverse plane during the stance phase; and
- Mean FR Sw (Mean Foot Rotation in Swing): the mean value of the foot position in the transverse plane during the swing phase.

To characterise the foot arch type, the computation of the arch index (AI) from the plantar pressure was conducted according to Cavanagh & Rodgers (1987). First, the foot-ground contact area (toes excluded) was divided into three regions, that

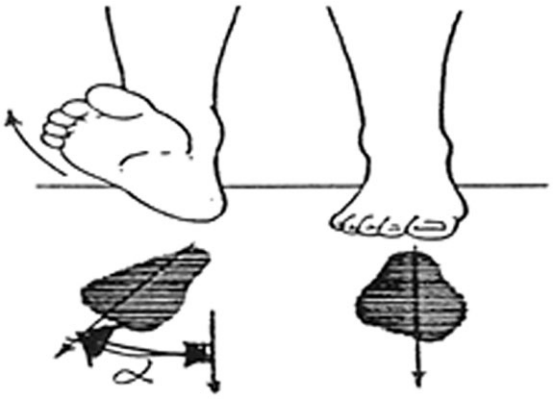

Figure 1 The foot rotation angle is the angle between the segment representative of the foot and the progression line of walking, projected on the transverse plane of the laboratory.

	DS (high/normal arch)	DS (low arch)	CG
No. of limbs (%)	13 (12%)	97 (88%)	30
AI	0.23 (0.01)	0.34 (0.04)*+	0.23 (0.01)
Age (years)	9.27 (1.85)	9.63 (1.72)	9.20 (5.70)
Height (m)	1.26 (0.14)+	1.27 (0.11)+	1.34 (0.15)
BMI (kg/m^2)	18.71 (2.62)+	20.36 (3.67)+	15.29 (3.71)

Table 1 Characteristics of the Down syndrome (DS) and control (CG) children

* $P < 0.05$, group with high/normal arch versus group with low arch; + $P < 0.05$, if compared with CG.
Data are expressed as mean (standard deviation).
AI, arch index; BMI, body mass index.

is, forefoot, midfoot and rearfoot. Three relative contact areas were estimated and then used to calculate AI according to the following equation (equation 1):

$$\mathrm{AI} = \frac{\text{Midfoot Area}}{\text{Rearfoot Area} + \text{Midfoot Area} + \text{Forefoot Area}} \quad (1)$$

To derive these three areas, a foot axis line is drawn from the middle of metatarsal 2 and 3 to the middle of the heel. Perpendicular to this foot axis, the foot excluding the toes is divided in three equal parts. Thus, AI was essentially a ratio of mid-foot 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 ≥ 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 pressure values matrices exported by the Tekscan system as text files.

Statistical analysis

According to the classification of plantar arches, the DS participants were divided into two sub-groups: a group with 'high/normal arch' (AI < 0.26; 13 lower limbs) and a group with 'low arch' (AI ≥ 0.26; 97 lower limbs).

All the parameters related to the foot rotation graph were computed for each participant (both for DS and control individuals) and the mean values and standard deviations of all indexes were calculated for each DS sub-group and for CG. According to Kolmogorov–Smirnov tests the parameters were not normally distributed so non-parametric analysis was used. Data of the two sub-groups and CG were compared using Kruskal–Wallis followed by *post-hoc* comparison, in order to detect significant differences. Null hypotheses were rejected when probabilities were below 0.05.

Results

The clinical characteristics of the two DS sub-groups and CG are listed in Table 1, showing that no statistical differences were found for age, height and body mass index (BMI: kg/m^2), with the exception of the AI. Both DS sub-groups were statistically different from CG for height and BMI, while only the DS sub-group with low arch showed higher AI value respect to the control children.

The comparison of foot rotation parameters (Table 2) revealed a significant group difference for mean foot position in the transverse plane during the stance (Mean FR St index) and swing phase (Mean FR Sw index). While the DS sub-group with high/normal arch had indices similar to the CG, the DS sub-group with low arch had indices values significantly higher than the other two groups. At initial contact (FR IC index) no significant difference was found between the two DS sub-groups, but the DS group with low arch was characterised by higher value respect to the CG, exhibiting a more extra-rotated foot at initial contact (Fig. 2). Plots of foot rotation angle during the gait cycle for three representative individuals (Fig. 2) illustrate the

Foot rotation parameters (°)	DS (high/normal arch)	DS (low arch)	CG
FR IC (°)	−14.57 (10.01)	−19.18 (10.14)$^{+}$	−11.23 (4.67)
Mean FR St (°)	−15.05 (9.99)	−20.73 (10.48)*$^{+}$	−13.51 (3.64)
Mean FR Sw (°)	−12.52 (8.02)	−21.85 (11.53)*$^{+}$	−16.12 (4.23)

Table 2 Comparison of selected foot rotation parameters between the two Down syndrome (DS) sub-groups and control group (CG)

* Significant difference ($P < 0.05$) between DS (low arch) and DS (high/normal arch).
$^{+}$ Significant difference ($P < 0.05$) between DS (low arch) and CG.
Values represent group means and standard deviation.
FR, foot rotation; IC, initial contact; St, stance phase; Sw, swing phase; CG, control group.

