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Kinematic gait deficits at the trunk and pelvis: characteristic features in children with hereditary spastic paraplegia

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ABBREVIATIONS

GPS Gait Profile Score

GVS Gait Variable Scores

HSP Hereditary spastic paraplegia

tGPS Gait Profile Score including trunk kinematics

[Abstract]

AIM To examine the kinematic gait deviations at the trunk and pelvis of children with hereditary spastic paraplegia (HSP).

METHOD This exploratory observational study quantified gait kinematics for the trunk and pelvis from 11 children with HSP using the Gait Profile Score and Gait Variable Scores (GVS), and compared the kinematics to data from children with typical development using a Mann–Whitney *U* test.

RESULTS Children with HSP (median age 11y 4mo, interquartile range 4y 0mo) demonstrated large deviations in the GVS for the trunk and pelvis in the sagittal and coronal planes when compared to the gait patterns of children with typical development (p=0.010–0.020). Specific deviations included increased range of movement for the trunk in the coronal plane and increased excursion of the trunk and pelvis in the sagittal plane. In the transverse plane, children with HSP demonstrated later peaks in posterior pelvic rotation.

INTERPRETATION The kinematic gait deviations identified in this study raise questions about the contribution of muscle weakness in HSP. Further research is warranted to determine contributing factors for gait dysfunction in HSP, especially the relative influence of spasticity and weakness.

What this paper adds

- Important HSP kinematic deficits were identified at the trunk/pelvis.
- Deviations included increased ranges of movement in the sagittal/coronal planes.
- Kinematics for the transverse plane showed little deviation from normal.
- Muscle weakness may contribute to gait patterns in HSP.

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[Text]

Hereditary spastic paraplegia (HSP) is an umbrella term for a group of neurodegenerative conditions that share similar symptoms and aetiology, in particular, mobility limitations and a disturbance in gait.[1] Although there is growing research that describes the gait characteristics of people with HSP,[2–6] the main focus has been on gait deviations of the lower limbs in the sagittal plane of movement.

Gait analysis has traditionally focused on the segments of the lower limb.[7] More recently, the importance of the trunk in the maintenance of a stable, upright position has become evident in the literature.[8] Two previous studies analyzed the trunk kinematics of people with HSP[2,5] – kinematic characteristics included deviations in trunk tilt velocity[5] and phasic deviations at the trunk and pelvis.[2] However, neither study included kinematics for the coronal or transverse planes of movement.

Bonnefoy-Mazure et al.[2] proposed that kinematic trunk deviations identified in the sagittal plane were a result of children with HSP using their trunk to compensate for the

difficulties they experienced in their lower limbs. Repetitive compensatory movements of the trunk can be associated with the development of secondary musculoskeletal issues.[9] In order to understand the contributors to gait dysfunction, develop the most appropriate interventions, and potentially prevent the development of secondary complications it is important to first describe the gait features of children with HSP, including the kinematics of the trunk, in all three planes of movement.[7,8]

Given the progressive nature of HSP and the suggestion that adults with late onset HSP may progress more rapidly than children,[1] sample variations in the current literature in chronological age as well as age at symptom onset could affect the generalizability of results. To our knowledge, no gait studies have reported characteristic coronal or transverse plane trunk movements in children with this condition. It is possible that trunk movements during gait demonstrate unique characteristics in children with HSP, and could be used as a biomarker for HSP.

This study aims to examine the kinematic gait deviations at the trunk and pelvis in all three planes of movement in a sample of children with HSP. Based on clinical observation it was predicted that children with HSP would demonstrate large amplitudes of movement at these segments.

METHOD

Participants

A sample of convenience was recruited through advertisements on the Australian HSP Research Foundation website (http://www.hspersunite.org.au) and via medical and allied health professionals throughout Australia. Eligible participants had a diagnosis of HSP and were aged between 6 and 19 years. Diagnoses were made by qualified medical practitioners involved in the ongoing medical care of the participants: these included paediatric neurologists, paediatricians, and paediatric rehabilitation physicians. Results from genetic testing were available for four participants: one child was confirmed to carry SPG3A; one SPG4; and remained inconclusive for two participants. Aside from those with genetic confirmation, HSP was diagnosed clinically on the basis of progressive childhood spasticity, and in the absence of alternative diagnoses suggested by neuroradiological or metabolic investigations and patient history.

