Available online at www.sciencedirect.com
ScienceDirect
Gait & Posture 28 (2008) 502–506
GAIT POSTURE
www.elsevier.com/locate/gaitpost

Joint stiffness and gait pattern evaluation in children with Down syndrome

Manuela Galli [a,d], Chiara Rigoldi [a,*], Reinald Brunner [b], Naznin Virji-Babul [c], Albertini Giorgio [d]

[a] *Bioeng. Dept., Politecnico di Milano, P.zza Leonardo da Vinci 32, 20133 Milano, Italy*
[b] *Down Syndrome Research Foundation, Vancouver, BC, Canada*
[c] *Children's University Hospital Basel, Basel, Switzerland*
[d] *San Raffaele Cassino, Tosinvest Sanità, Rome, Italy*

Received 20 February 2007; received in revised form 4 March 2008; accepted 5 March 2008

Abstract

Hypotonia, ligament laxity and motor alterations are characteristic for patients with Down syndrome (DS). The purpose of this study was the evaluation of typical gait pattern of subjects with Down syndrome and the quantification of their joint stiffness, connected with ligament laxity and hypotonia, as a possible compensation.

98 children with DS (mean age: 11.7 years; range: 6–15 years) and 30 healthy children (control group (CG); mean age: 11 years; range: 5–13 years) underwent full 3D gait analysis at self-selected speed.

Subjects with DS walked with more hip flexion during the whole gait cycle, knee flexion in stance phase, a limitation of the knee range of motion, and plantarflexion of the ankle at initial contact. Ankle power was limited as evident in terminal stance and pre-swing, represented by a low propulsive capacity at push-off, too. Hip joint stiffness was increased in general in patients with DS versus normal subjects while ankle joint stiffness revealed a lower value instead.

Keywords: Gait analysis; Down syndrome; Gait pattern; Joint stiffness

1. Introduction

Down syndrome (DS) is the most common non-inherited cause of mental impairment and occurs in 1 out of 1000 live births [1] 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 cardiac and respiratory conditions. Motor disability is widespread among individuals with DS. It includes longer motion and reaction times, balance and postural deficits, and cocontraction of agonist and antagonist muscles [2,3]. These deficits may have a causal link to delays in achieving motor development milestones in children. The motor dysfunction in individuals with DS involves impaired muscle control, which is frequently referred to as ''clumsiness'' by parents and health professionals [4]. The neuropathological basis for motor dysfunction in DS is unknown, but cerebellar dysfunction, delayed myelination, as well as proprioceptive and vestibular deficits have been suggested as possible causes [6,7]. The delay in motor development in DS is linked to the generalized muscle hypotonia and ligament laxity that is characteristic of the condition [5].

Early physiotherapy focuses on facilitating motor control and coordination in order to achieve developmental milestones. Once walking is established (which is often delayed by an average of 12–18 months) [8,9] regular physiotherapy is usually discontinued. There are, however, numerous reports in the literature suggesting that children with DS begin to develop orthopedic problems early in childhood and would benefit from specific biomechanical assessment and

* Corresponding author. Tel.: +39 02 2399 3359.
E-mail address: chiara.rigoldi@polimi.it (C. Rigoldi).

0966-6362/$ – see front matter © 2008 Elsevier B.V. All rights reserved.
doi:10.1016/j.gaitpost.2008.03.001

management. Caselli et al. [10] reported that walking in children and adolescents with DS was characterized by a ''Chaplinesque'' pattern with external rotation of the hips, increased knee flexion and valgus, and external rotation of the tibia. In childhood, pes planovalgus with marked pronation of the foot was observed, which could impact on postural stability and ambulation.

Foot deformity and resulting impeded function has been described as lever arm dysfunction in patients with neuromuscular disorders [20]. In adolescents and adults with DS, hallux valgus, hammer toe deformities, plantar fasciitis, and early onset of foot arthritis associated with severe flat feet were also observed, which impair ambulation and cause further dysfunction [21].

