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Postural stability during gait for adults with hereditary spastic paraparesis

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ABSTRACT

Individuals with hereditary spastic paraparesis (HSP) are often impaired in their ability to control posture as a result of the neurological and musculoskeletal implications of their condition. This research aimed to assess postural stability during gait in a group of adults with HSP. Ten individuals with HSP and 10 healthy controls underwent computerized gait analysis while walking barefoot along a 10-m track. Two biomechanics methods were used to assess stability: the center of pressure and center of mass separation (COP-COM) method, and the extrapolated center of mass (XCOM) method. Spatiotemporal and kinematic variables were also investigated. The XCOM method identified deficits in mediolateral stability for the HSP group at both heel strike and mid-stance. The group with HSP also had slower walking velocity, lower cadence, more time spent in double stance, larger step widths, and greater lateral trunk flexion than the control group. These results suggest that individuals with HSP adjust characteristics of their gait to minimize the instability arising from their impairments but have residual deficits in mediolateral stability. This may result in an increased risk of falls, particularly in the sideways direction.

1. Introduction

Hereditary spastic paraparesis (HSP; also known as hereditary spastic paraplegia) is a rare genetic disorder characterized by the gradual degeneration of neurons in the spinal cord (Salinas et al., 2008). This neural degradation worsens over time (Bonnefoy-Mazure et al., 2013), with the long neurons which innervate the lower limbs being most severely affected (Salinas et al., 2008). Because the ankle, knee and hip joints are the primary effectors for ensuring postural control, impaired communications between the cortex and the distal musculature may adversely affect postural stability. In individuals with HSP, muscle responses to balance perturbations appear to be delayed as a result of impaired afferent conduction, rather than slowed efferent conduction (Nonnekes et al., 2013). In addition, muscle spasticity (one of the more limiting motor deficits for people with HSP) reduces the elasticity of the muscles and limits their ability to move through a full range of motion. Over time this can result in permanent changes to the musculoskeletal architecture of the individual (Sankar and Mundkur, 2005). Together, the disrupted neural communication and musculoskeletal constraints may impair postural control and stability, resulting in significant challenges during tasks such as walking.

In clinical practice, outcome scales and standing posture measurements have been widely used to assess balance instabilities. Studies that assess static postural stability are often grounded in the model of the human body as an inverted pendulum (Winter et al., 1998), where the center of mass (COM) pivots above the center of pressure (COP; the vector of all ground reaction forces (GRF) (Winter, 2005)) but within the boundaries of the base of support (BOS) (Bruijn et al., 2013). The distance between the COP and the COM during standing is often used to study static balance control (Hass et al., 2004; Mok et al., 2011), however static balance tests do not correlate well with measures of dynamic gait stability and cannot predict falling risks (Owings et al., 2000; Kang and Dingwell, 2006). One possible reason for this discrepancy is that walking is inherently unstable, with the COM residing outside of the boundaries of the BOS for much of the gait cycle (Bruijn et al., 2013). Therefore, the performance of static balance tests may not be a good predictor of dynamic stability. Instead, the COP-COM distance (separation) can be applied to dynamic tasks. The COP-COM has been used to assess dynamic stability among an elderly population during walking and obstacle crossings (Hahn and Chou, 2004), and during gait initiation in a cohort of

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patients with Parkinson's disease (Hass et al., 2005). In both cases, those with greater impairment had lower COP-COM separation.

In a more recent study, Hof developed the extrapolated center of mass (XCOM) method to quantify dynamic stability (Hof et al., 2005). The XCOM method builds upon the concept of the inverted pendulum by considering the velocity of the COM (VCOM) relative to the height of the COM over the BOS, creating a new value known as the XCOM (Hof, 2008). A Margin of Stability (MOS) is quantified based on the distance between the XCOM and the outer boundary of the BOS. A negative MOS value indicates that the XCOM is outside of the BOS, thus the individual will need to make an adjustment, such as taking a step, to prevent a fall. The XCOM has been widely accepted as a method to assess dynamic postural stability in both clinical and healthy populations, including individuals with multiple sclerosis (Peebles et al., 2016), transtibial amputation (Beltran et al., 2014; Curtze et al., 2011), cerebral palsy (Delabastita et al., 2016; Dixon et al., 2016), stroke (Hak et al. 2013a, 2013b) and the elderly (Bierbaum et al., 2011).

