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Gait control in spinal palsy

Bernard Dan^{a,b,c,*}, Ethel Bouillot^{b,c}, Leena D. Mewasingh^b, Christine Devalck^d,
Ana Bengoetxea^c, Catherine Christophe^e, Guy Chéron^{b,c}

^aDepartment of Neurology, Hôpital Universitaire des Enfants Reine Fabiola, Free University of Brussels (ULB),
15 Avenue JJ Crocq, 1020 Brussels, Belgium

^bCentre d'Analyse du Mouvement, Campus Universitaire Brugmann, Free University of Brussels (ULB), Brussels, Belgium

^cLaboratory of Movement Biomechanics, ISEPK, Free University of Brussels (ULB), Brussels, Belgium

^dDepartment of Oncology, Hôpital Universitaire des Enfants Reine Fabiola, Free University of Brussels (ULB), Brussels, Belgium

^eDepartment of Imaging, Hôpital Universitaire des Enfants Reine Fabiola, Free University of Brussels (ULB), Brussels, Belgium

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Abstract

Developmental motor impairment with lower limb spasticity most commonly corresponds to cerebral palsy of the spastic diplegia type. Here we describe a 4-year-old girl whose locomotor phenotype reflects early cortico-spinal lesion at the spinal level. This child has developmental spastic paraparesis secondary to D4–D8 cord compression. We analysed her gait using the ELITE optoelectronic system and compared it to that of six normal age-matched controls and six age-matched children with leucomalacic spastic diplegia. Gait characteristics of the patient included preservation of head orientation and arm swing similar to findings in normal controls and contrasting with children with spastic diplegia. She also had truncal instability and displayed lack of selectivity in lower limb movement as in spastic diplegia and in contrast with normal controls. This may reflect differences in locomotor control between developmental spasticity of cerebral and spinal origin. The latter might correspond to spinal palsy defined as abnormal movement and posture secondary to non-progressive pathological processes affecting the immature spinal cord.

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1. Introduction

Cerebral palsy has been defined as a disorder of movement and posture secondary to non-progressive pathological processes affecting the immature brain [1]. Early classification schemes for cerebral palsy included a category labelled 'paraplegia' for patients with predominant impairment of both lower limbs. Some authors still refer to paraplegia when the arms are 'minimally' as opposed to 'perceptibly' affected in spastic diplegia [2]. This is a source of confusion as the term paraplegia classically indicates involvement of the motor pathways in the thoracic or upper

lumbar cord. Furthermore, some cases of cord injury with residual muscle power in the lower limbs are probably misdiagnosed as cerebral palsy, particularly of the spastic diplegia type [3]. With improvement of obstetric techniques, cases of early spinal injury have become rare. Recently, Misson, Leroy, Truscetti and Otto described two children who were initially believed to have spastic diplegia but whose normal brain imaging suggested an extra-cerebral origin eventually confirmed by abnormal spinal imaging (6^{ème} Congrès de la Société Européenne de Neurologie Pédiatrique, Rome, 1997). So far, discussion regarding the cerebral or spinal origin has been based on the topology of motor manifestations and the presence of associated features of cerebral impairment rather than on motor organisation. Here we describe a child whose locomotor phenotype reflects early cortico-spinal lesion at the spinal level ('spinal palsy') in contradistinction to early cerebral lesion.

* Corresponding author. Address: Department of Neurology, Hôpital Universitaire des Enfants Reine Fabiola, Free University of Brussels (ULB), 15 Avenue JJ Crocq, 1020 Brussels, Belgium. Tel.: +32-2-4773174; fax: +32-2-4772176.

E-mail address: bernard.dan@ulb.ac.be (B. Dan).

