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Faculteit Revalidatiewetenschappen

master in de revalidatiewetenschappen en de kinesitherapie

Masterthesis

Multisystem approach for balance problems in children with cerebral palsy: a cross-sectional case-control study

Jens Cuypers
Dennis Hertoghe

Scriptie ingediend tot het behalen van de graad van master in de revalidatiewetenschappen en de kinesitherapie, afstudeerrichting revalidatiewetenschappen en kinesitherapie bij musculoskeletale aandoeningen

PROMOTOR :

Prof. dr. Pieter MEYNS

BEGELEIDER :

Mevrouw Nina JACOBS



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Context

This duo thesis falls within the domain of 'gait & balance'. It is part of an ongoing PhD project led by Mrs. N. Jacobs which is titled: 'Understanding balance control in children with cerebral palsy on central and peripheral level: a synergistic approach using neuromechanics, brain imaging and functional assessment' (Project code: 92836). This project was started in November 2021, and approved by the Committee for Medical Ethics (CME) of Antwerp University Hospital (UZA)/ University of Antwerp (UAntwerpen), CME of Hasselt University (UHasselt) and the Ethics Committee Research UZ/ KU Leuven (B3002021000145). Funding for the project was provided by the Fonds Wetenschappelijk Onderzoek - Vlaanderen (FWO).

The goal of the overall project is to create insight into the central and peripheral determinants of balance problems in children with CP (CPc). This thesis can be seen as the starting point for further analysis of the effects of proprioception, muscle fatigue, and brain injury characteristics. The aim of this study is to compare balance performance with respect to Horak's multisystem framework for postural control between CPc and typically developing children (TDC). Furthermore, this study elaborates on the balance control of these children during the alternate stepping task using 3D movement analysis. The following research questions were formulated together with our attendant (Mrs. N. Jacobs):

1. Is there a difference in balance performance with respect to the different postural control systems between children with cerebral palsy and typically developing children?
2. Do children with cerebral palsy show different balance control during a stepping task in comparison to typically developing children?

This study was conducted in three movement analysis laboratories, the M²OCEAN-lab (UAntwerpen, UZA-campus Wilrijk), CMAL-lab (KULeuven, UZ Leuven campus Pellenberg), and the GRAIL (UHasselt, campus Diepenbeek). A Vicon system was used for the 3D movement analysis in these labs. Mrs. N. Jacobs performed all data collection and processing (using Vicon Nexus Software (v2.12.1 Vicon Inc.) and MATLAB (2022ab, Mathworks)). One of the authors of this thesis, Cuypers Jens, frequently assisted with assessments and actively helped with recruitment. The introduction and methodology were drafted by Cuypers Jens and further processed by Hertoghe Dennis. The results, discussion, conclusion, and abstract were written

together. Cuypers Jens mainly performed statistical analysis and the creation of figures and tables. Mrs. N. Jacobs provided written feedback on the first draft of the introduction and methods section, afterward she and Prof. Dr. Pieter Meyns also gave written feedback on the entire thesis. In total, six meetings were held together with Mrs. N. Jacobs to give oral feedback on the introduction, discussion, and statistical analyses

Abstract

Background: Balance control is crucial for developing motor skills necessary for all activities of daily life (ADL). Balance deficits are key aspects in the definition of cerebral palsy (CP). These deficits may result from impairments in one or more postural control systems. However, previous studies did not address all these underlying systems and balance control during tasks requiring anticipatory balance is only poorly understood.

Objectives: Firstly, this study aims to clinically investigate balance performance with respect to underlying postural control systems between children with CP (CPc) and typically developing children (TDc). The second objective is to further unravel dynamic balance control, on a biomechanical level, during an anticipatory stepping task.

Methods: In CPc and TDc, balance performance was evaluated using the Kids-Balance Evaluation Systems Test (Kids-BESTest)-extended version. During stepping, dynamic balance control was assessed by the Margin of stability in the anterior-posterior direction (MoS AP) using 3D movement analysis.

Results: 15 CPc (8.74 ± 0.42 ; boys/girls=7/8) and 21 TDc (8.33 ± 0.35 ; boys/girls=11/10) were included. CPc performed significantly worse than TDc on the Kids-BESTest ($z=-5.04$; $p<.0001$). During stepping, CPc exhibited more signs of instability ($z=-5.10$; $p<.0001$) and a longer performance time ($F=12.11$; $p=.0034$). Mean MoS AP was smaller ($F(1,56.2)=18.36$; $p<.0001$) and the variance of MoS AP was higher than TDc ($F(1,58.1)=24.17$; $p<.0001$).

Conclusion: The findings of this study suggest that CPc show deficits in all systems of postural control and that during stepping they exhibit less stable performances and a higher step variability compared to TDc.