This research aimed to compare dynamic postural stability during gait between adults with HSP and a healthy control group. We hypothesized that the dynamic postural instability in HSP can be quantified using COP-COM separation and MOS values. In comparison with the healthy control group, the COP-COM separation and MOS values from the HSP group will be lower.

2. Methods

2.1. Research participants

Ten individuals who had been genetically identified as individuals with HSP and 10 age-matched healthy controls underwent computerized gait analysis while walking barefoot along a 10-meter track. The research was approved by the Northern X Regional Human Research Ethics committee of New Zealand, and all participants gave their written informed consent. The HSP patients were recruited from the Auckland City Hospital Neurogenetics Clinic and the healthy controls were recruited from local communities. All HSP participants were rated as a 1 for the gastrocnemius on the Modified Ashworth Scale (slight increase in muscle tone affecting the end ranges of motion), with all but one affected in both legs. Two participants were also affected in the adductors, knee flexors, and knee extensors (MAS = 1). Participant demographics are displayed in Table 1.

2.2. Data collection

Data collection followed a standard protocol for gait analysis using the Cleveland Clinic marker set (Sutherland, 2002). Reflective spheres (14 mm diameter) were attached using double sided tape to mark the following bilateral positions: the acromioclavicular joint of the shoulder, mid triceps, lateral epicondyle of the elbow, posterior wrist, anterior superior iliac crest, lateral thigh (triad marker), lateral and medial knee, lateral shank (triad marker), lateral and medial malleoli of the ankle, calcaneous, the second metatarsal, and the sacrum (a single marker between the posterior superior iliac crests). Eight high-speed infrared cameras captured the movement of the participants, which was then transformed into a 3D computerized model of the body (Vicon Oxford Metrics, Oxford, UK).

First, a static trial was completed to calibrate the computerized model to each of the markers. For the static trial, the participant stood still for a few seconds while being recorded by the cameras. This allowed the positions of the ankle and knee markers to be determined relative to those markers on the shank and thigh, thus enabling the removal of the ankle and knee markers during subsequent walking trials. Virtual positions of joint centres were recreated from the positions of the remaining markers through a pointer-cluster method (Cappozzo et al., 1996). Participants were then required to walk barefoot at a self-chosen speed along a 10-m walking track. Force plates embedded in the track allowed for the COP location to be obtained from the GRF position data (Bertec Corporation Ohio, USA). The force plates recorded the vertical force and two components of the moment (around ML and AP directions respectively). The centre of pressure was then calculated based on these measures. For a trial to be accepted, force plate data needed to include a single, complete footstep, free from additional GRFs or noise. A minimum of 3 successful trials were obtained for each participant. The sampling rate of 100 Hz was used for the joint position data, and 1000 Hz for the force plate data.

2.3. Data processing

Each reflective marker was manually labelled using Vicon Workstation Software (Oxford Metrics, Oxford, UK). Data filtering was completed using Vicon Nexus 1.7.1 and a 4th order low-pass Butterworth filter with a cut off frequency of 6 Hz. The Cleveland Clinical Model (Sutherland, 2002) was then used to estimate locations of the joint centers and virtual markers. The COM position

Table 1
Participant demographics.

Participant	Sex	Age (years)	Leg Length (cm)	Weight (kg)	MAS
C1	F	62	725	71.8	N/A
C2	F	62	808	75	N/A
C3	F	62	754	61.2	N/A
C4	F	67	811	68	N/A
C5	M	63	772	78.2	N/A
C6	M	25	993	79.3	N/A
C7	M	28	830	63	N/A
C8	M	63	804	73.4	N/A
C9	M	70	791	69.3	N/A
C10	M	62	877	66.1	N/A
HSP1	F	54	676	61.1	1
HSP2	M	54	909	75.2	1
HSP3	F	40	772	67	1
HSP4	F	46	819	90.6	1
HSP5	F	58	799	90.1	1
HSP6	M	62	836	96.6	1
HSP7	M	72	877	106	1
HSP8	M	57	882	88.7	1
HSP9	M	32	808	63.2	1
HSP10	M	60	934	75	1

was calculated using a standard procedure as described by Winter, which locates the whole-body COM based on the COM from each individual body segment (Winter, 2005). Spatiotemporal gait parameters, lateral trunk flexion angle, and pelvic obliquity angle were generated with Vicon Workstation Software, using the kinematic and force data (OxfordMetrics, Oxford, UK).

