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Spine kinematics during gait in paediatric Hereditary Spastic Paraparesis

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ABSTRACT

Background: Many studies already addressed specific gait abnormalities in children affected by Hereditary Spastic Paraparesis (HSP). Some authors investigated the contribution of the upper body to walking pattern, but simplifying trunk and pelvis as two hinged rigid bodies. Recently, we developed a method to detail spinal kinematics in terms of anatomic curvatures and length; we were thus interested in applying such protocol to HSP.
Research question: how HSP influences spinal kinematics during gait?
Methods: we enrolled ten HSP patients (5–17 years, 8 males) and twelve Healthy Children (HC, 8–16 years, 4 males). Kinematic data were recorded with an optoelectronic system using the LAMB full body marker set, which included three physical markers placed on the spine, supplemented with a virtual one reconstructed on the coccix. Calculations included the spinal length (linear distance from C7 to coccix), the kyphosis and lordosis angles, the trunk tilt and obliquity, the pelvis and the shoulder-pelvis angles, as well as the joint angles of the lower limbs. For each variable, the average value and the range of motion (ROM) were extracted and compared between groups.
Results: the ROM of spinal length, the average value and ROM of kyphosis angle and the average value of trunk tilt significantly increased in HSP vs HC. A pathologic "double bump" pattern characterized the pelvic tilt traces, the lordosis angles and, with opposite sign, the kyphosis. Both the average value and ROM of pelvic tilt significantly increased in HSP, while ROM of lower limb angles was reduced.
Conclusion: spine kinematics were altered in HSP, who also showed an anterior trunk tilt. Therefore, the trunk should be considered an articulated system and not simplified to a rigid body, a perspective that could be also used in treating gait abnormalities.

1. Introduction

Hereditary Spastic Paraparesis (HSP) is a heterogeneous group of disorders caused by specific degeneration of corticospinal tracts [1], clinically divided into pure and complex forms [2]. Pure HSPs are characterized by slowly progressive lower extremity spasticity and weakness, while complex forms are characterized by the presence of additional neurological or non-neurological features with more variable phenotypes (symptoms may include impaired vision, deafness, peripheral neuropathy, ataxia, cognitive impairment, and epilepsy).

The diagnosis is based on clinical and genetic investigations [3]. In clinical practice, obtaining a probable or possible diagnosis of a HSP is relatively straightforward. However, it is often difficult to reach a precise diagnosis because of the high levels of clinical and genetic heterogeneity and the different patterns of inheritance [4]. Therefore, diagnosis relies mostly on clinical evaluations.

Considering that lower limb weakness and spasticity are the predominant signs and symptoms of HSP, causing motor deficit with reduced walking ability, many studies quantified specific gait abnormalities [5]. Some of these studies addressed children [6–11], but only a few have investigated the contribution of the upper body to the walking pattern [9–11], reporting several alterations in trunk movements in the sagittal and frontal planes [10,11]. The importance of the upper body has been also highlighted in asymptomatic adults by showing that variations in spinal curvatures and postural asset at rest correlate with specific changes in gait kinematics [12,13]. Moreover, trunk motion, generally seen as passively dependant on lower limb activity, seems instead to play an active role in counterbalancing the lower extremities

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in the swing phase, as well as in reducing the speed of rotation toward the contralateral side immediately before the contralateral heel strike [14]. However, trunk and pelvis were modelled as two hinged rigid bodies, thus ignoring the anatomically relevant curvatures of the spine.

Considering that we recently developed a method to detail spinal kinematics in terms of kyphosis, lordosis and length [15], it becomes of interest to apply this method in children with HSP, so as to disclose which are the determinants, in terms of spinal anatomic curvatures, of the already described increase in the angle between trunk and pelvis [10,11]. Should we find that the lordosis and kyphosis angles change in phase, it would be of interest to disclose which one has the larger range of motion, thus driving the trunk-pelvis angle; if instead the two spinal angles changed in antiphase, one of the two would drive the trunk-pelvis angle while the other one would act in a compensatory way. Since trunk movements are linked to pelvis kinematics [11], pelvis tilt, obliquity and rotation, as well as shoulder-pelvis angles, were also analysed. Finally, taking into account that different patterns may be recognized in the pathological gait of HSP children, according to the kinematics of pelvis and lower limbs in the sagittal plane [8], we evaluated the hip, knee and ankle angles in that plane so as to draw a direct comparison.