Children needed to be able to walk 10m without assistive devices during the gait assessment. Participants were excluded if they had undergone previous lower limb orthopaedic surgery or botulinum neurotoxin-A injections in the 6-month period preceding

the gait assessment. The study was approved by the associated Human Research Ethics Committees. All participants and their families gave informed consent.

Typically developing reference group

Gait data from the children with HSP were compared to data from a group of 28 children with typical development, a sub-cohort of those described by Baker et al.[10] (median age 10y 11mo, interquartile range [IQR] 4y 3mo). These data had been collected in a clinical gait laboratory using the same protocols and gait model as the HSP cohort. Trunk data were available from 10 of the children with typical development.

Data collection procedure

Gait analysis was performed using a standard set of 20 (13mm) reflective markers, with an additional three trunk markers (placed on the sternal notch of the manubrium and the spinous processes of T2 and T10). Marker placement was based on the protocol by Davis et al.[11] and the model provided by Vicon Plug-In-Gait (PiG) (Vicon Motion Systems, Oxford, UK). Data were collected with 12 MX F20/F40 cameras operating at 120Hz using a Vicon MX system and processed with PiG. The sequence proposed by Baker[12] was utilized to define pelvic kinematics. The orientation of the trunk and pelvis were defined in relation to the laboratory. In order to minimize variation in marker placement, the same physiotherapist (BA) applied all markers. Training in marker placement and data capture was provided by a clinician (JR) with extensive experience in clinical gait analysis before study commencement. Participants walked barefoot over a 10m walkway at their self-selected speed. At least five complete trials were captured for each leg of the participants.

A single representative trial was chosen to calculate the gait kinematics for each participant. This method was used, as opposed to calculating a mean gait trial, because in the presence of inter-trial variability a derived mean may not be representative of how the child actually walked.[13] The representative trial was defined as the first trial with a clean strike on the force plates and minimal marker drop-out. Walking speed and the stance-swing ratio of the representative trial were compared to the other trials of the same participant to ensure the kinematics were a reasonable representation for that child.[14]

Gait variables and outcome measures

The Gait Profile Score (GPS)[10] and Gait Variable Scores (GVS)[10] have been utilized to quantify the magnitude of kinematic deviation across the gait cycle in multiple studies of

neurological disorders,[8,15] including HSP,[3] and were employed in the current study. For this study, additional GVS were calculated for the trunk using the mathematical methods described by Baker et al.[10] and incorporated in the profile score (tGPS). The tGPS and GVS were presented in the Movement Analysis Profile (MAP).

The GVS can help to identify the joints and segments that experience the largest kinematic deviations throughout the gait cycle, nonetheless they do not provide information about the direction and timing of the gait deficits.[10] Therefore, the GPS and GVS were used in conjunction with discrete gait variables, based on those described by Wolf et al.[5] Similar to other studies of people with HSP,[2,4,5] the discrete gait variables described the kinematic peaks and amplitudes of movement in the gait cycle (Appendix SI, online supporting information). To account for the potential for phasic deviations,[2] the timing of kinematic peaks was also assessed.

Because the pelvis and trunk are segments common to the left and right lower limb, the kinematics for the left side were arbitrarily chosen to represent the movement of these segments.

Statistical analysis

Because of the relatively small number of participants, non-parametric statistical methods were employed to describe the distribution of the data throughout this study. Values for discrete kinematic variables from the group of participants with HSP were compared to children with typical development using a Mann–Whitney *U* test. This was an exploratory, observational study designed to examine potential trends in the gait kinematics of children with HSP; consequently, Bonferroni corrections were not applied. To allow more detailed examination of individual kinematic deviations the gait traces for each participant have been provided in Figure S1 (online supporting information).