The COP-COM distance was then determined by subtracting the horizontal position of the COM from the position of the COP in the mediolateral (ML) and the anteroposterior (AP) direction. To cater for left footed and right footed trials, the difference between the COP and COM was made positive for all trials in the ML direction (the COP always remained outside the COM). The average distance was normalized to each participant's leg length, calculated from the static trial as the distance between the left ankle joint center and left hip joint center.

Customized scripts written on Vicon BodyLanguage (Oxford Metrics, Oxford, UK) allowed for the MOS to be calculated using the following equation progression (Hof et al., 2005):

$$\omega_o = \sqrt{\frac{g}{l}} \tag{1}$$

ω_o represents the eigen frequency of the pendulum, calculated as the square root of gravity (9.81 $m \cdot s^{-2}$) divided by the length of the pendulum (the vertical distance between the COP and the COM as calculated from the static trial). The velocity of the COM (VCOM) was then divided by this value to obtain an individualized variable that reflects the individual's walking velocity relative to their height. The $COM_{position}$ refers to the location of the virtual COM marker in the AP or ML direction, based on the global coordinate system of the gait laboratory. Then the XCOM value was calculated by the following equation:

$$XCOM = COM_{position} + \frac{VCOM}{\omega o} \tag{2}$$

The MOS was finally determined as the distance between the XCOM and the border of the BOS (Eq. (3)). The anterior margin of the BOS was identified as the metatarsal marker of the stance foot, and the lateral margin estimated as 2 cm lateral to the second metatarsal marker.

$$MOS = BOS - XCOM \tag{3}$$

The Temporal Margin of Stability (MOST) reflects the length of time before the boundary of the BOS will be reached by the XCOM, based on the current velocity of the COM. This value provides an estimate of how long the individual can stay in their current state before action must be taken to prevent a fall.

$$MOS_T = \frac{MOS}{VCOM} \tag{4}$$

As the VCOM is very small in the ML direction, the MOS_T is approach indefinites during the mid-stance. Therefore, the MOS_T was only calculated in the AP direction. Values were obtained at heel strike and mid-stance for both techniques. Heel strike was identified as the first detectable point of contact with the force plate, and mid-stance was identified as the time where the GRF in the AP direction was equal to zero (i.e. the COM is directly above the COP). Fig. 1 diagrammatically compares the COP-COM method and the XCOM method.

2.4. Statistical analysis

All statistics were calculated using IBM SPSS software. Prior to statistical analysis, all data (spatiotemporal, kinematic, and stability parameters) were tested for normality using the Shapiro-Wilk test. For data that passed the normality test, an independent samples *T*-test was completed. Any non-normally distributed data were tested using the Mann Whitney *U* test. Equality of variances was determined using the Levene's test, and the appropriate *P*-value selected based on these results. A significant difference between groups was classified as a *P*-value equal to or less than 0.05.

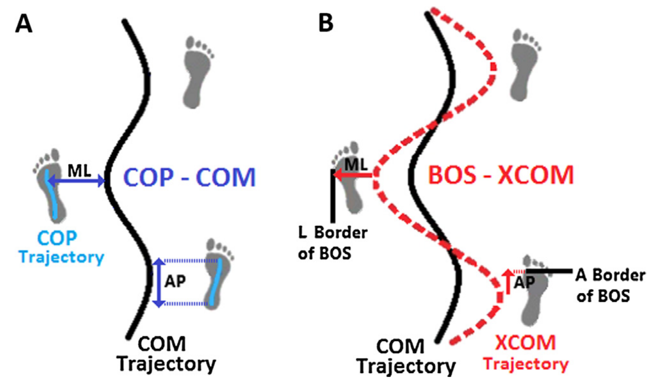

Fig. 1. Diagrammatic comparisons of the COP-COM method (A) and the XCOM method (B) in the mediolateral direction.

3. Results

There was no significant difference between the groups for age (56.4 ± 16.0 years for the control group, 53.5 ± 11.5 years for the HSP group, $p = 0.64$), body weight (70.5 ± 6.12 kg for the control group, 81.4 ± 15.2 kg for the HSP group, $p = 0.06$), and leg length (816.9 ± 74.8 cm for the control group, 831.5 ± 75.1 cm for the HSP group, $p = 0.67$). The mean COP-COM separation for both groups is depicted in Fig. 2. No between-group difference was detected in the AP direction at heel strike (0.305 ± 0.070 for the control group, 0.254 ± 0.070 for the HSP group, $p = 0.110$) or the AP direction at mid-stance (0.009 ± 0.042 for the control group, 0.003 ± 0.050 for the HSP group, $p = 0.738$). There was also no significant difference between the groups for the COP-COM separation in the ML direction at heel strike (0.051 ± 0.033 for the control group, 0.075 ± 0.034 for the HSP group, $p = 0.118$) or mid-stance (0.072 ± 0.029 for the control group, 0.064 ± 0.021 for the HSP group, $p = 0.481$).