2. Methods

2.1. Participants and procedure

Patients were selected and enrolled at the "Istituto Neurologico Carlo Besta" of Milan. The main inclusion criteria were: ascertained clinical diagnosis of pure Type I HSP, preservation of standing and walking without assistance; an IQ $\geq$ 70, i.e., children were sufficiently cooperative and able to understand the required tasks. Moreover, twelve healthy children (HC), typically developing, were enrolled as a control group. Demographic and anthropometric data are reported in Table 1. No statistical differences were found between both groups (Mann-Whitney *U* test, p-value set at 0.05).

The experimental procedure was carried out in accordance with the standards of the Declaration of Helsinki. The Ethical Committee "Comitato Etico di Ateneo dell'Università degli Studi di Milano" approved both the study and the written consent procedure (counsel 23/23). Before each acquisition, a child neuropsychiatrist and the experimenters explained the aim of the study and the details of the procedure to the parents as well as to the child. Only those children who agreed with the study and whose parents/guardians consented their participation were recruited in the study; they were free to abandon the experimental procedure at any time. All participants/parents/guardians signed the written, informed consent form.

Participants were instructed to walk along a linear eight-meter pathway at a self-selected speed. The task was repeated 2–8 times according to individual compliance. Children were allowed to rest in between trials.

Table 1
Demographic and anthropometric measurements of the HSP (Hereditary Spastic Paraparesis) and HC (healthy children). Data are shown as median value (min - max).

Group	HSP	HC
Gender (Nmales/Ntot)	8/10	4/12
Age (yrs)	13.5 (5.0 – 17.0)	11.5 (8.0 – 16.0)
Height (cm)	162.6 (101.3–173.0)	149.7 (124.7–177.5)
Weight (kg)	53.2 (20.3–89.7)	42.2 (28.5–64.9)
BMI (kg/m^2)	21.0 (16.0–30.9)	18.3 (16.4–23.3)

2.2. Kinematic recordings

Kinematic data were collected by means of a motion analysis system (SMART-DX, BTS, Italy) with 8 cameras (sampling frequency of 100 Hz), using the LAMB full-body marker set [16,17].

Following the methodology described in detail by Palmisano et al. [15], spinal variables were computed thanks to three physical markers, placed on: (1) the seventh spinous vertebral process (C7); (2) the point of maximum kyphosis (MAX_KYPH); (3) the middle position between the two posterior superior iliac spines (PSIS_MX), and one virtual marker reconstructing the coccyx position. The position of the coccyx was estimated, relative to the PSIS_MX, in the pelvis local reference system (px, py and pz, see Fig. 1) using the results of previous radiography studies in adults [18,19] and resizing them taking into account the changes in pelvis dimensions according to children age [20]. The resulting point was then reprojected into the laboratory reference system.

Upper body kinematic measurements included [15,21]:

a) the *spinal length* (length of the vector connecting C7 to coccyx markers), expressed in % of subject's height;
b) the *angle of kyphosis* (between the vector connecting C7 to MAX_KYPH and the vector connecting MAX_KYPH to PSIS_MX) and the *angle of lordosis* (between the vector described by MAX_KYPH and PSIS_MX and the vector from PSIS_MX to the coccyx); both angles were projected to the pelvis sagittal plane to account for small deviations of the individual orientation with respect to the laboratory axes;
c) the angles of *trunk tilt* and *obliquity,* as well as of *pelvic tilt*, *obliquity* and *rotation*, all measured in relation to the planes of the laboratory;
d) the angles of *shoulder orientation,* computed as the angles between the vector connecting the two acromion markers and the vector connecting the two anterior superior iliac spines, projected to the frontal ($\alpha 4$) and horizontal ($\alpha 6$) planes of the pelvis reference system.