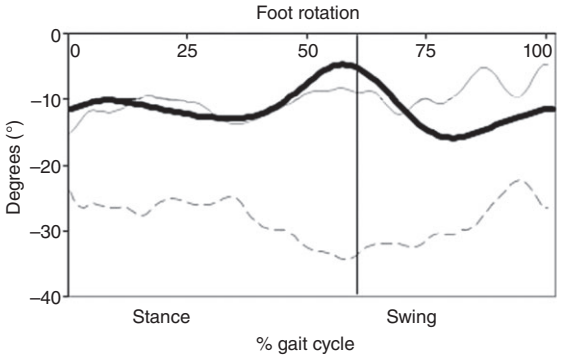

Figure 2 Foot rotation plot representative of a child with high/normal arch (solid line), a child with low arch (dashed line) and a control group child (thick line). The vertical line is representative of toe-off and stance and swing phases are represented.

observed difference in foot rotation for DS children with low arch compared with DS children with high/normal arch and the CG.

Discussion

The aim of this study was to assess if the presence of flat foot in children with DS could influence the foot position during walking, and in particular the foot rotation, that is, internal-external rotation of the foot on the transverse plane, using data obtained from three-dimensional (3D) gait analysis.

In literature, the majority of studies related to the effects of flat foot on gait patterns have been conducted on healthy individuals. Several investigations have compared subjects with flat foot to those with normal foot posture (Williams *et al.* 2001; Hunt & Smith 2004; Tweed *et al.* 2008; Cobb *et al.* 2009; Twomey & McIntosh 2012), but the results of these studies have been inconsistent because of the variations in foot posture classification and the biomechanical modelling methods used (Levinger *et al.* 2010). To our knowledge, only two studies were conducted in children (Shih *et al.* 2012; Twomey & McIntosh 2012); the authors of these studies concluded that while there was a suggestion of restraint of motion rather than an excessive motion, differences were small between the symptomatic pes planus and normal subjects across parameters related to ankle and foot position during gait.

In our evaluation, significant differences were found between DS children with high/normal arch and those with low arch in terms of foot rotation. While DS participants with high/normal arch globally displayed a foot position in the transverse plane close to CG during the whole gait cycle, the DS group with low arch was characterised by higher extra-rotation of the foot in comparison with the DS group with high/normal arch and CG.

Our results suggest that the presence of flatfoot lead the children to extra-rotate their feet more than the children without flat foot. In addition, the abnormal foot position in the transverse plane, together with other factors like ligament laxity and hypotonia, may have a direct influence on ankle kinetic ability, which has been previously demonstrated to be reduced in DS children with flat foot (Galli *et al.* 2013).

A flatfoot condition is commonly explained by hypotonia and ligamentous laxity, which are typically observed in DS children, and which is likely to cause a collapse of the medial longitudinal arch. Such problems might be exacerbated by a mass

excess that is another issue often encountered in children with DS (Weijerman & de Winter 2010), and indeed a significantly larger BMI was found in our results in the DS group. However it was found that obesity did not significantly modify the AI in young individuals with DS, thus indicating that, contrary to what is observed in the case of healthy individuals, the excess of mass makes no further contribution to medial arch flattening. This is because of a saturation effect caused by the joint and ligament laxity associated with DS. In addition it was found that children with DS exhibited very high AI values even in normal-weight conditions. Thus, the foot arch is already so flattened that even further loads are unable to modify its configuration (Pau *et al*. 2013). Moreover, as the growth curves of the children with DS and of the developing children are not consistent, it would be misleading to make a direct comparison between them, for example by using the typical cut-off points proposed by Cole *et al*. (2000) to define the overweight or obese condition. We can suppose that the anomalies in foot position related to the flat foot obtained in the present study in DS children may be directly connected to the syndrome and to its features, because no adaptation during gait in developing children with flat foot was observed (Shih *et al*. 2012).

From a clinical point of view, these results could enhance the rehabilitative programmes in DS. As one of the primary causes of flatfoot 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, these subjects should be encouraged to walk for its positive impact on muscle mass and strength and energy balance, to optimise gait patterns and prevent the onset of compensatory strategies. Our results could be used also in the design optimisation of orthotic devices or foot insoles, especially in the most severe flat foot. For example, for the most serious cases the use of proper foot insoles could not only correct the excessive external foot rotation in children with flat foot but also globally improve their gait pattern.

This study has some limitations. First, the degree of muscular hypotonia, weakness and ligament laxity has not been measured nor compared between sub-groups, thus hindering interpretation of the findings. Another weakness related to the choice of control group could be that the developing individuals were of the same chronological age and not in terms of mental age (IQ). However, it was found that when the comparison group is matched on mental age, there are no significant differences in the overall motor performance (Croce *et al*. 1996; Angelopolou *et al*. 1999). However, the main purpose of our investigation was to compare DS children with and without flat foot, in order to identify possible differences that could be useful to differentiate and enhance the rehabilitative programmes of these subjects.

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