RESULTS

Fourteen children with HSP responded to invitations for this study. Three children were excluded because of previous lower limb surgery or an inability to complete the gait assessment. One participant had undergone bilateral soft tissue procedures to improve a cavus, varus, and adducted foot position 7 years before participation in this study. This participant was included in the current study because the surgery targeted foot alignment, did not include concomitant calf lengthening, and, based on reports by Stebbins et al.,[16] was predicted to have had minimal impact on the proximal gait kinematics. Data from 11

participants were included in the final analyses (mean age 11y 4mo, IQR 4y 0mo) (Table I). Seven participants were male and four female, and six reported an onset of HSP symptoms before they were 3 years of age. Regardless of the environment, most children did not use assistive devices (Appendix SII, online supporting information).

The values for the tGPS and GVS are provided (Table II) and displayed in the MAP (Fig. 1). The GVS that were not the focus of this study are provided in Appendix SIII (online supporting information). When compared to children with typical development, the participants with HSP exhibited large deviations in their overall gait kinematics as reflected by the tGPS (left tGPS p=0.006, right tGPS p=0.002). With the exception of the variables in the transverse plane, the GVS differed when comparing children with HSP to the group with typical development (p=0.010–0.020).

Between-group differences were found for 8 of the 30 discrete gait variables (Table III). In the sagittal plane, participants with HSP generally demonstrated a larger amplitude of trunk movement (p=0.003) and a larger degree of posterior trunk lean (p=0.009) when compared to children with typical development. The amplitude of pelvic movement was also increased in the sagittal plane (p<0.001), with a larger degree of maximum anterior tilt (p=0.029).

In the coronal plane, participants with HSP demonstrated an increased amplitude of trunk obliquity (p=0.001), large peaks of trunk obliquity during swing phase of the left leg (p=0.011), and delayed maximal pelvic rise in the coronal plane (p=0.002). In the transverse plane of movement, participants with HSP demonstrated a delay in the timing of maximal pelvic rotation (p=0.009).

DISCUSSION

This study is one of the first to examine and describe the three-dimensional movements of the trunk and associated movements of the pelvis in children with HSP. Specific deviations included increased excursion of the trunk and pelvis in the sagittal and coronal planes, with increased posterior trunk lean, anterior pelvic tilt, and larger amplitudes of trunk obliquity during the swing phase.

When the kinematics were considered across the entire gait cycle, the children with HSP demonstrated significant deviations in the movements of the trunk and pelvis in the sagittal and coronal planes. In addition, the between-group differences in tGPS were found to be larger than the minimum clinically important difference of 1.6° previously reported for the GPS.[17] Contrary to our hypotheses, although children with HSP demonstrated small

kinematic gait deviations in the transverse plane, these deficits did not differ from the data from the group of children with typical development.

Inspection of the results for the discrete variables and the kinematic gait traces of individuals suggested that some participants were asymmetrical in their presentation. For example, approximately half of the participants demonstrated greater trunk obliquity when the left leg was in swing phase compared to the right leg. In clinical scenarios it is rare for people to be completely symmetrical, therefore it is likely that this gait asymmetry was a chance occurrence rather than a characteristic feature of HSP gait. Although hip kinematics in the transverse plane were not specifically analyzed in this study, examination of individual gait traces identified that over half of the participants presented with asymmetric rotational profiles of their hips, with a tendency for more internal rotation of their right hip than their left, six of whom also demonstrated asymmetric trunk obliquity. Internal hip rotation (sometimes related to hip dysplasia) can be associated with lever arm dysfunction of the hip abductors, resulting in increased trunk obliquity to compensate for decreased hip abduction in swing.[18] The purpose of the current study was to describe kinematic deviations, therefore radiological results were not collected. While the clinical histories and brief physical examinations performed during this study did not suggest significant hip dysplasia, further formal investigation may be warranted to examine the incidence of hip dysplasia in children with this condition.