Regarding lower limb kinematics, local coordinate systems were defined for the thigh (tx, ty and tz, see Fig. 1), the shank and the foot. For the shank and the foot, only the longitudinal axes were considered (sy and fy, see Fig. 1). Then, the anatomical angles of the joints in the sagittal plane were obtained as:

$$\theta_{Flex-Extension}(hip) = \cos^{-1}\left(\frac{tx \bullet py}{|tx|\ |py|}\right) \quad (1)$$

$$\theta_{Flex-Extension}(knee) = \cos^{-1}\left(\frac{tx \bullet (sy \wedge tz)}{|tx|\ |sy \wedge tz|}\right) \quad (2)$$

$$\theta_{Dorsi-plantar\ flexion}(ankle) = \cos^{-1}\left(\frac{fy \bullet sy}{|fy|\ |sy|}\right) \quad (3)$$

2.3. Data analysis and statistics

Data were low pass filtered at 10 Hz with a 3rd order Butterworth filter and interpolated with a cubic spline. In each participant, left and right gait cycles were identified thanks to the markers placed on the heels. The spatiotemporal parameters (step length and duration; stride length, duration, mean velocity and width; duration of stance, swing and double support) were calculated and separately averaged for the right and left sides.

To inquire for possible asymmetries between right and left cycles, the right and left values of each parameter were compared within each group with a paired Wilcoxon test. Also, the absolute difference between the two values were calculated and compared across the two groups with a Mann-Whitney *U* test. Given the symmetry of gait found in the two groups (see Results), left and right values of the spatiotemporal parameters were averaged in each participant and comparison were drawn