2. Material and methods

2.1. Study population

2.1.1. Patient with early spinal lesion

The present analysis was conducted in a 4-year-old girl with isolated spastic paraparesis secondary to spinal compression by a posterior mediastinal neuroblastoma stage 4 diagnosed at 4 months. The tumour was successfully treated with chemotherapy (carboplatin, etoposide, cyclophosphamide, adriamycin, vincristine). The patient walked without support from 27 months. On spinal magnetic resonance imaging the cord appeared narrow from D4 to D8 (Fig. 1). Cerebral magnetic resonance imaging was normal. Neurological examination showed lower limb spasticity with distal predominance, hyperactive reflexes, bilateral Babinski sign, no fixed contractures and no other abnormality. In particular, there were no clinical signs of sensory, visual, auditory or autonomic disturbance. She had 2–3 weekly sessions of Bobath-type physiotherapy since the age of 17 months.

2.1.2. Normal controls

The normal control group consisted of six 4-year-old children (3 girls, 3 boys) with normal development and no disabilities.

2.1.3. Children with leucomalacic spastic diplegia

Another control group consisted of six 4-year-old children (2 girls, 4 boys) with spastic diplegia associated with periventricular leucomalacia and preterm birth (27–33 weeks' gestation; median 30.1). Independent sitting appeared from 10 to 17 months (median 12.8). Independent walking appeared between 21 and 36 months (median 29.0). All these patients had spastic hypertonia and hyperactive jerks more marked in the lower limbs and predominating distally, with ankle clonus and bilateral Babinski sign. They had mild axial hypotonia. There was no evidence of dystonia. None of these patients showed any fixed contractures. All the patients had Bobath-type physiotherapy at a frequency of 1–3 sessions per week since the first year of life.

2.2. Locomotor task

The subjects were asked to walk as naturally as possible at comfortable speed, looking straight forward, on the laboratory floor coated with a red band of linoleum (0.6 m wide and 8 m long). For each subject 10 trials were recorded in the same condition. Only the steps realised in the median 4 m of the ground band were considered.

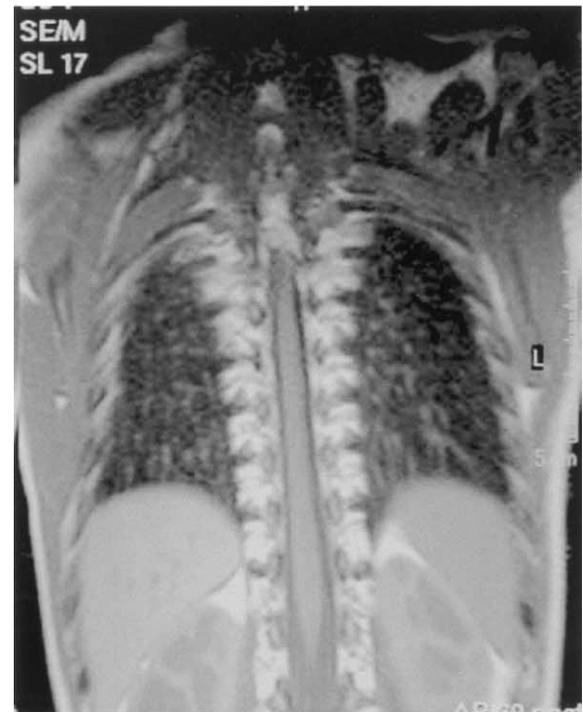
2.3. Gait recording

2.3.1. Recording equipment

The task was recorded using the optoelectronic ELITE system [4] following a standard protocol [5]. This system



(a)



(b)

Fig. 1. Spinal magnetic resonance imaging (coronal T1-weighted SE sequence, 600/30). (a) Pre-chemotherapy scan at 4 months of age showing a large right posterior and superior mediastinal tumour entering the spinal canal through neural foramina and compressing the spinal cord. (b) Post-chemotherapy scan at 7 months of age showing disappearance of the tumour.

consists of two charge-couple devices, infrared light-emitting cameras that detect reflective markers at a sampling rate of 100 Hz. The dedicated computer processes TV signals in real time by recognising the infrared reflectors on the basis of their shape and computing the 3D coordinates of the centres of these markers. The cameras were placed 4 m from the subjects. In the working field, accuracy was 0.67 mm.