Keywords: Cerebral Palsy, Balance, Margin of Stability, Kids-BESTEST

1. Introduction

Balance control is crucial for the development of motor skills that are necessary for all activities of daily life (ADL), such as walking, running, swimming, and ball games. Balance control is the ability to regulate the position of the body in space, with the main purposes being stability and orientation (Shumway-Cook & Woollacott, 2017; Dewar et al., 2017). To ensure balance during these tasks, the center of mass (CoM) needs to be controlled within the base of support (BoS) (Shumway-Cook & Woollacott, 2017). The process of controlling the COM (i.e., balance control) is complex as it depends on multiple sensorimotor mechanisms. In the multisystem framework, Horak et al. (2006) described six systems/resources required for balance control, depending on the type of postural control task (Horak, 2006; Horak et al., 2009). Firstly, there are biomechanical constraints that can have an impact on balance control. These can be seen as 'requirements' that are needed for good balance control (e.g., lower limb strength, range of motion, ...) and will therefore influence the control of the CoM within the BoS. Secondly, a person uses several movement strategies to react to external (Reactive Postural Adjustments or RPA) and internal perturbations (Anticipatory Postural Adjustments or APA). Examples of RPA are the ankle and hip strategy for brief, in-place perturbations, and the stepping strategy for when the CoM falls outside of the BoS. APA are elicited when, for example, someone tries to take a step. The postural muscles in the trunk and standing leg will activate in advance of the stepping leg, providing stability to complete the task. Thirdly, sensory strategies play a crucial role in understanding complex sensory environments. Depending on the sensory context, a person needs to quickly re-weight somatosensory, visual, and vestibular information. This can be demonstrated when a person needs to maintain postural control in a dark room, where he will need to rely more on somatosensory and vestibular information, yet less on visual information. Fourthly, orientation in space is important. This is the orientation of the body with regard to gravity, the support surface, and the internal references of the person. Fifthly, there is the control of dynamics, which is the complex control of a moving CoM during gait and changing postures. Finally, cognitive processing is required for every balance task (Horak, 2006; Woollacott & Shumway-Cook, 2002). These systems and their development are self-evident for healthy children and adults. However, in children with cerebral palsy (CPc), a lesion in the developing brain induces negative effects down the chain of musculoskeletal and neural components

underlying postural control (Dewar et al., 2015). Hence, it is not surprising that balance deficits are key aspects in the definition of cerebral palsy (CP) (Rosenbaum et al., 2007). This severely affects their functional abilities, such as sitting, standing, and walking, and often results in falls (Morgan & McGinley, 2018; Gilson et al., 2014). These balance deficits/problems are therefore one of the most frequent requests for help in physiotherapeutic clinical practice (Dewar et al., 2017).

CP is the most common movement disability in children (Christensen et al., 2014), of which spastic CP is the most prevalent common clinical phenotype occurring in 53-85% (Patel et al., 2020; Perra, 2021). It is established that CPc differ from typically developing children (TDc) in maintaining balance in sensory-challenging environments. For example, they use a vision-steered postural control strategy, with balance problems increasing in the absence of visual input (Pavão et al., 2013; Özal et al., 2022). Furthermore, CPc appear to have smaller anterior limits of stability, which limits their balance and the reach distance of their arm movements in a standing position (Ledebt & Savelsbergh, 2014). As such, like other developmental disorders (e.g., DCD) (Verbecque et al., 2021), it can be hypothesized that CPc show task-specific postural control/balance deficits, situated across all the different postural control systems. However, in the current studies, balance deficits were not evaluated by addressing these underlying systems. Also, results remain conflicting and heterogeneous, which makes postural control in CP only poorly understood. To fill in this gap, this study aims to clinically investigate balance performance with respect to the underlying postural control systems between CPc and TDc.

Moreover, In tasks requiring precise anticipatory control, CPc perform poorer compared to TDc. These children display poorly modulated APA, characterized by less muscle activation in the preparatory/APA phase of movements (Bigongiari et al., 2011; Tomita et al., 2013). As such, they might be limited in their ability to control COM during tasks and, therefore, exhibit poor task performance. However, previous studies solely focussed on the preparatory APA phase of bilateral arm flexion and sit-to-stand tasks (Bigongiari et al., 2011; Pavão et al., 2015). It is therefore insufficiently known how this further affects dynamic balance control in more challenging tasks, like stepping. Therefore, the second objective is to further unravel dynamic balance control on a biomechanical level during an anticipatory stepping task.

2. Method

2.1. Experimental design

This study was conducted in three movement analysis laboratories, the M²OCEAN-lab (UAntwerpen, UZA-campus Wilrijk), CMAL-lab (KU Leuven, UZ Leuven campus Pellenberg), and the GRAIL (UHasselt, campus Diepenbeek). The lab was chosen depending on the region of recruitment. The study has a cross-sectional case (CPc)-control (TDc) design. Children participated voluntarily and after written informed consent from the parents. This study was started in November 2021, and approved by the Committee for Medical Ethics (CME) of Antwerp University Hospital (UZA)/ University of Antwerp (UAntwerpen), CME of Hasselt University (UHasselt) and the Ethics Committee Research UZ/ KU Leuven (B3002021000145).

2.2. Participants

CPc and TDc, aged between five years and zero months until 12 years and 11 months, were included if they met the predefined eligibility criteria/selection criteria. TD children were included when they (1) were born at term (>37 weeks of gestation), (2) had an Intelligence Quotient (IQ) greater or equal to 70, and were cognitively capable of understanding and participating in assessment procedures. They were excluded (1) in case of a diagnosis of a neuromotor or neurodevelopmental disorder (e.g., developmental coordination disorder (DCD), Autism Spectrum Disorder (ASD), or Attention Deficit Hyperactivity Disorder (ADHD)), (2) uncorrected visual or vestibular impairments, or (3) any other neurological, orthopedic, intellectual (IQ<70) or medical conditions potentially impeding balance control. A global questionnaire was used, in which parents could evaluate if a condition was present (e.g., intellectual deficits, since no specific test was used). The Strengths and Difficulties Questionnaire (SDQ full version) for parents was used to assess the behavior at school and home (e.g., to identify potential behavioral problems/disorders if not (yet) diagnosed). These children were recruited from the researcher's (and master student's) lab environment, elementary schools (Vrije Basisschool Lutselus, Diepenbeek, Belgium), and youth organizations through acquaintances and social media in the regions of Hasselt, Antwerp, and Leuven.