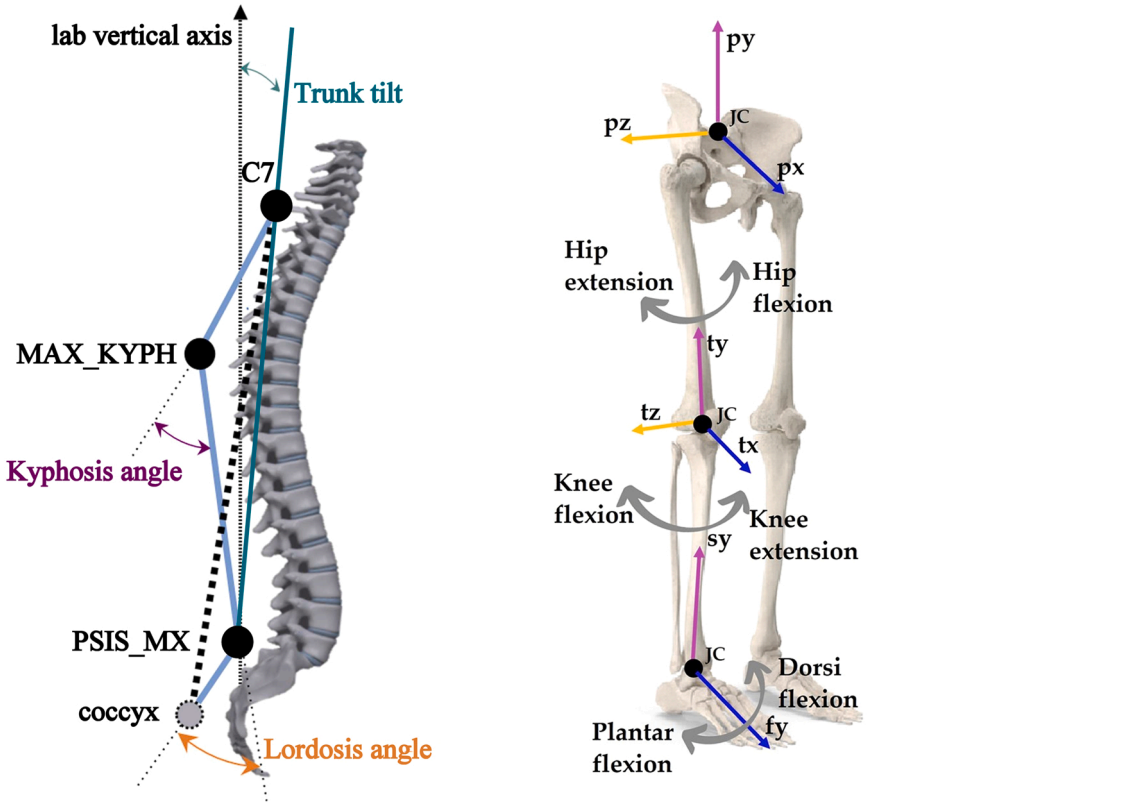

Fig. 1. On the left, spinal variables in sagittal view. Physical markers (black) were placed at the seventh cervical vertebra (C7), the point of maximum kyphosis (MAX_KYPH) and the midpoint between the posterior superior iliac spines (PSIS_MX). A virtual marker (gray) was reconstructed on the coccyx. The vectors connecting these points allowed the calculation of kyphosis and lordosis angles. Trunk angles were obtained by considering the inclination of the vector connecting PSIS_MX and C7 with respect to the vertical axis of the laboratory. The projections on the sagittal and frontal planes defined trunk tilt and obliquity, respectively. The spinal length (dashed line) was calculated as the length of the vector between the coccyx and C7. Adapted from [15]. On the right, local coordinate systems for the pelvis (p) and the thigh (t). JC: joint center; x: anteroposterior axis (blue), y: vertical axis (pink), z: mediolateral axis (yellow).

between the population data of the two groups (Mann-Whitney *U* test).

Similarly, the angles of the left and right lower limbs were calculated throughout their respective gait cycles, then traces of both sides were time-normalised using cubic spline interpolation and averaged. Instead, for the upper-body kinematics (spine, shoulders, trunk and pelvis), the normalised right gait cycle was arbitrarily chosen as the representative time base.

In each individual, for each kinematic variable we extracted its average value and its peak-to-peak amplitude, i.e, the range of motion (ROM). Average values, as well as ROM, were then compared between groups with Mann-Whitney U tests. The level of significance was set to 0.05 for all statistical tests. All computations were performed with ad hoc algorithms written in Matlab® (R2021b, The Mathworks, USA).

3. Results

3.1. Gait spatiotemporal parameters

The analysis of spatiotemporal parameters showed symmetry between the two body sides in both healthy and HSP children. In fact, for each parameter (step length and duration; stride length, duration, mean velocity and width; duration of stance, swing and double support) the difference between the average right and left values did not reach significance within each group.

Compared to healthy peers, the HSP group showed reduced walking velocity, swing duration, step and stride length (Table 2). In contrast, the stride width and the duration of stance and double support phases increased significantly. No difference was found in step and stride duration.

Table 2
Median (min - max) values of gait spatiotemporal parameters in the HSP and HC group. *: $p < 0.05$, Mann-Whitney *U* test HSP *vs* HC.

	HSP	HC
Step length (m)	**0.46*** (0.27 –0.63)	0.61 (0.53 –0.78)
Step duration (s)	0.52 (0.46–0.66)	0.52 (0.48–0.63)
Stride length (m)	**0.92*** (0.54 –1.28)	1.23 (1.06 –1.56)
Stride duration (s)	1.05 (0.92 –1.31)	1.05 (0.95 –1.25)
Stride velocity (m/s)	**0.85*** (0.59 –1.12)	1.09 (0.87 –1.47)
Stride width (m)	**0.12*** (0.04 –0.15)	0.07 (0.01 –0.09)
Stance duration (% gait cycle)	**63.29*** (60.21 –67.39)	61.24 (57.42 –63.72)
Swing duration (% gait cycle)	**37.09*** (32.61 –39.78)	38.6 (36.27 – 42.57)
Double support duration (% gait cycle)	**13.12*** (10.75 –17.39)	11.16 (7.92 – 13.72)

3.2. Body kinematics

During the gait cycle, no difference was found between HSP and HC participants with regard to the average value of spinal length while the ROM greatly increased in patients (about 3 times, Table 3). For what concerns the kyphosis angle, both average and ROM values were larger in HSP. No significant changes were found in the lordosis angle, although its ROM increased in patients.

Table 3
Body kinematics during walking. Average value (AVG) and range of motion (ROM) are shown as the median value (min – max) for HSP children and HC; *: $p < 0.05$, Mann-Whitney *U* test HSP *vs* HC.