The mean MOS values in both directions for each group are displayed in Fig. 3. The MOS in the ML direction was significantly lower for the HSP group at heel strike (−0.039 ± 0.035) than the control group (0.050 ± 0.053, $p = 0.023$); and at mid stance (−0.038 ± 0.037 for the HSP group and 0.055 ± 0.053 for the control group, $p = 0.023$). A statistically significant difference was not

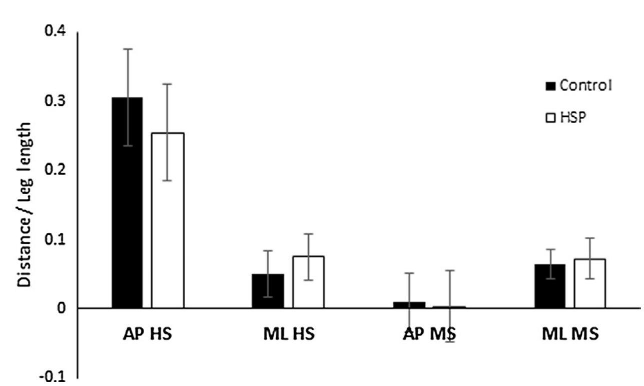

Fig. 2. The mean COP-COM separation relative to leg length. Black bars = Control group; White bars = HSP group; AP = Anteroposterior; ML = Mediolateral; HS = Heel strike; MS = Mid-stance. ***p < 0.001.

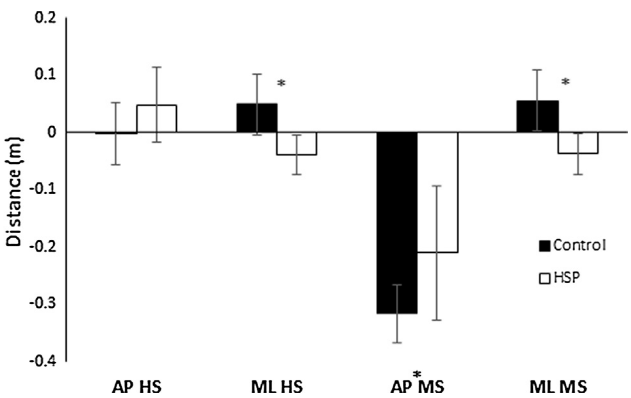

Fig. 3. The mean Margin of Stability obtained using the XCOM method. Black bars = Control group; White bars = HSP group; AP = Anteroposterior; ML = Mediolateral; HS = Heel strike; MS = Mid-stance. ***p < 0.001.

identified between the control group and HSP group for the AP MOS at heel strike (0.049 ± 0.065 for the HSP group, −0.003 ± 0.054 for the control group, $p = 0.072$), but there was a significant difference at mid-stance (−0.210 ± 0.116 for the HSP group, −0.316 ± 0.050 for the control group, $p = 0.039$). There was also a statistically significant difference between the MOS_T in the AP direction at heel strike (Fig. 4). The HSP group took longer to reach the limits of stability (0.068 ± 0.080) than the control group (0.000 ± 0.040, $p = 0.028$). A significant difference was not identified for the MOS_T in the AP direction at mid-stance (−0.214 ± 0.100 HSP group; −0.251 ± 0.039 control group, $p = 0.291$).

Gait characteristics significantly differed between the two groups (Table 2), with the HSP group walking significantly slower (0.95 ± 0.28 versus 1.27 ± 0.16, $p = 0.007$), stepping with a lower cadence (103.60 ± 18.26 versus 123.30 ± 15.48, $p = 0.018$), having a longer step time (0.60 ± 0.12 versus 0.49 ± 0.07, $p = 0.029$), and with more time spent in the double support phase (0.31 ± 0.11 versus 0.17 ± 0.04, $p = 0.002$) than the control group. The HSP group also had a significantly wider step width than the control group (13.80 ± 4.53 versus 7.65 ± 2.56, $p = 0.023$). Step length did not significantly differ between the group with HSP and the controls (55.40 ± 10.54 versus 62.50 ± 4.55, $p = 0.066$), nor did single support time (0.44 ± 0.10 versus 0.41 ± 0.05, $p = 0.353$). A significant difference was identified in the lateral trunk flexion, with the HSP group demonstrating more flexion than the control group (7.15 ± 3.46 versus 2.65 ± 1.37, $p = 0.001$). Pelvic obliquity did not significantly differ between the HSP group and the control group (1.65 ± 0.58 versus 1.76 ± 0.56, $p = 0.679$).