2.3.2. Marker placement

The markers were adhesive plastic spheres (15 mm in diameter) covered with reflecting material. The subjects were barefoot, wearing only pants or light cloth that would not cover the markers or interfere with the movement. Markers were placed on the subjects' skin overlying the following anatomical landmarks: the lateral aspect of the nose at the height of the infra-orbital edge, the ear tragus, the upper limit of the acromion, the lateral epicondyle of the elbow, the styloid process of the wrist, the antero-superior iliac spine, the greater trochanter, the lateral condyle of the knee, the lateral malleolus and the distal end of the fifth metatarsal. Nine links between markers were considered as body segments in order to define a highly simplified multi-segmental model of the subjects. The position in space of the 10 markers were recorded and processed for real time recognition. Image coordinates of the marker centroids were reconstructed in three-dimensions.

2.4. Data analysis

2.4.1. General features of movement

After validation of the tridimensional reconstructs the sagittal, frontal and horizontal projections of the links and marker trajectories were inspected in order to appreciate the general features of the recorded movements. Walking speed, step length and cadence, which are descriptive linear stride measurements classically used as standard gait parameters, were defined according to classic recommendations [6].

2.4.2. Mathematical and statistical analysis

Mathematical and statistical analysis was performed using the Statistica Software for Windows 5.1 (Softcom, Tulsa, OK). Angular head orientation was calculated as the angle between the horizontal and line joining the orbital and ear tragus markers in the sagittal plane. Angular trunk orientation was calculated as the angle between the vertical and line joining the iliac spine and acromion markers in the sagittal plane. Thigh elevation angle (α_t) was calculated as the angle between the vertical and the trochanter-knee marker segment, shank elevation angle (α_s) as the angle between the vertical and the knee-malleolus marker segment and foot elevation angle (α_f) as that between the vertical and the malleolus-metatarsal marker segment (Fig. 2D). These parameters were investigated because the temporal waveform of the elevation angles of the lower limb

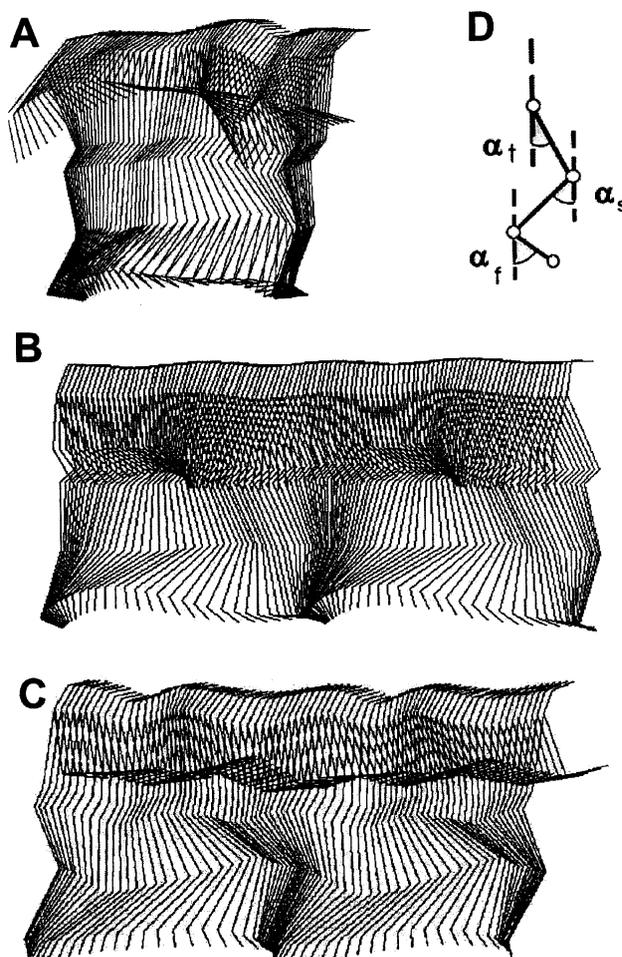


Fig. 2. Kinograms (stick diagrams) representing sagittal projection of locomotion. In these diagrams, two consecutive segments represent 20 ms. (A) One gait cycle of the child with spastic paraparesis. (B) Two gait cycles of a normal child. (C) Two gait cycles of a child with spastic diplegia. (D) Absolute angles of elevation of the thigh (α_t), shank (α_s), and foot (α_f) with respect to the vertical in the sagittal plane.