CPc were included if they had a diagnosis of (1) spastic (2) uni- or bilateral CP in accordance with The Surveillance for Cerebral Palsy in Europe classification (SCPE). (3) Magnetic

Resonance Imaging (MRI) was necessary to confirm the diagnosis. (4) The children had minimal understanding of verbal instructions and (5) were classified as level I or II on the Gross Motor Function Classification System (GMFCS) (i.e., children who can walk and climb stairs with(out) aid, they can run and jump, but speed, balance, and coordination are limited (Level I) or they are only limited in running and jumping (Level II)). Children with GMFCS level III were also allowed to participate if they were able to walk independently for at least six meters with(out) a walking aid and to stand independently for at least one minute. Children were excluded from the study if they had (1) ataxic or dystonic CP, (2) a GMFCS IV-V classification, or if they could not walk 6 meters independently, (3) a severe intellectual disadvantage (as reported by the parents using a global questionnaire and the SDQ), (4) severe musculoskeletal deformities, (5) upper or lower extremity surgery in the past year, and (6) botulin toxin injections in the past 12 weeks. These children were recruited in the regions of Hasselt, Antwerp, and Leuven and more specifically via the CP referral centers of Leuven (CP-care program) and Antwerp (CePRA), rehabilitation centers, regular or specialized schools (e.g., Sint-Gerardus, MFC Mytyl, Heder, and Pulderbos), private practices (e.g., Vossenbergt1team), acquaintances and through social media.

2.3. Balance Evaluation Systems Test for Children (Kids-BESTest), extended version

Balance performance was assessed using an extended version of the original full Kids-BESTest (Dewar, Claus, Tucker, Ware, et al., 2017; Dewar et al., 2019). The Kids-BESTest is a comprehensive criterion-referenced test to identify and classify balance deficits across six domains (36 items) according to the multisystem framework defined by Horak: (1) biomechanical constraints (5 items), (2) limits of stability and verticality (5 items), (3) transitions and anticipatory postural adjustments (5 items), (4) reactive postural responses (5 items), (5) sensory orientation (5 items) and (6) stability in gait (7 items). Every item is scored from zero (worst performance) to three (best performance) on a four-point rating scale, using age- and task-specific scoring criteria. The item scores are added up to achieve a score for each domain (6 domain scores) and a maximum total score of 108 points across all domains. All scores were normalized ($((\text{achieved score}/\text{total score}) * 100)$) to ensure comparability across all five age bands (i.e., age 5, age 6, age 7, age 8-10, age 11-14). The kids-BESTest is found to have good intrarater, interrater, and test-retest reliability in TDc and CPc (ages 7-18) (Johnson et al., 2023). Concurrent validity has been tested for individual items (e.g., Forward Reach

Test, Lateral Reach Test, (Modified) Clinical Test of Sensory Interaction in Balance) and is shown to be poor compared with center of pressure measures in CPc and TDc (Dewar, Claus, Tucker, Ware et al., 2017; Dewar et al., 2019; Dewar et al., 2021; Dewar et al., 2022; in Johnson et al., 2023).

2.4. Anticipatory balance

To assess anticipatory balance when stepping to a target, CPc and TDc performed a preselected task of the Kids-BESTest: i.e., alternate stair touches as fast as possible until four successful touches for each foot. Children performed the alternate stair touch task barefoot, on footprints (shoulder width apart), with their hands on their hips. They were instructed to not touch the staircase without transferring their weight (height: 15cm). The children were, however, allowed to look at their feet during the task and if they took their hands off their hips, a cue was given to put them back.

All CPc and TDc had an instrumented 3D movement analysis during the stepping task. This 3D movement analysis was conducted using laboratory-based optoelectronic motion capture (Vicon Motion Systems Inc, Oxford Metrics), comprising 10 high-speed (100Hz) infrared cameras. Participants wore the Plug-In Gait (PIG) full-body marker model, using 14-mm reflective markers. (RFHD: right forehead, LFHD: left forehead, RBHD: right back head, LBHD: left back head, CLAV: clavicle, STRN: sternum, C7, T10, RSHO: right shoulder, LSHO: left shoulder, RUPA: right upper arm, LUPA: left upper arm, RELB: right elbow, LELB: left elbow, RFRM: right forearm, LFRM: left forearm, RWRB: right wrist marker B, LWRB: left wrist marker B, RWRA: right wrist marker A, LWRA: left wrist marker A, RASI: right anterior superior iliac spine, LASI: left anterior superior iliac spine, RPSI: right posterior superior iliac spine, LPSI: left posterior superior iliac spine, RTHI: right thigh, LTHI: left thigh, RKNE: right knee, LKNE: left knee, RTIB: right tibia, LTIB: left tibia, RANK: right ankle, LANK: left ankle, RHEE: right heel, LHEE: left heel, RTOE: right toe, LTOE: left toe).

During the alternate stair touch task, anticipatory balance was assessed by (1) clinical Kids-BESTest item score (four-point Likert scale) and (2) anterior-posterior Margin of Stability (AP MoS) at foot touchdown.

2.4.1. Kids-BESTest item score

The clinical Kids-BESTest item score for the alternate stair touch task is scored on a four-point Likert scale ranging from 0 to 3 based on quantitative and qualitative performance. The assessor considers the time to finish the task (quantitative performance), the number of steps (quantitative performance), and the frequency of the signs of instability (qualitative performance). Examples of these signs of instability are excessive trunk movements, hesitation, and inadequate weight shift to the stance leg. For each item score, the scoring criteria are presented in Table 1.