		HSP	HC
Spinal length (% of height)	AVG	33.03 (28.92 –40.95)	32.65 (31.02 –35.96)
	ROM	**1.73*** (0.62 –3.76)	0.64 (0.45 – 1.03)
Kyphosis angle (°)	AVG	**11.10*** (2.74 –20.40)	17.85 (8.83–22.17)
	ROM	**5.75*** (2.82 –12.31)	2.65 (1.39 –6.97)
Lordosis angle (°)	AVG	38.24 (25.04–54.02)	37.29 (27.28–42.88)
	ROM	6.42 (2.01–13.29)	4.08 (2.48–5.20)
Shoulder α4 (°)	AVG	−0.62 (−4.99– 3.40)	0.22 (−1.17 – 4.73)
	ROM	14.36 (10.95 – 32.59)	12.86 (6.28 – 21.50)
Shoulder α6 (°)	AVG	1.29 (−3.92 – 9.38)	1.44 (−0.60 – 6.20)
	ROM	18.72 (9.91 – 27.89)	16.03 (7.06 – 25.31)
Trunk tilt (°)	AVG	**8.71*** (2.88 – 15.37)	5.84 (2.36–14.50)
	ROM	3.99 (1.42 – 4.78)	3.30 (2.32 – 4.72)
Trunk obliquity (°)	AVG	−0.20 (−1.67 – 0.76)	0.11 (−0.60 – 0.74)
	ROM	5.83 (3.77 – 16.68)	5.81 (3.18 – 7.16)
Pelvic tilt (°)	AVG	**11.96*** (−1.56 – 23.20)	4.15 (−2.85 – 11.65)
	ROM	**7.37*** (2.09 – 12.86)	3.06 (1.74 – 5.85)
Pelvic obliquity (°)	AVG	0.76 (−2.26 – 3.43)	0.85 (−1.40 – 4.18)
	ROM	**5.44*** (3.36 – 6.93)	10.15 (4.53 – 14.00)
Pelvic rotation (°)	AVG	**3.20*** (−10.78 – 6.24)	0.14 (−2.08 – 3.87)
	ROM	17.64 (8.74 – 25.71)	13.41 (7.04 – 19.34)
Hip flex-extension (°)	AVG	**17.22*** (6.46 – 30.31)	10.32 (1.37 – 18.40)
	ROM	**40.01*** (35.93 –46.55)	45.59 (40.37 –53.80)
Knee flex-extension (°)	AVG	23.23 (20.79–30.25)	22.21 (18.81–26.70)
	ROM	**42.35*** (31.80–56.18)	58.49 (52.01–63.89)
Ankle dorsi-plantarflexion (°)	AVG	−2.62 (−8.09–3.78)	−3.08 (−8.14 – 0.01)
	ROM	**20.58*** (14.12–36.83)	33.71 (29.53 – 38.99)

No differences were found for shoulder angles (α4 and α6, Table 3). Regarding the trunk, only the average value of tilt was significantly larger in HSP, with no difference in the ROM; obliquity was unchanged.

For what concerns the pelvis, both the average pelvic tilt and its ROM increased significantly in patients compared to healthy peers (of about 5° and 4°, respectively) while the ROM of obliquity halved (no changes in the average value). Average value of pelvic rotation was significantly larger and oriented to the left (about 3°) in the patient group, while no difference was found in the ROM.

Interestingly, the pelvic tilt traces in HSP showed a "double bump", with a first peak at 20 % of the gait cycle and the second at the beginning of the swing phase (Fig. 2): a well-known pathological feature [8,10]. The same pattern was present in the lordosis and, with opposite sign, in the kyphosis angle.

Regarding the lower limbs (Fig. 3), only the average of the hip flexion-extension significantly increased in HSP, while the ROM of all joints (hip, knee and ankle) was statistically reduced (Table 3).