The children with HSP had a tendency to lean more posteriorly than children with typical development, and demonstrated greater amplitudes of trunk movement in the sagittal plane. Notably, 7 of the 11 children demonstrated a ‘double-bump’ trunk pattern with large peaks of movement occurring twice throughout the gait cycle. This finding is consistent with the theory proposed by Bonnefoy-Mazure et al.[2] that children with HSP use their trunk to compensate for lower limb deficits. The group with HSP also demonstrated larger amplitudes of anterior pelvic tilt than children with typical development. This could be explained by the increased lumbar lordosis commonly reported as a feature of HSP.[1] In addition, weak hip extensors and hamstrings have been reported in several papers, which could exacerbate a hyperlordotic posture.[1,19,20]

Previous studies of HSP gait have often chosen to compare gait data from people with HSP to patterns observed in people with cerebral palsy.[2,4–6] While it is recognized that similarities exist between the gait characteristics of people with these two conditions, comparisons of data with other known gait patterns may enable a better understanding of the potential contributing factors for gait dysfunction in HSP. Graham[21] described a sagittal plane

gait pattern, labelled 'hip extensor avoidance gait', in people with spina bifida, whereby people with an L4-level lesion rely on increased flexion of the joints of the lower limbs, an anteriorly tilted pelvis, and a posterior trunk lean to help compensate for weakness of the hip extensors and dorsiflexors during gait. Some of the features of the 'hip extensor avoidance gait' pattern, particularly those described for the trunk and pelvis, were also evident in the gait patterns demonstrated by children with HSP in the current study. These similarities raise the possibility that weakness, as well as spasticity, may contribute to gait dysfunction in HSP, which warrants further investigation.

The data for the coronal plane of movement supported the hypotheses generated for this study, and although the trunk movement patterns of the group with HSP were similar to children with typical development, the amplitudes of movement were often significantly larger. Marsden et al.[20] assessed muscle strength in adults with HSP and found generalized weakness of the flexors and extensors of the lower limb. Although the hip abductors were not assessed by Marsden et al.,[20] the propensity for weakness in the HSP population suggests that other muscle groups could also be affected. Strategies such as excessive lateral trunk flexion or recruitment of other muscles, such as latissimus dorsi, are sometimes used to compensate for weak hip abductors.[22] Excessive lateral trunk flexion has also been described in the gait patterns of adults with osteoarthritis, as a means to hitch the contralateral side of the pelvis and compensate for a pelvic drop during swing.[23] Inspection of the kinematic data in the current study suggested that children with more trunk movement in the coronal plane had relatively little movement at the pelvis, while three children exhibited larger amplitudes of pelvic obliquity with relatively smaller amplitudes of trunk obliquity. It is possible that children with HSP utilized lateral trunk flexion in an attempt to compensate for weak hip abductors and assist with limb clearance in the swing phase, similar to the 'hip abductor avoidance gait' pattern described in children with spina bifida.[21]

Increased trunk obliquity and a 'hip abductor avoidance' gait pattern have been associated with increased loading on the joints of the lower limb, especially the knee joint, in people with spina bifida.[21,24] Previous studies have shown that the use of assistive devices may help to decrease trunk obliquity and excessive joint loading.[25] Based on the kinematic deviations identified in the current study, children with HSP who exhibit excessive lateral and posterior trunk movement may also find that the use of mobility devices could help conserve energy and prevent future joint deterioration.

This study has certain limitations that affect the generalizability of the results, including the small sample size and wide age range of participants with HSP. Similar small

sample sizes and variations in age range have been demonstrated by previous influential gait studies of children with cerebral palsy[7,8] as well as the majority of gait studies of children with HSP.[2,3,6] One previous study with a large sample (n=29) of people with HSP[5] included children as well as adults (age range 5–63y) which limits the generalizability of the results specifically to children. Because of the relatively low prevalence of the condition it is likely that these limitations are inherent in studies of this population.

In order to identify true trunk and pelvic gait kinematics in our study it was necessary for children to complete the gait assessment without an assistive device. This particular criterion also meant that children who were more functionally limited were not included in the sample. It is therefore possible that more severely involved children with HSP may demonstrate different kinematic characteristics compared to those identified in this study. The kinematic deficits demonstrated in this study suggest that muscle weakness could be an additional contributing factor to gait deviations in children with HSP. Nevertheless, muscle strength was not formally assessed, limiting the conclusions that can be drawn about its contribution to gait dysfunction in the current study. The comparison of grouped data may have resulted in some 'wash out' of individual gait deviations, therefore we have provided individual data (available in Figure S1, online supporting information).