Table 1
Alternate Stair Touch Task Scoring Criteria

Score	Criteria
0	<8 steps, constant signs of instability, or inability to execute the movement
1	8 steps (>20 seconds) and/or has repeated signs of instability
2	8 steps (10-20 seconds) and/or has occasional signs of instability
3	8 steps (<10 seconds) and/or has no signs of instability

2.4.2. Margin of Stability

The AP MoS (in mm) of the standing leg was calculated at foot touchdown of the stepping leg (determined by the distal phalanx II peak of the stepping foot) using the following formula (Curtze et al., 2024).

$$AP\ MoS = XCoM - BoS_{AP}$$

To calculate the extrapolated CoM (XCoM), firstly, the CoM trajectory was determined and exported using Vicon Nexus Software (v2.12.1 Vicon Inc.). Next, MATLAB (2022ab, Mathworks) was used for further data analyses and processing. Using the velocity of the CoM (vCoM), derived from the CoM position with respect to time, the XCoM was then calculated using the following formula.

$$XCoM = CoM + vCoM / \omega_0 \text{ in which } \omega_0 = \sqrt{g/l} \text{ is the eigenfrequency of the pendulum}$$

The anterior-posterior BoS (BoS_{AP}) boundary, on the other hand, was determined as the anterior border of the stance foot, defined by the coordinates of the distal phalanx II marker trajectory. If the XCoM surpasses this anterior border (BoS_{AP}), then MoS values get positive,

expressing that the XCoM lies outside the BoS. Reversely, this indicates that negative values express that the XCoM lies inside the BoS (CAVE: a smaller negative value indicates that the XCoM is closer to the boundary of the BoS).

The MoS was reported for both the dominant and non-dominant stance leg. For each leg, the mean and variance of MoS across the four successful steps were calculated and used as an outcome parameter, representing dynamic balance control.

2.5. Statistical analysis

Sample size calculations (G*Power 3.1.9.7) for the first research question, based on the expected mean difference (CPc vs TDc) in the total Kids-BESTest score (4-point Likert scale), showed that a minimum sample size of 5 CPc was needed to see if there was a significant inter-group difference of 22.2% (with a mean of $69.4 \pm 13.8\%$ in CPc and $91.6 \pm 4.8\%$ in TDc), providing an effect size of 2.15. This was based on mean and standard deviation (SD) data of the total Kids-BESTest score from [Dewar et al. \(2022\)](#). Power ($1 - \beta$) was set to 0.80 with a significance (α) of 0.05.

To investigate if there was a difference in MoS for the second research question, MoS data (non-dimensional) during gait from [Rethwilm et al. \(2021\)](#) was used. A minimum of 10 CPc is required to detect a significant intergroup difference in mean MoS of 0.055 (with a mean of 0.139 ± 0.037 in CPc and 0.084 ± 0.017 in TDc) with an effect size of 0.84, assuming a power ($1 - \beta$) of 0.80 and a significance (α) of 0.05.

Statistical analyses were executed in JMP Pro (version 17.0). The data distribution was tested using Shapiro-Wilk's test and the Normal Quantile Plot (QQ-plot) for normality. For homoscedasticity, the Brown-Forsythe test was used. To verify homogeneity between the groups, two-mean independent t-tests (two-sided) were used to compare the means of the anthropometric characteristics (age, weight, and height), granted that the assumptions (normality and homoscedasticity) were met. The Chi-square test was used to compare gender between both groups. To investigate group differences in the kids-BESTest total score and the domain scores, the Rank-sum test was used. Significance was set to $\alpha = .05$, however, a Bonferroni correction was applied afterward setting the significance to $\alpha = .007$ ($.05 / 7$). To examine group differences in item score and performance time during the alternate stair touch task (domain 3, item 5), the Rank-sum test and the Welch test were used, respectively.

A two-way repeated measures ANOVA model was used to examine the differences in mean and variance of MoS AP between groups (between-factor: CP/TD) CPc and TDc, depending on limb dominance (within-factor: dominant/non-dominant leg). The assumption of normality had to be met. If there was an interaction effect between group and leg dominance, post-hoc tests were carried out using the Bonferroni correction to correct for multiple comparisons.

3. Results

3.1. Group characteristics

Gender ($X^2(1, N = 36) = 0.114, p = .735$), age ($t(34) = 0.734, p = .468$), weight ($t(34) = 0.384, p = .704$), and height ($t(34) = 0.356, p = .724$) were similar between both groups; this is shown in Table 2 and confirms the homogeneity of the anthropometric data.

Table 2
Group Characteristics

Group	Gender		Anthropometric data (M \pm SD)			GMFCS
	Girl	Boy	Age (years)	Weight (Kg)	Height (cm)	
CP	8	7	8.74 \pm 0.42	30.42 \pm 9.68	133.51 \pm 12.17	I = 9 II = 5 III = 1
TD	10	11	8.33 \pm 0.35	29.35 \pm 7.04	132.14 \pm 10.71	/
P-value	.735 ⁺		.468 [°]	.704 [°]	.724 [°]	/

Note. CP = Cerebral Palsy, TD = Typically Developing, M = Mean, SD = Standard Deviation, GMFCS = Gross Motor Function Classification System.

⁺Chi-square test, [°]independent t-test

3.2. Balance performance (Kids-BESTest)

The total Kids-BESTest score was significantly lower in CPc compared to TDc ($z = -5.04, p < .0001$)(mean group-difference: 33.01%). Also, CPc performed significantly worse across all domains, with the largest effect observed for ‘Stability in gait’ (mean group-difference: 42.51%). For each score, the descriptive (median and interquartile ranges) and statistical results are presented in Table 3 and visualized in Figure 1 and Figure 2 (A-F).