Parker and Bronks [11] studied the gait pattern of six young children (mean age: 4.2 years) with DS using video analysis. Poor heel–toe rocking during the stance phase and exaggerated abduction of the lower limb to facilitate foot clearance were observed. The gait pattern of 63 children with DS showed prolonged hip flexion during the gait cycle, an increase of knee flexion in the sagittal plane at initial contact and significant changes in ankle movement during the gait cycle [12]. These findings fit with the lever arm dysfunction in other disorders where inadequate plantarflexion results in crouch. The gait is further characterized by a significant decrease in plantarflexor moments and of absorbed and generated ankle power [12]. These abnormalities may reflect muscle hypotonia, ligament laxity, weakness of the plantarflexors and dysfunction of the foot as a lever arm.

While the specific orthopaedic and biomechanical limitations have been clearly identified, little is known about the nature of the relationship between muscle hypotonia, ligament laxity and the resulting joint stiffness in children with DS. The purpose of this study was to document the gait characteristics of children with DS and to quantify the hip and ankle joint stiffness that characterize gait in individuals with DS.

2. Patients

Ninety-eight children with DS (mean age: 11.7 years; range: 6–15 years) and 30 healthy ones (control group: CG; mean age: 11 years; range: 5–13 years) participated in this study. All patients were independent ambulators. The characteristics of the subjects are listed in Table 1.

The parents of all children provided informed consent to participate in the study and this study was approved by the Ethical Committee of the hospital IRCCS ''San Raffaele-Pisana'', Rome, Italy.

Table 1
Characteristics (mean ± S.D.) of analyzed subjects

Subjects	Height (cm)	Weight (kg)
CG	132.44 ± 10.92	29.81 ± 6.10
DS	141.69 ± 12.96	47.65 ± 14.13

3. Methods

Three-dimensional kinematic data were obtained using a 12-camera optoelectronic system with passive markers (ELITE 2002, BTS, Milan, Italy [13], sampling rate of 100 Hz). Two force platforms (Kistler, Winterthur, CH), embedded in the walkway were used to obtain kinetics. All trials were videotaped using a video system, synchronized with the optoelectronic system and force platforms (Videocontroller, BTS, Milan, Italy). Seventeen passive markers were placed according to Davis [15].

All subjects were asked to walk barefoot at their self-selected speed along a 10 m walkway. Six trials were collected for each subject. Kinematic and kinetic data were computed using Euler angles and Euler's equations of motion, respectively [15]. The kinematic and kinetic data of the hip, knee and ankle joints in the sagittal plane were studied, as they represent push-off capacity. All the graphs were normalized for percentage of gait cycle.

Temporal spatial parameters were compared between the two groups. Hip, knee, ankle joint kinematics and kinetics (range of motion, maximum/minimum of flexion extension angles values during gait cycle instants, hip, knee and ankle flexion extension joint moments, generated ankle power) were analyzed.

In order to evaluate the effect of ligament laxity and hypotonia on joint kinetics and kinematics, hip and ankle joint stiffness (hip joint stiffness: K_h; ankle joint stiffness: K_a) were expressed by plotting the values of flexion–extension moment versus flexion–extension angle over the gait cycle interval (Fig. 1a and b). The interval between the 10% and 30% (corresponding to the second rocker) of gait cycle was selected and the linear regression was fitted (Fig. 1c and d); the angular coefficient of linear regression corresponded to the joint stiffness index as described in previous studies [16,17]. Knee stiffness was not included in this study because of the lack of linear relation between kinematics and kinetics. As the weight differed significantly between the two groups (BMI: DS, 27.44 ± 3.8 kg/m^2; CG, 21.5 ± 1.49 kg/m^2) (Table 1), the kinetic data were normalized for weight. The individual mean and standard deviation of the parameters of interest were calculated before the mean and standard deviation of the groups.

Kinematic and kinetic parameters were compared using the Student's *t*-test (parametric data) or the Wilcoxon test (non-parametric data). Statistical significance was set at $p < 0.05$.

4. Results

4.1. Gait evaluation

Subjects with DS showed a significant decrease in gait speed (0.42 ± 0.08 s-1; $p < 0.05$) and stride length (0.29 ± 0.04; $p < 0.05$) in comparison with the control group (gait speed: 0.85 ± 0.06 s-1; stride length: 0.89 ± 0.09).