According to the results from this study, some children with HSP demonstrate kinematic gait deficits at the trunk and pelvis, and in particular, large amplitudes of movement, especially in the sagittal and coronal planes. The previous intervention literature focused predominantly on the management of muscle spasticity in the HSP population, with few recommendations about muscle weakness. Although muscle spasticity is a key feature of HSP, some of the kinematic deficits identified in the current study support the idea that weakness could be another important contributing factor to gait dysfunction in children with this condition. In order to develop the most appropriate treatment strategies for children with HSP, further research is warranted to identify potential contributing factors and examine their relationship with gait in this population.

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accommodation costs to allow some interstate participants to attend a gait assessment. The HSP Research Foundation was not involved in study design, data collection, the analysis of results, or the manuscript drafting process.

Conflict of interest

This study forms a component of the research conducted as part of Brooke Adair's PhD. Dr Jennifer McGinley and Professor Kerr Graham were involved in the development of the GPS. No further conflicts of interest exist for this study.

SUPPORTING INFORMATION

The following additional material may be found online:

Figure S1: Individual kinematic gait traces.

Appendix SI: Discrete angular and temporal gait variables assessed throughout study.

Appendix SII: Distribution of functional mobility ratings for children with HSP (*n*=11), based on the number of children assigned each rating.

Appendix SIII: Between-group comparison of the GVS which were not the focus of this study.

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Table I: Demographic characteristics of participants with hereditary spastic paraplegia

	Characteristics	***n***	Median (IQR)	Range
Sex	Male	7		
	Female	4		
Age (y)			11y 4mo (4y 0mo)	7y 2mo–18y 7mo
Height (m)			1.55 (0.27)	1.27–1.89
Weight (kg)			46.5 (35.0)	25.0–84.5
Gait speed (m/s)			1.32 (0.13)	1.04–1.42
Family history[a]		9		
Symptom onset	<3y	6		
	3–10y	4		
	>10y	1		

[a]Indicates the number of participants with other family members with known gait dysfunction. IQR, interquartile range; gait speed, average velocity for both legs of the participant during three-dimensional gait analysis symptom onset, age when symptoms were first noticed according to parental report.

Table II: Between-group comparison of the Gait Profile Score that included trunk kinematics and Gait Variable Score for participants with hereditary spastic paraplegia and typically developing children

	Side	HSP (*n*=11)					TD (*n*=8)[a]					Between-group difference (*p*-value)
		Median	Range		Q1	Q3	Median	Range		Q1	Q3	
			Min	Max				Min	Max			
tGPS	L	7.11	5.18	10.21	6.59	9.70	4.64	2.90	8.44	4.08	6.21	**0.006**
	R	7.14	5.85	9.72	6.23	9.23	4.69	2.76	9.23	4.22	5.70	**0.002**
GVS												
Trunk tilt (sagittal)	L	7.13	1.35	15.75	5.76	11.92	4.71	2.02	7.77	3.43	5.56	**0.020**
Pelvic tilt	L	7.34	3.56	17.56	4.11	8.52	4.03	0.64	14.19	2.15	5.64	**0.018**
Trunk obliquity (coronal)	L	3.38	1.08	7.72	2.28	4.27	1.22	0.81	4.26	1.02	2.58	**0.014**
Pelvic obliquity	L	2.19	1.52	4.49	1.78	2.96	1.70	0.49	5.63	1.16	2.11	**0.010**
Trunk rotation	L	3.94	2.07	9.35	2.80	5.77	3.13	0.60	8.84	1.63	6.51	0.360
Pelvic rotation	L	3.30	1.26	8.48	2.21	4.34	3.64	1.73	7.73	2.62	4.56	0.382

[a]Trunk data from the reference group were collected from 10 children rather than 28. HSP, hereditary spastic paraplegia; TD, children with typical development; Min, minimum; Max, maximum; Q1, first quartile; Q3, third quartile; tGPS, GPS that included trunk kinematics; GVS, Gait Variable Score; L, left; R, right.