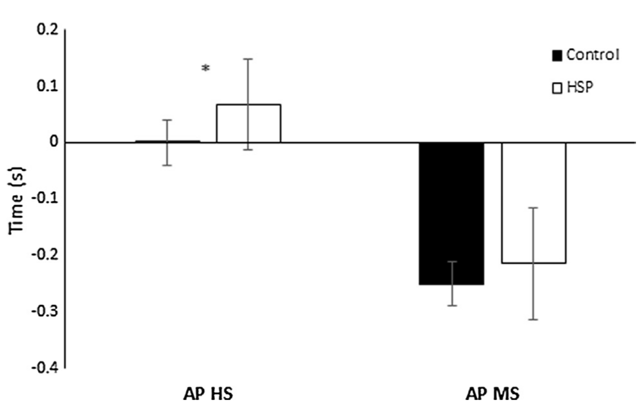

Fig. 4. The mean Temporal Margin of Stability obtained using the XCOM method. Black bars = Control group; White bars = HSP group; AP = Anteroposterior; HS = Heel strike; MS = Mid-stance. *p < 0.05.

4. Discussion

This research aimed to evaluate the postural stability of adults with HSP. Our hypothesis that postural instability for the group with HSP could be quantified was partially confirmed by a significantly lower MOS in the lateral direction for the HSP group. This research is the first to use the XCOM technique among a cohort of participants with HSP, providing novel insight into the control of posture among these individuals.

4.1. The COP-COM separation

No between-group differences were identified in the AP and ML directions at heel strike or mid-stance. These results are surprising given the wider step widths of the HSP group compared to the controls, an adaptation which is common in those with postural control deficits as an attempt to increase stability (Mcandrew Young and Dingwell, 2012). The lack of difference in lateral COP-COM separation may be related to the significantly higher lateral trunk motion identified in the HSP group, as is consistent with other work amongst HSP populations during gait (Adair et al., 2016; Bonnefoy-Mazure et al., 2013). Lateral trunk flexion may be increased to promote the hitching ability of the pelvis, a compensation for the proposed hip abductor weakness in those with HSP (Adair et al., 2016). During gait, the hip abductors are the primary source of control for corrections in ML stability (Winter, 1995). Stability of the pelvis via the hip musculature is vital for postural control as the pelvis forms the base from which the rest of the postural muscles act. An increased lateral trunk flexion would consequently shift the COM outwards towards the foot in stance, 'cancelling out' the increased COP-COM separation occurring from the wider steps. In our results, pelvic obliquity did not significantly differ between the two groups while lateral flexion did, supporting the idea that pelvic stability is prioritized over trunk stability.

4.2. The Margin of stability

In the AP direction, a significant difference in stability was identified between the two cohorts at mid-stance. Typically, the AP MOS is primarily negative during gait, returning to a positive state only briefly during the double stance phase. Comparable stability values between the two groups are likely the result of adjustments made to gait: lower walking speed, lower cadence, longer total stepping time, and more time spent in the double support phase for the HSP group. These adjustments expand the time spent in double support and therefore the time that the XCOM is contained within the boundaries of the BOS, thereby increasing stability.

A significant between-group difference was identified for the average MOS in the ML direction at both heel strike and mid stance, with the values being negative for the group with HSP and positive for the control group. Generally, in the ML direction, stability decreases during mid-stance as the XCOM moves closer to the edge of the BOS, then increases with the change in XCOM direction towards the upcoming stance foot. The lower lateral MOS values in the HSP individuals stand out from those found previously in other clinical populations, where the ML MOS values matched or even exceeded those of healthy controls due to compensations made during gait (Beltran et al., 2014; Curtze et al., 2011; Hak et al., 2013; Peebles et al., 2016; Delabastita et al., 2016). The significant difference identified in the lateral MOS also contradicts the results of the ML COP-COM separation, which did not significantly differ between the two groups. It is likely that the velocity of the COM is responsible for this discrepancy, as this is the major defining feature between the COP-COM and the XCOM techniques (Eq. (2)). During walking, there can be up to a 4 cm dif-

Table 2
Spatiotemporal and kinematic gait data. *Mann Whitney U test.