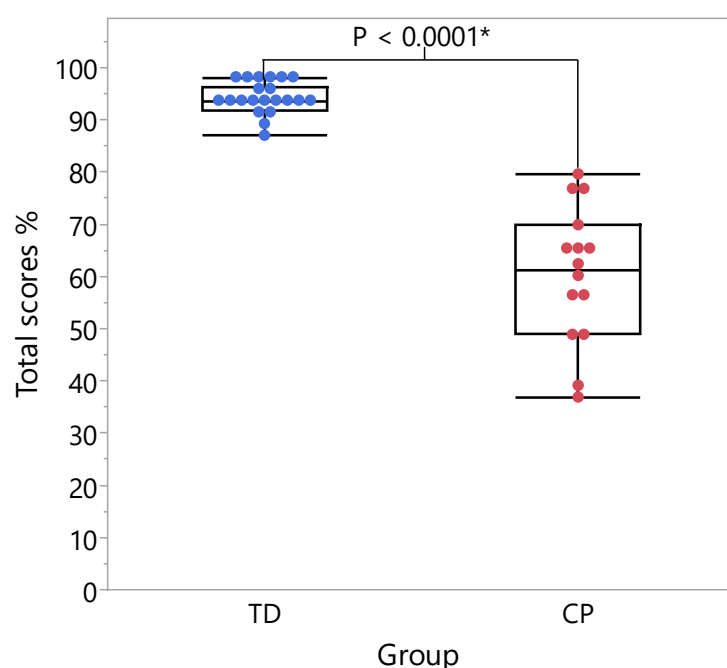
Table 3*Kids-BESTest Domain and Total Scores*

Domain	P-value ⁺	Z-value	Median ± IQR (%)	
			TD	CP
Biomechanical constraints	<.0001*	-5.00	100 ± 6.67	80 ± 6.67
Limits of stability and verticality	<.0001*	-4.90	90.48 ± 14.29	52.38 ± 30.48
Transitions and anticipatory postural adjustments	<.0001*	-4.95	93.33 ± 12.22	61.11 ± 22.22
Reactive postural responses	<.0001*	-4.99	100 ± 2.78	83.33 ± 44.44
Sensory orientation	<.0001*	-5.19	100 ± 6.67	60 ± 13.33
Stability in gait	<.0001*	-5.05	94.44 ± 6.67	50 ± 33.33
Total score	<.0001*	-5.04	93.52 ± 4.52	61.11 ± 20.87

Note. TD = typically developing, CP = cerebral palsy, IQR = interquartile range.

⁺Rank-sum test

* p <.007 due to Bonferroni correction (0,05/7).

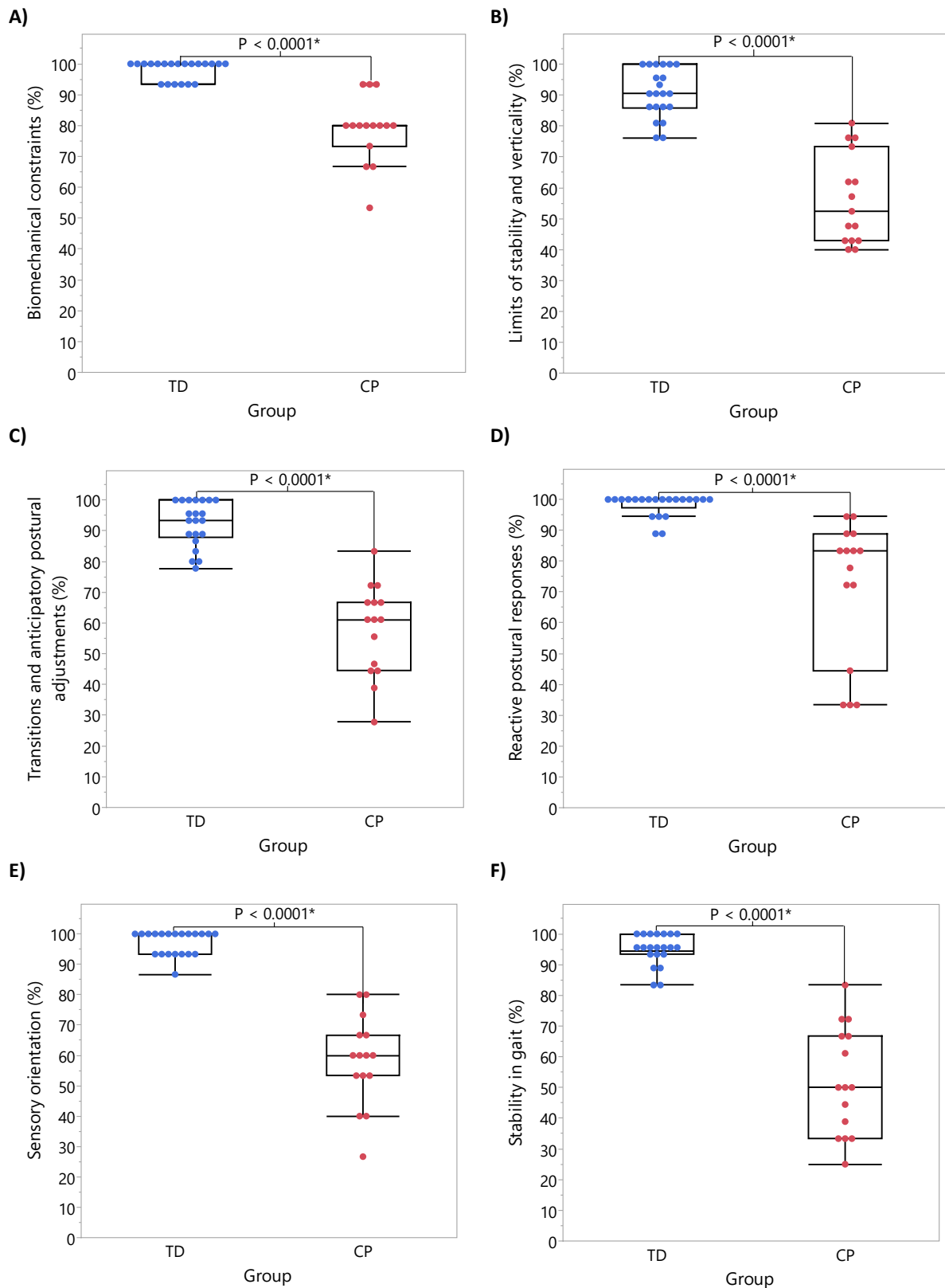
Figure 1*Box Plots of Total Kids-BESTest Scores for TD and CP*