Table III: Distribution of discrete gait variables for the trunk and pelvis

		HSP (*n*=11)			TD (*n*=28)[a]		
		Median	Range		Median	Range	
			Min	Max		Min	Max
Trunk tilt (sagittal)[b]							
Max tilt	L	10.05	1.73	23.38	10.99	6.65	20.16
Timing max tilt[c]	L	47	0	98	38	0	90
Min tilt	**L**	**0.12**	**-9.97**	**20.44**	**7.05**	**2.59**	**17.34**
Timing min tilt[c]	L	65	14	74	57	8	100
ROM sagittal plane	**L**	**9.55**	**2.94**	**16.62**	**3.43**	**2.82**	**7.18**
Trunk obliquity (coronal)							
Max down[d]	L	4.05	2.24	10.19	2.49	-0.26	7.18
Timing max down[c]	L	20	13	47	39	12	48
Max up[e]	**L**	**7.26**	**1.15**	**15.34**	**1.98**	**-2.23**	**4.35**
Timing max up[c]	L	69	61	98	76	0	95
ROM coronal plane	**L**	**10.51**	**5.16**	**23.91**	**4.12**	**2.97**	**11.45**
Trunk rotation (transverse)[f]							
Max rotation	L	6.88	1.88	19.20	4.56	-1.01	12.33
Timing max rotation[c]	L	38	27	100	40	24	57
Min rotation	L	-6.10	3.61	-11.27	-3.90	4.06	-9.76
Timing min rotation[c]	L	77	0	100	88	0	100
ROM transverse plane	L	12.22	3.27	22.69	8.66	4.21	11.95
Pelvic tilt (sagittal)[g]							
Max tilt	**L**	**21.51**	**7.72**	**35.12**	**14.94**	**0.43**	**25.27**
Timing max tilt[c]	L	30	0	100	81	0	100
Min tilt	L	14.75	2.23	24.00	11.32	-3.63	22.36
Timing min tilt[c]	L	47	17	94	54	0	100
ROM sagittal plane	**L**	**5.14**	**4.03**	**11.26**	**3.37**	**1.71**	**7.70**
Pelvic obliquity (coronal)							
Max drop	L	3.62	1.97	5.73	4.20	1.13	9.65
Timing max drop[c]	L	64	0	100	64	30	70
Max rise[h]	L	4.59	-0.38	8.66	4.09	-2.68	8.43
Timing max rise[c]	**L**	**48**	**14**	**87**	**14**	**10**	**100**

ROM coronal plane	L	7.90	3.20	14.40	8.21	3.75	14.11
Pelvic rotation (transverse)[i]							
Max rotation	L	10.45	5.12	12.70	6.47	1.74	16.31
Timing max rotation[c]	L	16	0	100	5	0	100
Min rotation	L	-7.00	-3.59	-14.61	-8.02	-0.55	-17.47
Timing min rotation[c]	**L**	**56**	**48**	**88**	**50**	**14**	**73**
ROM transverse plane	L	16.75	9.74	27.32	15.27	4.31	30.68

Bold indicates variables that demonstrated statistically significant between-group differences. [a]Trunk data from the reference group were collected from 10 children rather than 28. [b]Positive values indicate the trunk is anteriorly tilted, negative values indicate posterior tilt. [c]Timing indicates the point (%) in the gait cycle where the peak occurs when normalized over 100%. The timing in the gait cycle has been rounded to the nearest whole percentage. [d]Down indicates the left side of the trunk is laterally flexing towards the left limb; negative values indicate the left side of the trunk is in an upwards position i.e. laterally flexing towards the right side. [e]Up indicates the left side of the trunk is laterally flexing towards the right; negative values indicate the left side of the trunk is in a downwards position i.e. leaning towards the left side. [f]Positive values indicate the trunk is anteriorly rotated on the left side, negative values indicate the trunk is posteriorly rotated. [g]Positive values represent an anterior tilt of the pelvis, negative values indicate posterior pelvic tilt. [h]Negative values indicate the left side of the pelvis is dropped. [i]Positive values indicate anterior rotation of the left side of the pelvis, negative values indicate posterior rotation. HSP, hereditary spastic paraplegia; TD, children with typical development; Min, minimum; Max, maximum; L, left; ROM, range of movement.