Sagittal hip kinematics (Fig. 2a) showed more hip flexion in DS patients (initial contact: DS, $37.0 \pm 8.1^\circ$; CG,

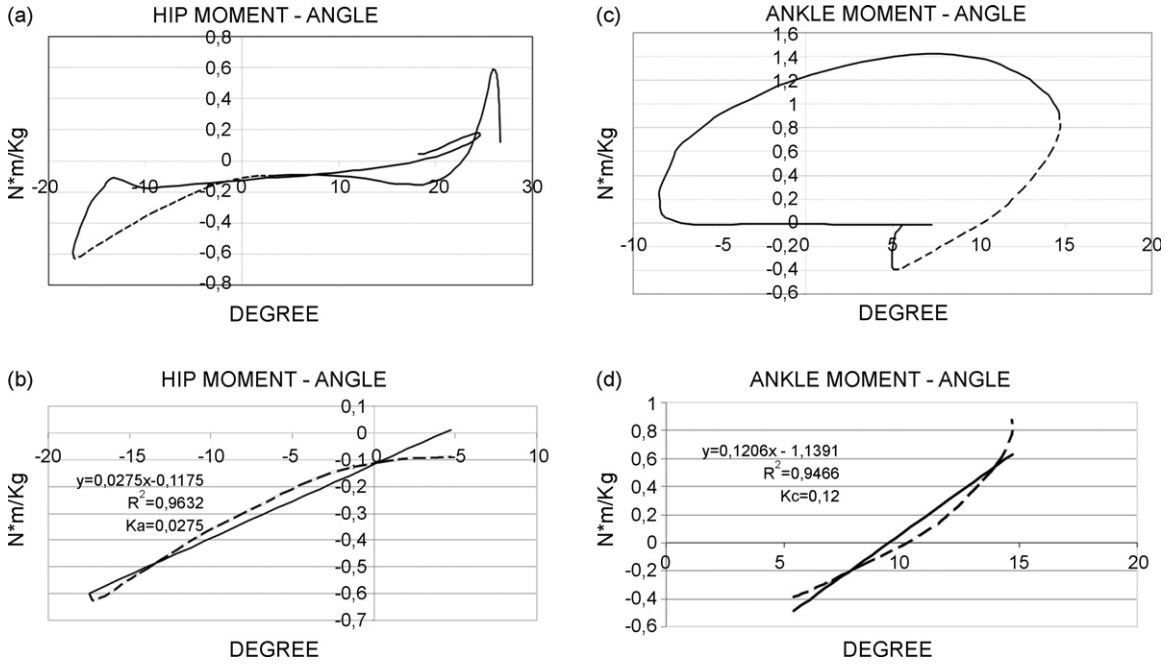

Fig. 1. (a) The hip angle–moment plot during the gait cycle for a healthy subject. (b) The ankle angle–moment plot during the gait cycle for a healthy subject. (c) The hip angle–moment plot cycle during second rocker (dashed line) for a healthy subject. (d) The ankle angle–moment plot cycle during second rocker (dashed line) for a healthy subject. The slope of the joint moment plotted as a function of joint angle during second rocker represents ankle joint stiffness.

$29.0 \pm 5.2^{\circ}$; $p < 0.05$; hip maximum extension in stance: DS, $11.5 \pm 9.7^{\circ}$; CG, $-6.6 \pm 6.4^{\circ}$; $p < 0.05$) and a reduced range of motion (DS: $27.4 \pm 7.1^{\circ}$; CG: $37.9 \pm 4.1^{\circ}$; $p < 0.05$).

Sagittal knee kinematics (Fig. 2b) showed an increased flexion at initial contact for the DS group (DS: $10.3 \pm 7.0^{\circ}$; CG: $6.3 \pm 4.7^{\circ}$; $p < 0.05$) an increased flexion at mid stance (DS: $14.7 \pm 9.3^{\circ}$; CG: $6.5 \pm 4.2^{\circ}$) and a reduction of the knee flexion (not statistical significant) in swing.

Sagittal ankle kinematics (Fig. 2c) showed a reduced first rocker and a reduced peak of ankle plantarflexion at toe-off for DS (DS: $-4.6 \pm 9.3^{\circ}$; CG: $-9.9 \pm 10.1^{\circ}$).