Note. Box plots with individual data points for the total Kids-BESTest scores. TD = typically developing = Blue, CP = cerebral palsy = Red.

* p <.007 due to Bonferroni correction

Figure 2

Box Plots of the Kids-BESTest Domain Scores (1-6) for TD and CP



Note. Box plots with individual data points for the six Kids-BESTest domains and distinction between unilateral and bilateral CP. TD = typically developing = Blue, CP = cerebral palsy = Red.

* $p < .007$ Due to Bonferroni correction.

3.3. Anticipatory balance control

For the alternate stair touch task, there is a significant difference in item score ($z = -5.10$, $p < .0001$). CPc have a lower score compared to TDc (CPc scores range from 0-2 (majority 60% score 1) and TDc range from 2-3 (majority 76% score 3) and it also takes them more time to complete the task ($F = 12.11$, $p = .0034$). Table 4 compares their median and mean score with the interquartile range and standard deviation, respectively. This is also visualized in Figures 3 and 4.

Table 4

Analysis of the Alternate Stair Touch Task (Domain 3, Item 5)

	Test	P-value	Z-value	Median \pm IQR	
				TD	CP
Item 5	Rank-sum	<.0001*	-5.10	3 \pm 0.5	1 \pm 0

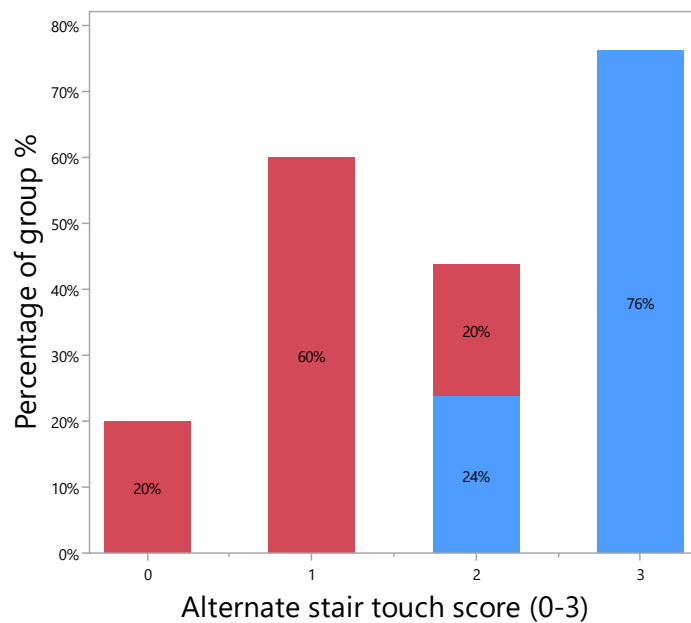
	Test	P-value	F-ratio	Mean \pm SD (Sec)	
				TD	CP
Step Time (sec)	Welch	.0034*	12.11	5.30 \pm 0.68	9.59 \pm 0.80

Note. Tests used for statistical analysis of the alternate stair touch task and their outcome. Domain 3: Transitions and anticipatory postural adjustments, TD = typically developing, CP = cerebral palsy, IQR = interquartile range, SD = standard deviation, Sec = seconds.

* $p < .05$

Figure 3

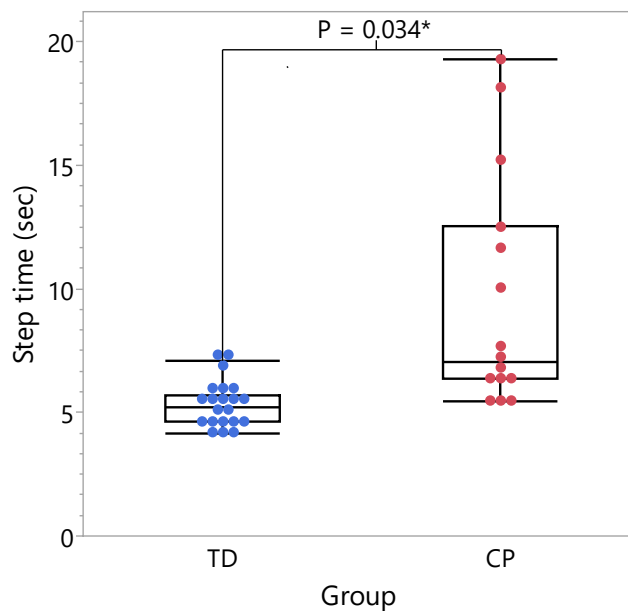
Distribution of the Alternate Stair Touch Score for TD and CP



Note. Stacked bar graph for the distribution of the Alternate stair touch task score (domain 3, item 5) with labeled percentages. TD = typically developing = Blue, CP = cerebral palsy = Red.