4. Discussion

Although Hereditary Spastic Paraparesis is known as a condition that primarily affects the lower limbs, our results showed that alterations are also evident in the upper body kinematics. Previous studies [9–11] have shown changes in the trunk kinematics, but none have yet assessed the postural alignment of the spine regions, i.e. the angles of kyphosis and lordosis, which were shown to relevantly impact gait performance [15]. In agreement with Bonnefoy-Mazure et al. [10], our results revealed increased trunk tilt while walking in the HSP children. Even if no spinal symptoms are clinically related to HSP, present results provide evidence that the alterations in lower limb movements reflect into altered spine kinematics during walking.

By observing the average value of spinal length, kyphosis angle and lordosis angle, only kyphosis was significantly larger in HSP. In contrast, the ROM was greater in HSP for all the three variables (although not significantly for lordosis).

Of note, the time course and alignment of these spine variables may indicate a different relationship between the spinal curvatures and spinal length in the two groups. In HC, kyphosis and lordosis angles were almost in phase and with a small ROM. In contrast, in HSP the two angles changed in antiphase, with the kyphosis angle resembling the pattern observed in HC, but with a larger excursion. Both kyphosis and lordosis angles contribute to spinal length, as the larger they are the shorter the spine. It might then be speculated that phase opposition between kyphosis and lordosis in HSP might be seen as a strategy to compensate for excessive changes in the spinal length over the gait cycle. If so, one should have expected that the percentage increase in the ROM of the spinal length was lower than the corresponding increase in the kyphosis and lordosis angles. Instead, the increase in spinal length in HSP *vs.* HC was larger than that of the other two variables, actually in contrast with such a possible compensatory mechanism. An alternative explanation for the antiphase changes in spinal curvatures is that, given the stiffness in their lower limbs, HSP children adapted their kyphosis and lordosis so as to keep their head vertically aligned to the pelvis, thus preserving balance. In fact, it has been shown that walking is worse in adults with spinal deformities associated with a misalignment of the head-pelvis axis [22].

Going down the kinematic chain, the pelvic tilt in HSP also showed the characteristic "double bump" [8,10] with larger amplitudes of anterior pelvic tilt than HC. This could be related to the weakness of hip extensors and hamstrings, altering the loading response, but may also reflect the reduced ankle ROM and/or an insufficient third rocker. Pelvic changes were also found in the frontal and transverse planes. These two parameters have been described as belonging to the group of "determinants of gait", i.e. parameters capable of minimizing the movement of the center of mass, producing energy-efficient locomotion [23,24]. In fact, the rotation of the pelvis would reduce the magnitude of the fall of the center of mass that occurs during double support while the movement of the pelvis in the frontal plane lowers it during single leg stance [25]. Abnormal pelvic movements in neurological patients may be due to compensation for weakened and spastic muscles [26–29]. Indeed, in children with cerebral palsy (CP), the increase in pelvic tilt ROM is associated with proximal spasticity and a contracture-related hip extension deficit, while the increase of the average pelvic tilt is rather a consequence of the strength imbalance between the knee flexors and extensors [30]. The many similarities in spatiotemporal and kinematic gait parameters found between CP and HSP children [6,8] might suggest that also the causes of the abnormal pelvic movements could be similar.

Moving towards the lower extremities, interesting data were collected by Pulido et al. [8], who clustered the result from a large number of HSP, identifying six kinematics patterns. In our population