segments (pelvis, thigh, shank and foot) relative to the vertical is much more stereotypical across trials, speeds, and subjects than the corresponding waveform of either the joint angles [7,8] or the EMG patterns [8]. Moreover, the temporal changes of these elevation angles do not evolve independently of each other, but they are tightly coupled together [7]. When the elevation angles of the thigh, shank and foot are plotted one versus the others, they describe a regular gait loop, which lies close to a plane. The plane orientation and the shape of the loop reflect the phase relationship between the different segments and therefore the timing of intersegmental coordination [17], on which postural stability with respect to gravity and dynamic equilibrium for forward progression depend. The plane orientation shifts in a predictable way with increasing speed of walking [17]. Moreover, it reliably correlates with the mechanical energy expenditure [17]. Sets of data of all trials were pooled by groups (2.1–3). Means were compared by

analysis of variance (ANOVA). We analysed the characteristics of the lower limb elevation angles covariation by means of principal components analysis (for more details, see Ref. [5]). Briefly, this approach may be viewed as the performance of a rotation of the original space of variables in order to maximise the variance of a factor which would best represent the whole data set, thereby minimising the variance around this factor, i.e. first principal component. The same operation is repeated in order to extract another factor that maximises the remaining variance, extracting the second principal component and so on. Principal components are linear combinations of variates whose coefficients are the elements of the corresponding eigenvector of the sample covariance matrix. The principal components of the covariation of α_t , α_s and α_f during walking were computed by pooling together the samples of time-varying angles after subtraction of the mean value. The eigenvectors of the first two principal components characterise the plane fitting the data best. The normal vector of a particular plane corresponds to the third eigenvector. For each session the best-fitting plane of angular covariation was identified. The three-dimensional orientation of this plane in each child was compared with the mean orientation of the corresponding plane of the normal controls by computing the angle between the respective plane normal vectors data (for more details, see Refs. [5,20]).

2.5. Ethical aspects

This project has been approved by the local ethics committee. Informed consent was obtained from the parents.

3. Results

3.1. Standard gait parameters

The mean \pm SD of walking velocity, cadence and step length of the patient with spastic paraparesis, the normal controls and the children with spastic diplegia are displayed in Table 1. There were no significant differences in walking velocity and cadence between the three sets of data. Step length was significantly greater in normal controls ($P < 0.005$) than in the patient with spastic paraparesis and in

Table 1
Standard gait parameters in spinal palsy, normal controls and spastic diplegia

	Spinal palsy	Normal controls	Spastic diplegia
Walking velocity (ms^{-1})	0.55 ± 0.42	0.88 ± 0.20	0.71 ± 0.25
Walking cadence (steps/min)	55.8 ± 6.2	61.4 ± 6.6	68.7 ± 13.3
Step length (m)	0.59 ± 0.25	0.86 ± 0.23	0.62 ± 0.14

children with spastic diplegia, between whom there was no significant difference.

3.2. Global description of the movement

Fig. 2 shows sagittal kinograms of one gait cycle of the patient with spastic paraparesis (for clarity of the display) and two consecutive gait cycles of a normal child and a child with spastic diplegia. The patient with paraparesis (Fig. 2A) maintained stable angular orientation of the head but had marked variation in trunk inclination. The shoulder was maintained in extension and the arm swing was particularly ample at the level of the elbow. The hip, knee and ankle tended to keep a semi-flexed attitude throughout the walking cycle. In particular, the knee remained flexed during the swing phase. Normal children (Fig. 2B) also maintained the angular orientation of their head throughout the gait cycle, but also that of the trunk. The arm swing was characterised by shoulder extension and minimal elbow flexion at the beginning of the stance phase smoothly changing to flexion that increased through the swing phase. The hip was flexed at the onset of the stance phase and extended at the end of it, with a mirror pattern seen during the swing phase. The knee and ankle angular movements consisted of alternating flexion and extension, knee extension being maximal at the heel strike and at the toe-off. In contrast, children with spastic diplegia (Fig. 2C) showed marked variation in both head and trunk orientation. The upward deviation of the head orientation corresponded to a backward displacement of the trunk, that was maximal at the time of ground contact and at mid-stance (corresponding to the other foot ground contact). Conversely, a downward deviation of the head orientation was associated with a forward trunk displacement. There was no arm swing. Instead, the patients tended to maintain a stereotyped flexor attitude of the shoulder associated with fixed elbow flexion throughout the gait cycle. Like the patient with spastic paraparesis, these children retained a semi-flexed attitude of the hip throughout the cycle. Similarly, the knee and the ankle remained flexed during the whole cycle, notably resulting in the absence of heel strike.