Figure 1: The Movement Analysis Profile for the group of children with hereditary spastic paraplegia (HSP). The top of the shaded bars indicate the median score for each gait variable and the error bars show the interquartile range for that variable for the group of children with HSP. According to the convention described by Baker et al.,[10] the top of the black solid bars indicate the mean variable score for the reference group of typically developing children. *Gait Variable Scores that were found to be significantly different ($p<0.05$) when comparing children with HSP to typically developing peers. TT, trunk tilt (sagittal plane); PT, pelvic tilt; HF, hip flexion; KF, knee flexion; ADF, ankle dorsiflexion; TL, trunk obliquity (coronal plane); PO, pelvic obliquity; HA, hip abduction; TR, trunk rotation (transverse plane); PR, pelvic rotation; HR, hip rotation; FP, foot progression; tGPS, Gait Profile Score that includes trunk kinematics.

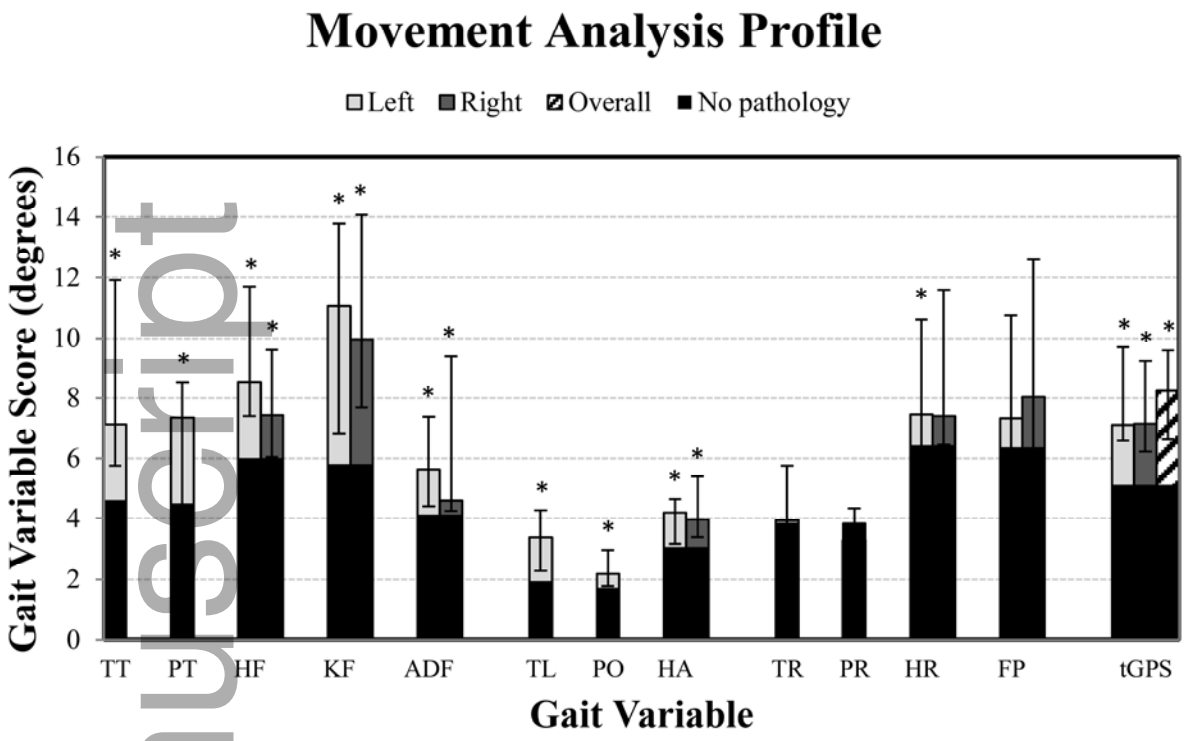

Figure 1: The Movement Analysis Profile (MAP) for the group of children with HSP.