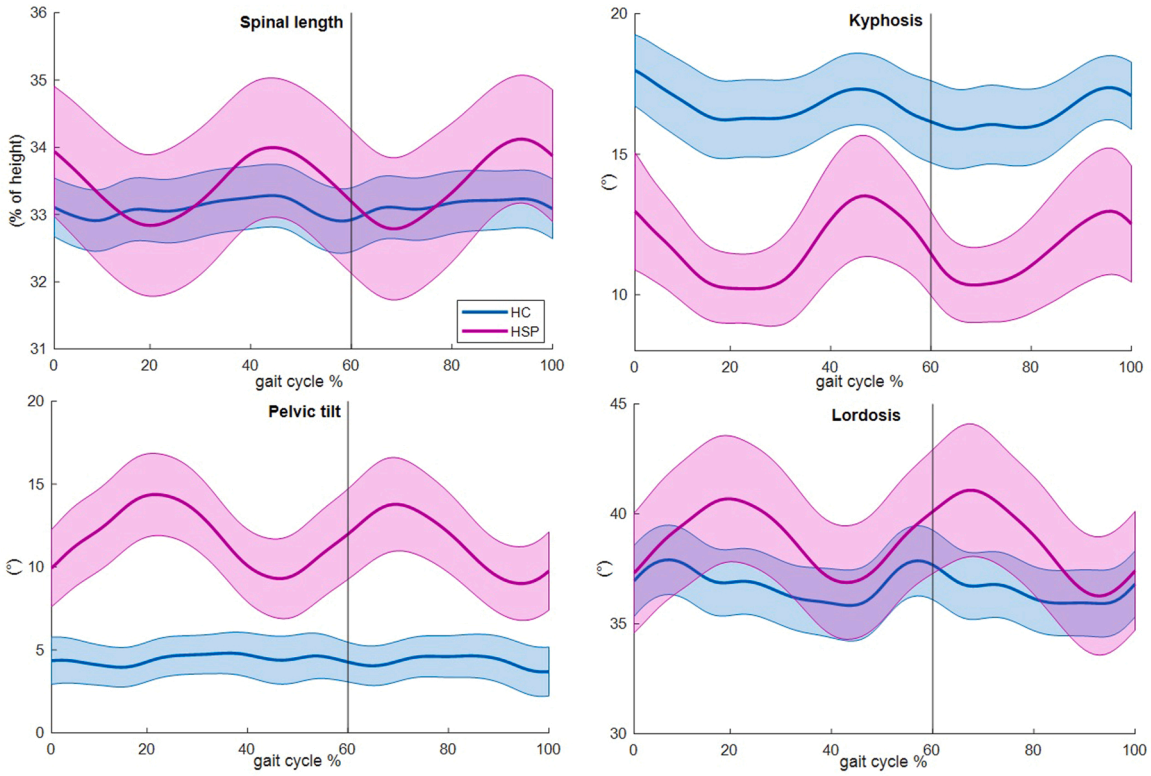

Fig. 2. Average curve (solid line) and standard error (shaded area) of spinal length and kyphosis (upper plots) and of pelvic tilt and lordosis (lower plot) for the two cohorts; in blue the control group (HC), in purple the HSP participants. All curves are plotted against the percentage of the gait cycle.