3.3. Head and trunk stability

Peak-to-peak angular deviation of the head during walking was minimal and similar in the child with paraparesis ($5.4 \pm 4.9^\circ$) and normal controls ($4.9 \pm 3.4^\circ$). It was significantly greater ($P < 0.01$) in children with diplegia ($10.7 \pm 6.1^\circ$). However, peak-to-peak trunk angular deviation was significantly smaller ($P < 0.01$) in normal controls ($7.3 \pm 2.0^\circ$) than in the patient with paraparesis ($15.2 \pm 6.6^\circ$) and children with diplegia ($9.8 \pm 3.8^\circ$).

3.4. Lower limb intersegmental coordination

The mean values and SDs of the percentage of data variance accounted for by the first, second and third

principal components (PV1, PV2 and PV3, respectively) showed no significant differences between sets of data: 66.87 ± 5.41 , 31.50 ± 5.14 and 1.62 ± 0.34 , respectively, for the patient with paraparesis, 75.1 ± 4.6 , 23.8 ± 4.5 and 1.1 ± 0.8 , respectively, for normal controls and 71.9 ± 8.4 , 26.5 ± 7.9 and 1.5 ± 1.3 , respectively, for patients with spastic diplegia. PV3 was close to zero in all sets of data, indicating that the orthogonal planar regression accounted for close to 99% of the data variance, hence planar covariation. The three-dimensional orientation of the planar covariation is another important parameter of the intersegmental coordination because it reflects the phase relationship between the different segments. It was calculated as the angle between the best-fitting plane representing the three-dimensional plot of the time-courses of the elevation angle of the thigh versus that of the shank versus that of the foot for each walking trial and the mean plane best-fitting the corresponding angles in the control group. The angular orientation of the covariation planes was similar in the child with spastic paraparesis ($10.08 \pm 5.55^\circ$) and children with spastic diplegia ($17.3 \pm 14.7^\circ$), significantly differing from normal controls ($P < 0.01$).

4. Discussion

Analysis of gait of this patient with chronic motor impairment due to early occurrence of a stable lesion to the spinal cord showed a different pattern from that found in normal controls and in patients with leukomalacic spastic diplegia. The most remarkable features were head stability despite marked truncal instability and non-selective (i.e. global) intersegmental lower limb coordination (Table 2).

Head stability has been described as one of the most consistent features of locomotion [9] and various other motor tasks performed by normal subjects [10,11]. Head stability implies stability of afferents from the visual, orbital, auditory, vestibular and somatosensory systems, which has been hypothesised to ease integration of afferent information into a neural code representing the location of the head and body with respect to the outside world [12]. This process appears particularly crucial with respect to gravity [13] although stability of head orientation during

movement of other body parts also persists in weightlessness [14]. The angular stability of the head is normally ensured by three reflexes that integrate afferent information for head control, namely the tonic neck reflex, the opto-cervical reflex and the vestibulo-colic reflex, compensating for perceived deviations by appropriate contraction of neck muscles. The efferent pathways of these reflexes are under hemispheric control, which might account for the lack of head stability observed in patients with spastic diplegia in the present study as well as during other movements [11].