Figure 4

Box Plots of Step Time for CP and TD



Note. Box plots of time needed to complete the alternate stepping task. CP = cerebral palsy = Red, TD = typically developing = Blue.

* $p < 0.05$

3.4. Margin of Stability AP

3.4.1. Mean MoS AP:

For the mean MoS AP, no significant interaction effect for Group * Leg dominance ($F(1, 47.8) = 0.16, p = .6865$) was found. During stepping, MoS was higher (and thus smaller) in CPc compared to TDc (Difference CPc-TDc = 24.33 mm), with a significant main effect of Group ($F(1, 56.2) = 18.36, p < .0001$). However, these results were similar for both legs ($F(1, 47.8) = 2.75, p = .1035$). Table 6 provides information on their mean and standard deviation. This is visualized in Figure 5 (A).

3.4.2. Variance MoS AP:

A two-way repeated measures ANOVA revealed that there is no significant interaction effect Group * Leg dominance on variance MoS AP ($F(1, 49.6) = 0.01, p = .9106$). For the main effects, there was a significant effect of Group ($F(1, 58.1) = 24.17, p < .0001$), but no effect of Leg dominance ($F(1, 49.6) = 0.58, p = 0.4495$). During the alternate stepping task, MoS was significantly more variable across steps compared to TDc (Difference CPc-TDc = 13.41). Table 6 provides information on their mean and standard deviation. This is visualized in Figure 5 (B).

Table 5
Statistical Analysis of the Margin of Stability Data

MoS AP	Factor	P-value	F-ratio
Variance	Group (CP vs TD)	<.0001*	24.17
	Leg dominance	.4495	0.58
	Group * Leg dominance	.9106	0.01
Mean	Group (CP vs TD)	<.0001*	18.36
	Leg dominance	.1035	2.75
	Group * Leg dominance	.6865	0.16

Note. Outcome two-way repeated measures ANOVA. MoS AP = Margin of Stability Anterior-Posterior, TD = typically developing, CP = cerebral palsy.

* $p < .05$

Table 6

Descriptive Values of the Margin Of Stability AP

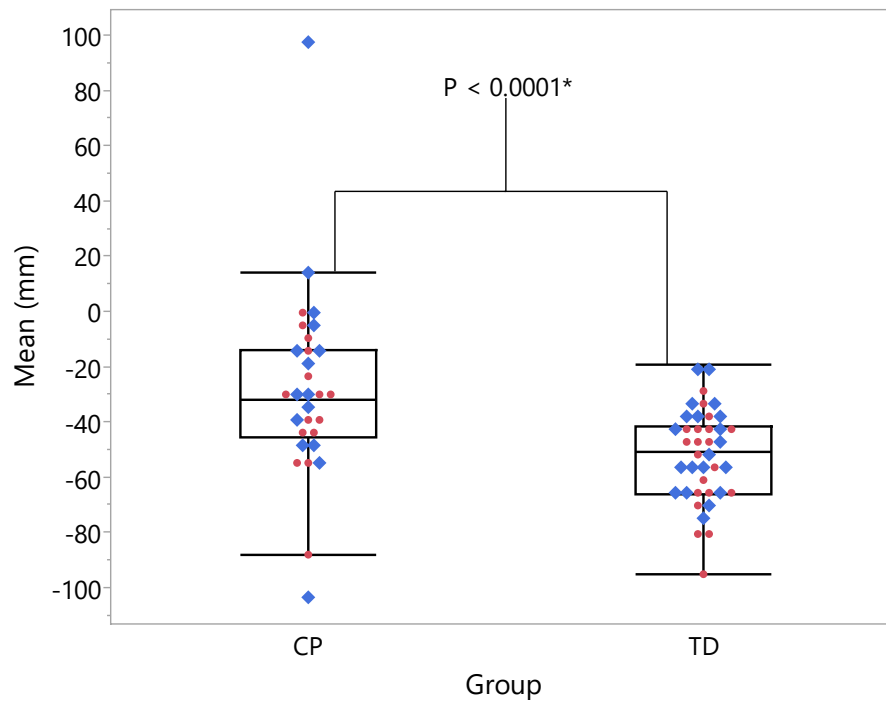
MoS AP	Group	Mean \pm SD		
		Dominant	Non-Dominant	Total
Variance	CP	24.66 \pm 15.46	26.36 \pm 14.40	25.51 \pm 14.71
	TD	10.96 \pm 6.38	13.25 \pm 10.31	12.10 \pm 8.54
Mean (mm)	CP	-23.34 \pm 43.16	-34.59 \pm 22.26	-28.96 \pm 34.22
	TD	-49.88 \pm 16.01	-56.71 \pm 16.81	-53.29 \pm 16.57

Note. MoS AP = Margin of Stability Anterior-Posterior, TD = typically developing, CP = cerebral palsy, SD = standard deviation, Dominant = dominant leg, Non-dominant = non-dominant leg