There is an increase in maximum hip flexor moment in hip kinetics (Fig. 3a), at initial contact for the DS group (DS: 0.7 ± 0.3 N m/kg; CG: 0.3 ± 0.4 N m/kg; $p < 0.05$) which is followed by a rapid decrease at approximately 15% of the gait cycle. During the greatest part of stance, however, DS patients had an increased extensor moment.

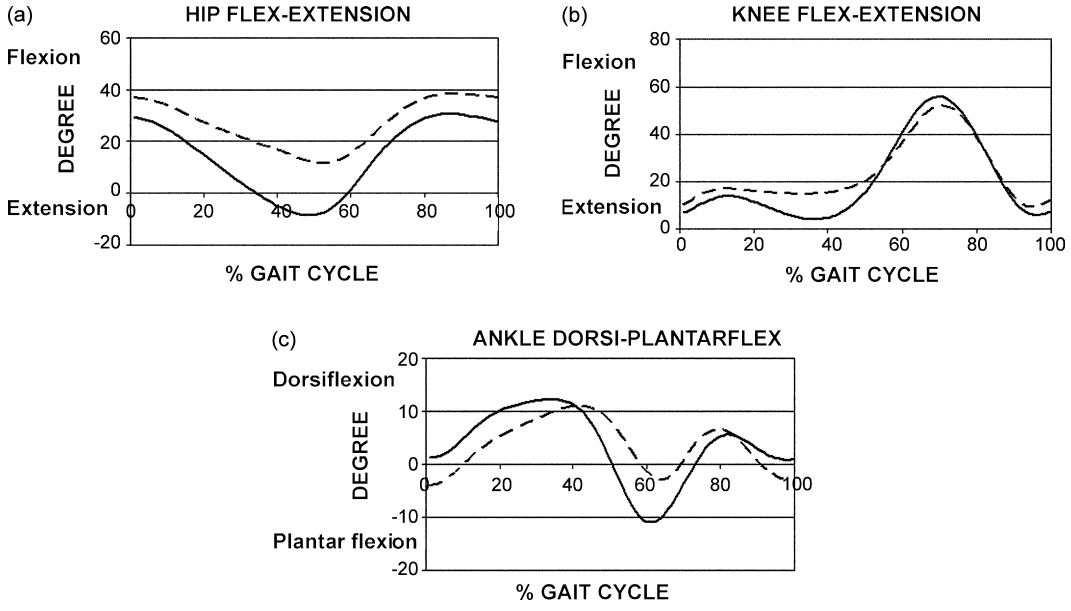

Fig. 2. Joint kinematics: plots of group mean data, DS (dashed line) and control group (solid line).

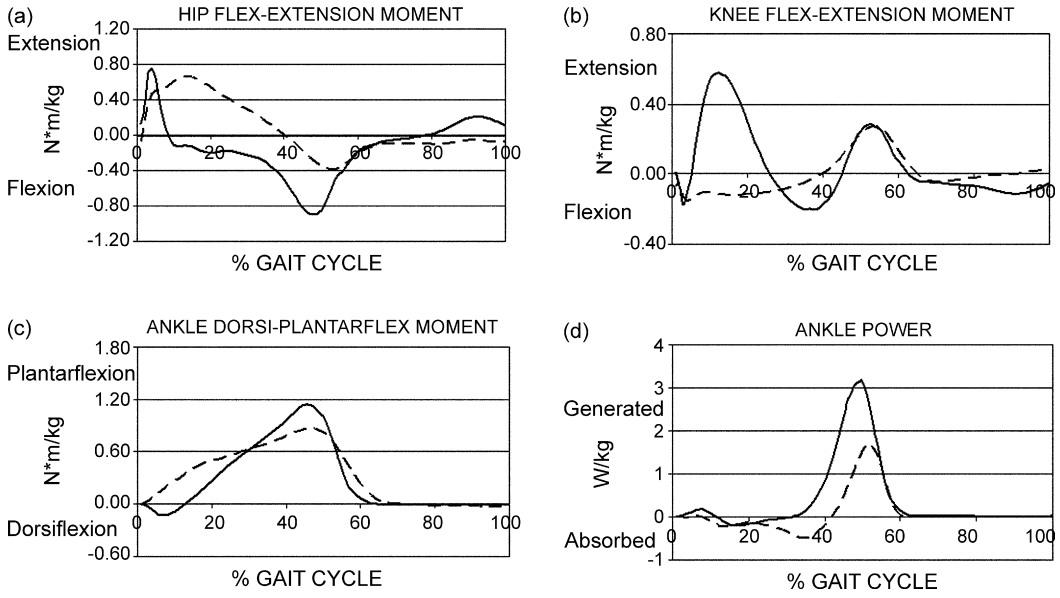

Fig. 3. Joint kinetics: averaged plots for the group with DS (dashed line) and control group (solid line).