Investigations of the stabilisation of human body position have indicated that the main object of regulation for maintaining an erect posture is the trunk [15]. The level of trunk stability is also a distinctive feature of mature versus immature movement [5,16]. Trunk stabilisation emerges gradually with reduction in pitch and roll displacements between 3 and 6 months following the onset of walking [5] and postural stabilisation has been shown to improve the mechanical efficiency of walking [16]. The planar covariation of the elevation angles of the lower limb segments has been shown to be functionally significant for the mechanics of walking [17,18]. In particular, the covariation plane orientation reliably correlates with the mechanical energy expenditure [17]. As an expression of lower limb intersegmental coordination patterns, this parameter has been demonstrated to reflect a consistent motor control strategy for the reduction in kinematic degrees of freedom in human walking in healthy adults [18] and children [5]. It has also been studied in various conditions including Parkinson's disease [19], hereditary spastic paraparesis [20] and Angelman syndrome [21]. The covariation plane orientation reflects the phase relationship between segments and therefore the timing of intersegmental coordination, on which postural stability with respect to gravity and dynamic equilibrium for forward progression depend. The planar covariation may represent an aspect of phase coupling between units of central pattern generators (CPGs) driving the limb segments during locomotion. CPGs are neuronal networks that are able to generate rhythmic commands producing organised rhythmic movement [22]. Such rhythmic patterns can be recorded at several levels during walking in mammals, suggesting spinal and supraspinal control [23]. In the patient with early onset spastic paraparesis, abnormal CPG control at the spinal level is likely. In this child, the covariation plane differed significantly from normal controls but was similar to patients with spastic diplegia. Lack of condition-specificity in alteration of plane orientation was noted previously in adult hereditary spastic paraparesis [20] and Parkinson's disease [19]. This would be consistent with abnormal tuning of the highly organised CPG exerted at different levels by different neural structures in the different conditions.

The correlation between trunk deviation with respect to the vertical and covariation plane orientation has been demonstrated in healthy toddlers [5]. It has also been studied in patients with Parkinson's disease, in whom trunk

Table 2
Summary of gait features in spinal palsy, normal controls and spastic diplegia

	Spinal palsy	Normal controls	Spastic diplegia
Head stability	+	+	–
Trunk stability	–	+	–
Arm swing	+	+	–
Lower limb planar covariation	+	+	+
Lower limb selectivity	–	+	–

inclination and covariation plane orientation practically normalised following apomorphine infusion or pallidal stimulation [19].

Postural control of the trunk has been shown to be particularly crucial in paraplegia, where anticipatory postural adjustments to movement are longer and involve more trunk muscles than in healthy controls [24]. In our patient, the role of arm swing may have been of utmost importance in this respect. This can be contrasted with the absence of arm swing in patients with spastic diplegia and seems to be reinforced by the clinical observation that crossing the hands over the shoulders resulted in deteriorating perceived balance during walking in the patient with spastic paraparesis. This manoeuvre tended to improve perceived balance in patients with spastic diplegia and did not alter it in normal controls.

5. Conclusion

This study suggests that there may be differences in locomotor control between developmental spasticity of cerebral and spinal origin. Distinctive features include relative preservation of head orientation, trunk instability, preserved arm swing and lack of selectivity in lower limb movement. The concept of cerebral palsy [1] has proved clinically useful, particularly for management purposes. There may be a case for proposing a germane concept of spinal palsy defined as abnormal movement and posture secondary to non-progressive pathological processes affecting the immature spinal cord. This would apply to our patient because although the initial insult was due to a tumour, the lesion occurred at an early stage (i.e. before gait development) and remained non-evolutive, as the tumour was successfully treated. This proposed entity is likely to be a rare occurrence. More studies are required to refine this concept.