	Control Mean (SD)	HSP Mean (SD)	p value
Cadence (steps/min)	123.3 (15.48)	103.6 (18.26)	0.018
Speed (m/s)	1.27 (0.16)	0.95 (0.28)	0.007
Step time (s)	0.49 (0.07)	0.6 (0.12)	0.029
Single Support Time (s)	0.41 (0.05)	0.44 (0.1)	0.353*
Double Support Time (s)	0.17 (0.04)	0.31 (0.11)	0.002
Step Length (cm)	62.5 (4.55)	55.4 (10.54)	0.066
Step Width (cm)	7.65 (2.56)	13.8 (4.53)	0.023*
Lateral Trunk Flexion (°)	2.65 (1.37)	7.15 (3.46)	0.001
Pelvic Obliquity (cm)	17.59 (5.63)	16.51 (5.83)	0.679

ference between the location of the COM and XCOM, where the XCOM approaches the border of the COP more closely than that of the COM (Hof et al., 2005) and with a sharper change in trajectory as compared to the smooth trajectory of the COM (Curtze et al., 2011). A higher velocity of the COM may promote the risk of falls (Quach et al., 2011). Any unexpected increases in COM velocity (e.g., stepping on an unstable surface, tripping over an item) require rapid corrective action if the COM velocity is not well controlled. With the increased lateral trunk flexion in the group with HSP, it is likely that the lateral COM displacement approaches the border of the BOS more closely than that of the controls. Any deficits in the ability to hold the COM along a smooth trajectory will be evident while using the XCOM method, thus the increased risk of falls in this cohort are better represented through the XCOM method than that of the COP – COM separation.

4.3. The temporal margin of stability

The larger AP MOS_T for the group with HSP at heel strike was likely due to the slower walking velocity. Because step length did not significantly differ between the two cohorts, a slower forward velocity of the COM may have increased the time before the COM moved beyond the anterior border of the foot. During mid-stance, the AP MOS_T was negative for both groups due to the COM residing in front of the anterior base of support. This indicates an unstable state, where an expansion of the base of support (i.e. another step forward) is required to return to a positive stability margin.

4.4. Implications of these findings

These results suggest the presence of lateral instability in those with HSP despite the compensations made during gait to increase stability. This ML instability may increase the risk of lateral falls, which warrants a cautious approach during tasks where the lateral BOS is reduced, such as standing with a reduced step width, walking with feet in tandem or balancing on one foot, as may be required when navigating through tight spaces or stepping over obstacles. The XCOM technique is useful to identify and stratify those most at risk of falls, aiding in the allocation of resources to those more severely affected.

Very few training studies have been carried out in this cohort, but there is some evidence that therapy results are promising for improving gait parameters. A hydrotherapy rehabilitation program improved walking speed, step length, and kinematic variables in a group of adults with HSP through exaggerations of the compensatory strategies already being used during gait (Zhang et al., 2014). Also, a robotic gait training program improved balance, walking capacity, and quality of life in a group of adults with HSP (Bertolucci et al., 2015). Further research may build upon the results of these studies by using the XCOM method to objectively quantify changes in postural control following a rehabilitation program. Applying the XCOM method in a research setting would provide a means to critically evaluate the changes in postural stability pre- and post-intervention. To inform best clinical practice this information is vital, and could improve the quality of rehabilitation programs provided to individuals affected by HSP.

4.5. Limitations

The technique used in this study retain several of the over-simplified characteristics of the inverted pendulum model. The BOS during the double stance phase is ignored, and instead we assume that the COP alternates instantaneously during heel strike of the forward foot. It also ignores any displacement of the COP within each step (Hof, 2008). Additionally, the task required the walking trial to be completed with little or no assistive aid, limiting the applicability of the technique to individuals who are severely affected by HSP.

5. Conclusion

To our knowledge, this research is the first study to investigate postural stability of adults with HSP. Despite compensations made to increase stability, a lower MOS was also identified in the ML direction for the group with HSP. The application of these two techniques is useful to quantify instability in patients with HSP and has the potential to be highly useful in future clinical research applications and the treatment of those with HSP.

Conflict of interest

We declare that we have no proprietary, financial, professional or other personal interest of any nature or kind in any product, service and/or company that could be construed as influencing the position presented in the manuscript.

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Appendix A. Supplementary material

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jbiomech.2019.03.001.

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