Sagittal knee kinetics (Fig. 3b) revealed absence of the first extensor moment peak: in CG the first peak corresponded to 0.8 ± 0.2 N m/kg, while in DS, at the same percentage of gait cycle, an extensor moment was observed (-0.1 ± 0.2 N m/kg).

A short dorsiflexor peak for the ankle joint at the beginning of stance and a reduction of ankle moment maximum index (DS: 0.9 ± 0.3 N m/kg; CG: 1.3 ± 0.4 N m/kg; $p < 0.05$) for the DS group were seen in sagittal ankle kinematics (Fig. 3c). This was correlated with a reduction of power generating capacity at push-off (DS: 1.8 ± 0.3 W/kg; CG: 3.2 ± 0.8 W/kg; $p < 0.05$) (Fig. 3d).

4.2. Joint stiffness

K_h showed a statistically significant increase in the DS group (0.058 ± 0.025 (N m)/(kg degree)) in comparison with the CG (0.028 ± 0.007 (N m)/(kg degree), $p < 0.05$). In contrast, there was a statistically significant decrease in K_a for subjects with DS (0.058 ± 0.05 (N m)/(kg degree)) in comparison with the control group (0.103 ± 0.014 (N m)/(kg degree); $p < 0.05$). This finding was similar when data were normalized for gait speed.

5. Discussion

Patients with Down syndrome show ligament laxity, resulting from the connective tissue disorder, that characterizes the condition. Muscle hypotonia is another characteristic of these patients. The combination of these problems impedes dynamic joint stabilization and explains the increased incidence of musculo-skeletal deformities. Patients need to compensate for their muscle and ligament dysfunction in order to cope with daily activities and maintain function. Gait becomes unsteady, and the increased cautiousness during walking may lead to low velocity and short strides as observed in the present study.

There were characteristic changes of gait pattern in patients with DS. Kinematics revealed increased knee flexion at initial contact in comparison to the CG. This was associated with absence of the first peak of the knee extensor moment, indicative of relative weakness in stabilizing the knee. Knee flexion was increased throughout the stance phase but without a corresponding extensor moment. This also indicates insufficient strength of the knee extensors to stabilize the knee.

DS subjects walked with increased hip flexion throughout the gait cycle. In comparison with controls, subjects with DS had a prolonged hip extension moment in the second part of stance. This was probably due to a trunk forward lean in order to reduce the external knee flexion moment. The reduction of the hip range of motion is associated with the reduced stride length. Kinematic and kinetic changes were also observed at the ankle joint. In the DS group, the ankle was in more plantarflexion at initial contact. This was interpreted as an attempt to control knee extension. The ankle moment at loading response in the CG showed a short dorsiflexion pattern. This was absent in the DS group due to the reduced first rocker. In these patients, in fact, the ground reaction force was anteriorly positioned with respect to the ankle, resulting in an immediate internal plantarflexion ankle moment. At push of, however, plantarflexion was reduced as a consequence of a reduction in the propulsive force.

These gait alterations indicate a general functional muscle weakness. The increased joint stiffness that was observed may represent a compensatory mechanism for muscle weakness.

Interestingly a difference in joint stiffness patterns between the hip and ankle joint was found in this study.