References

- [1] Bax MCO. Terminology and classification of cerebral palsy. *Dev Med Child Neurol* 1964;6:295–7.
- [2] Stanley F, Blair E, Alberman E. Cerebral palsies: epidemiology & causal pathways. *Clinics in developmental medicine* no. 151. London: Mac Keith Press; 2000.
- [3] Brett EM. Cerebral palsy, perinatal injury to the spinal cord and brachial plexus injury. In: Brett EM, editor. *Paediatric neurology*, 2nd ed. Edinburgh: Churchill Livingstone; 1991. p. 285–316.
- [4] Ferrigno G, Pedotti A. ELITE: a digital dedicated hardware system for movement analysis via real-time TV signal processing. *IEEE Trans Biomed Eng* 1985;32:943–50.
- [5] Cheron G, Bouillot E, Dan B, Bengoetxea A, Draye JP, Lacquaniti F. Development of a kinematic coordination pattern in toddler locomotion: planar covariation. *Exp Brain Res* 2001;137:455–66.
- [6] Winter DA. *The biomechanics and motor control of human gait: normal, elderly and pathological*, 2nd ed. Waterloo, IA: University of Waterloo Press; 1991.
- [7] Borghese NA, Bianchi L, Lacquaniti F. Kinematic determinants of human locomotion. *J Physiol* 1996;494:863–79.
- [8] Grasso R, Bianchi L, Lacquaniti F. Motor patterns for human gait: backward versus forward locomotion. *J Neurophysiol* 1998;80:1868–85.
- [9] Pozzo T, Berthoz A, Lefort L. Head stabilization during various locomotor tasks in humans. I. Normal subjects. *Exp Brain Res* 1990;82:97–106.
- [10] Pozzo T, Levik Y, Berthoz A. Head and trunk movements in the frontal plane during complex dynamic equilibrium tasks in humans. *Exp Brain Res* 1995;106:327–38.
- [11] Dan B, Bouillot E, Bengoetxea A, Noël P, Kahn A, Cheron G. Head stability during whole body movements in spastic diplegia. *Brain Dev* 2000;22:99–101.
- [12] Andersen RA, Essick GK, Siegel RM. Encoding of spatial location by posterior parietal neurons. *Science* 1985;230:456–8.
- [13] Massion J. Movement, posture and equilibrium: interaction and coordination. *Prog Neurobiol* 1992;38:35–56.
- [14] Amblard B, Assaiante C, Vaugoyeau M, Baroni G, Ferrigno G, Pedotti A. Voluntary head stabilisation in space during oscillatory trunk movements in the frontal plane performed before, during and after a prolonged period of weightlessness. *Exp Brain Res* 2001;137:170–9.
- [15] Oddsson LI. Control of voluntary trunk movements in man. Mechanisms for postural equilibrium during standing. *Acta Physiol Scand Suppl* 1990;595:1–60.
- [16] Ledebt A, Bril B, Wiener-Vacher S. Trunk and head stabilization during the first months of independent walking. *NeuroReport* 1995;6:1737–40.
- [17] Bianchi L, Angelini D, Orani GP, Lacquaniti F. Kinematic coordination in human gait: relation to mechanical energy cost. *J Neurophysiol* 1998;79:2155–70.
- [18] Lacquaniti F, Grasso R, Zago M. Motor patterns in walking. *News Physiol Sci* 1999;14:168–74.
- [19] Grasso R, Peppe A, Stratta F, Angelini D, Zago M, Stanzione P, Lacquaniti F. Basal ganglia and gait control: apomorphine administration and internal pallidum stimulation in Parkinson's disease. *Exp Brain Res* 1999;126:139–48.
- [20] Dan B, Bouillot E, Bengoetxea A, Cheron G. Effect of intrathecal baclofen on gait control in human hereditary spastic paraparesis. *Neurosci Lett* 2000;280:175–8.
- [21] Dan B, Bouillot E, Bengoetxea A, Cheron G. Lower limb coordination patterns in locomotion in Angelman syndrome. *Mov Disord* 2000;15(Suppl 3):234–5.
- [22] Grillner S. Locomotion in vertebrates: central mechanisms and reflex interaction. *Physiol Rev* 1975;55:247–304.
- [23] McCrea DA. Spinal circuitry of sensorimotor control of locomotion. *J Physiol* 2001;533:41–50.
- [24] Bouisset S, Do MC, Zattara M. Posturo-kinetic capacity assessed in paraplegics and Parkinsonians. In: Woollacott M, Horak F, editors. *Posture and gait: control mechanics*. Portland, OR: University of Oregon Books; 1992.