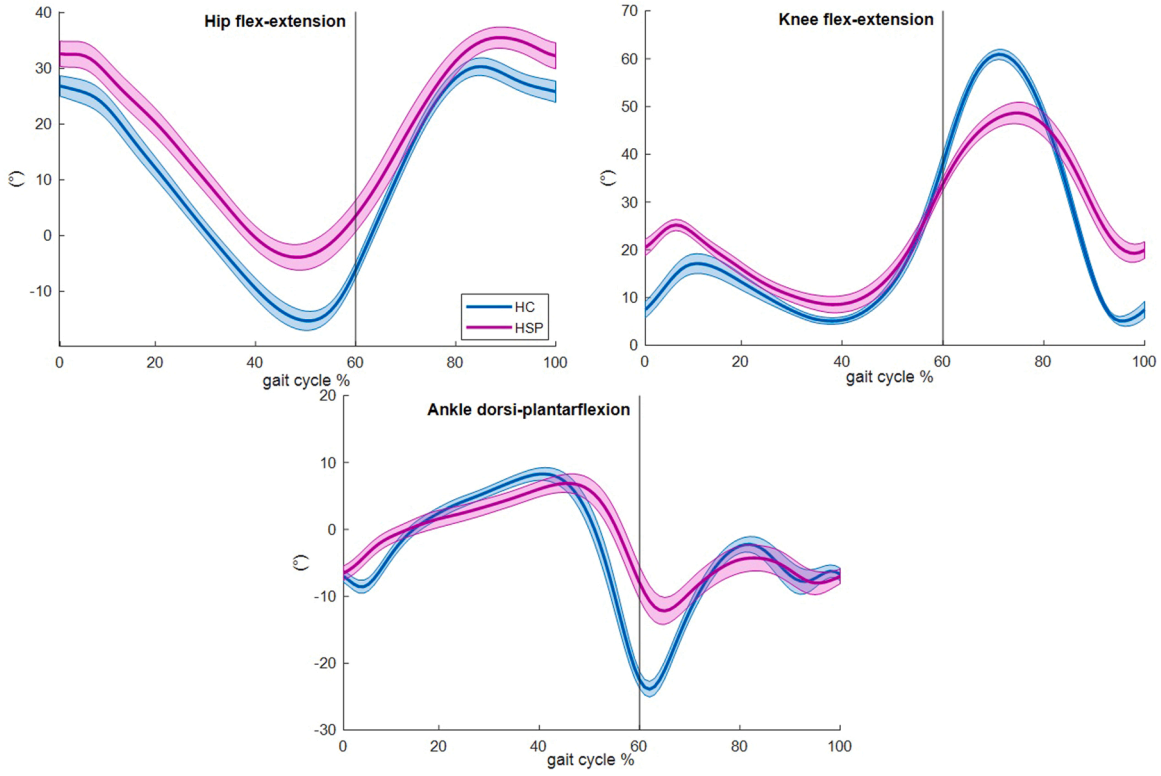

Fig. 3. Average curve (solid line) and standard error (shaded area) of hip and knee flex-extension (upper plots) and of ankle dorsi-plantarflexion (lower plot); in blue the control group (HC), in purple the HSP group. All curves are plotted against the percentage of the gait cycle.

the hip, knee and ankle kinematics were similar to Pulido's pattern IV, characterized by insufficient hip extension and increased knee flexion in stance, followed by decreased and delayed knee flexion in swing, increased ankle dorsiflexion before swing and decreased and delayed plantar flexion. In fact, all these features were present in our recordings but, contrary to Pulido's findings, pelvic tilt was larger in our patients than in the control group. According to Pulido et al., [8], such pattern resembles the "crouch gait" that has been described in CP patients and attributed to weakness of hip, knee and ankle physiological extensor muscles [31,32].

A major limitation of this study might be the small sample size of the HSP group. However, we adopted a numerosity which is comparable to that of other studies addressing the same topic [6,10,11]. Another limit is that the spine was considered as composed of three linear segments (C7 to MAX_KYPH; MAX_KYPH to PSIS_MX; PSIS_MX to coccyx), i.e., the minimum set required to estimate the relevant anatomical curvatures. Clearly, with more markers placed on the spine a better description could have been obtained. Furthermore, this study did not include spino-pelvic radiographic data to support the kinematic results with postural alignment, preventing a direct comparison with the results obtained in healthy adults [12,13] as well as in patients with Cerebral Palsy [33,34]. Finally, we restricted the investigation of lower limb angles to the sagittal plane because we were mainly interested in comparing our results to the seminal classification study of Pulido et al. [8], who identified different patterns basing on pelvis and lower limb kinematics in that plane. We are nonetheless aware that a full 3D evaluation would have allowed a more comprehensive description of gait deficits.

In conclusion, despite the small sample size, we observed that in HSP the alterations in the lower limbs also affect the spine, generating a picture of total body motor impairment. Our results show that it is important to consider the trunk as an articulated system and not to approximate it as a rigid body hinged to the pelvis, a perspective that could be useful also in the treatment of gait abnormalities. Future research should then explore targeted integrated rehabilitation approaches and assistive technologies that address both upper and lower body impairments. Understanding the interplay between upper and lower body dynamics in HSP is imperative for developing comprehensive therapeutic strategies aimed at improving their mobility and quality of life [35].

CRediT authorship contribution statement

V. Farinelli: Writing – review & editing, Writing – original draft, Software, Investigation, Data curation, Conceptualization. **C. Palmisano:** Writing – review & editing, Software, Formal analysis. **C. Dosi:** Writing – review & editing, Resources. **I. Pedrinelli:** Resources. **E. Pagliano:** Resources. **R.Esposti:** Writing – review & editing, Supervision. **P.Cavallari:** Writing – review & editing, Supervision, Funding acquisition, Conceptualization.

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Declaration of Competing Interest

The authors declare the following financial interests/personal relationships which may be considered as potential competing interests: Paolo Cavallari reports financial support was provided by Pierfranco and Luisa Mariani Foundation. All other authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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