Table 2
Joint stiffness ankle and hip in normals and DS

Subjects	K_h (N m/(kg degree))	K_a (N m/(kg degree))
DS	0.058 ± 0.025	0.058 ± 0.05
CG	0.028 ± 0.007	0.103 ± 0.014

Overall, joint stiffness was increased at the hip but was decreased at the ankle joint (Table 2). While hypotonia, and ligament laxity are thought to be the hallmarks of DS, these features may not be observed at every joint and under all conditions. For example, Webber et al. [18] reported ''postural stiffness'' during standing and higher overall stiffness for adults with DS compared to normals. It may be that these patients increased postural stability by increasing cocontraction [17]. In the present study increased hip joint stiffness was found, which is consistent with the literature. However, ankle joint stiffness was reduced. Limitations of the gait biomechanical model may have contributed to this finding. The foot with its complex anatomical structure is represented by a single rigid body. Typically in patients with DS, the foot is highly unstable and deformed. This additional hypermobility may mask the true magnitude of joint stiffness at the ankle. The functional problem of foot instability may further contribute to the lack of push-off force produced by these patients.

6. Conclusion

Patients with Down syndrome present with joint laxity and muscle hypotonia which cause functional weakness. The increased hip joint stiffness found in this study may be one mechanism of compensation.

Conflict of interest

All authors have no conflicts of interest and financial interest.

References

[1] Lai FM, Woo BH, Tan KH. Birth prevalence of Down syndrome in Singapore from 1993 to 1998. Singapore Med J 2002;43:70–6.
[2] Shumway-Cook A, Woollacott MH. Dynamics of postural control in the child with Down syndrome. Phys Ther 1985;65:1315–22.
[3] Aruin AS, Almeida GL, Latash ML. Organization of a simple two-joint synergy in individuals with Down syndrome. Am J Ment Retard 1996;101:256–68.
[4] Latash ML, Corcos DM. Kinematic and electromyographic characteristics of a single joint movements of individuals with Down syndrome. Am J Ment Retard 1991;96:189–201.
[5] Carr J. Mental and motor development in young Mongol children. J Ment Defic Res 1970;14:205–20.
[6] Molnar GE. Analysis of motor disorder in retarded infants and young children. Am J Ment Defic 1978;83:213–22.
[7] Bodensteiner JB, Smith SD, Schafer GB. Hypotonia, congenital hearing loss and hypoactive labyrinths. J Child Neurol 2003;18:171–3.
[8] Donoghue EC, Kirman BH, Bullmore GHL, Laban D, Abbas KA. Some factors affecting age of walking in mentally retarded population. Dev Med Child Neurol 1970;12:781–92.
[9] Oster J. Mongolism. Copenhagen: Einar Munksgaaard Forlag; 1953.
[10] Caselli MA, Cohen-Sobel E, Thompson J, Adler J, Gonzalez L. Biomechanical management of children and adolescents with Down syndrome. J Am Pediatr Med Assoc 1991;81:119–27.
[11] Parker AW, Bronks R. Gait of children with Down syndrome. Arch Phys Med Rehabil 1980;61:345–51.
[12] Galli M, Albertini G, Tenore N, Crivellini M. Gait analysis in children with Down syndrome. Progr Rep-Intern Rev Med Sci 2001;13:21–7.
[13] Cioni M, Cocilovo A, Rossi F, Paci D, Valle MS. Analysis of ankle kinetics during walking in individuals with Down syndrome. Am J Ment Retard 2001;106:470–8.
[15] Davis RB, Ounpuu S, Tyburski DJ, Gage JR. A gait analysis data collection and reduction technique. Hum Mov Sci 1991;10:575–87.
[16] Davis RB, De Luca A. Gait characterization via dynamic joint stiffness. Gait Posture 1996;4:224–31.
[17] Frigo C, Crenna P, Jensen LM. Moment–angle relationship at lower limb joints during human walking at different velocity. J Electromyogr Kinesiol 1996;6:177–90.
[18] Webber A, Virji-Babul N, Edwards R, Lesperance M. Stiffness and postural stability in adults with Down syndrome. Exp Brain Res 2004;155:450–8.
[20] McNee A, Shortland A, Eve L, Robinson R, Gough M. Lower limb extensor moments in children with spastic diplegic cerebral palsy. Gait Posture 2004;20(2):171–6.
[21] Roizen NJ, Patterson D. Down's syndrome. Lancet 2003;361(9365):1281–9.

Further reading

[14] Ferrigno G, Pedotti A. Elite: a digital dedicated hardware system for movement analysis via real-time TV signal processing. IEEE Trans Biomed Eng 1989;11:943–50.