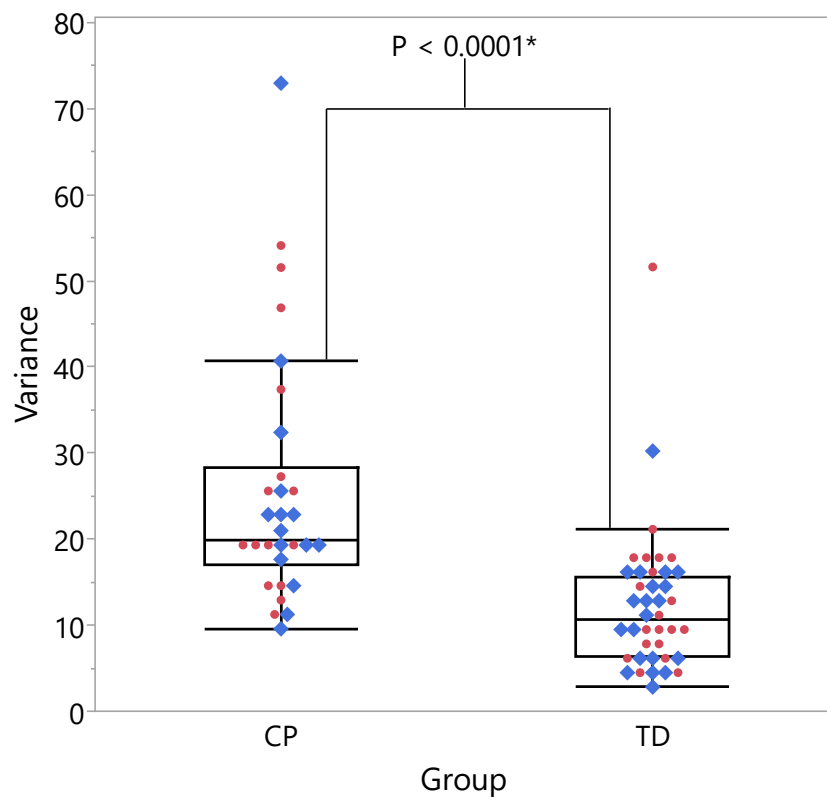
Figure 5

Box Plots of MoS AP Mean and Variance for CP and TD

A)



B)



Note. Box plots with individual data points for MoS AP variance and mean, distinguishing between the dominant (Blue) and non-dominant leg (Red). MoS AP = Margin of Stability anterior-posterior, CP = cerebral palsy, TD = typically developing.

* $p < .05$

4. Discussion

4.1. Balance performance: multisystem approach

This study aimed to see if there was a difference in balance performance with respect to the different postural control systems between CPc and TDc. Additionally, we focused on comparing the ability to control balance during a stepping task. Although extensive research has been conducted on balance performance, only recently researchers have been shifting towards more comprehensive methods of balance evaluation. This study aimed to evaluate balance by using a multisystem approach. Therefore, comprehensively addressing postural control.

As hypothesized, CPc showed poorer balance performance across all postural control systems. This study has shown that CPc median scores were lower for the total Kids-BESTest score as well as for every Kids-BESTest domain score than TDc. What stands out in Figure 2 is the difference in interquartile range between TDc and CPc. In the CPc boxplots the variability is more prominent, pointing out that between CPc, there are a lot of individual differences in performance. This variability stood out the most in Figure 2 (D), portraying the reactive postural control scores of CPc of the Kids-BESTest. The between-group differences, on the other hand, were the most notable in the assessment of the limits of stability (Figure 2(B)) and the stability in gait (Figure 2(F)). These outcomes accentuate the importance of a comprehensive balance assessment, estimating potential deficits in multiple involved systems in postural control. Future research should look into the possibility of mapping out an individual balance profile for CPc. As previously mentioned, the Kids-BESTest is based on Horak's multifactorial system for explaining postural control ([Horak et al., 2009](#); [Horak, 2006](#)). It is not a surprise that CPc score lower than TDc when assessing potential biomechanical constraints since trunk-pelvic malalignment, foot deformities, inadequate muscle strength, and range of motion restrictions in the lower limbs are common characteristics of CPc ([Abd El-Nabie & Saleh, 2019](#)). These restraints are specifically examined in the first domain since they might have an influence on the BoS and sensorimotor mechanism underlying postural control and, therefore, could have an influence on their performance ([Horak, 2006](#)). The specific influence of biomechanical restraints on other postural control systems can be an interesting direction for further research.

In addition, CPc performed worse than TDC when pushing their limits of stability in anterior and lateral directions without changing their BoS as well as when keeping an upright position with their eyes closed. CPc showed an even worse performance in controlling their stability limits and ability to stay upright than in other domains. It is known that CPc experience difficulties relying only on somatosensory and vestibular information since they rely predominantly on visual input ([Lidbeck et al., 2016](#)), making them over- or undershoot the vertical position. This could also affect the ability of the child to preserve balance in sensory-challenging conditions such as static standing with eyes open/closed and on a stable/unstable surface, as was shown in our study. Our results reflect those of Donker et al. (2008) who also found that CPc showed a greater amount of postural sway and more signs of instability when visually deprived. To preserve balance during these tasks, a child should be able to adapt its reliance on sensory input for postural control (visual, vestibular, and somatosensory) in situations of inter-sensory conflict, also known as, “sensory reweighting” ([Peterka, 2018](#)). However, CPc show, depending on the location of the lesion, more deficits in their sensory processing abilities. These can be attributed to lesions in the thalamocortical connections ([Knijnenburg et al., 2023](#)).

Furthermore, our study showed that CPc experience difficulties with balance during gait. More specifically, when confronted with numerous perturbations (e.g., head movements, dual tasks, changing speeds, obstacles crossing, ...). Slowing down before and during obstacle crossing, walking with a bigger base of support, a variable walking path during head movements, and no or asymmetric arm swing were signs of instability measured during our assessment. In accordance with these present results, previous studies have also reproduced the same conclusion. For example, Law and Webb (2005) showed that CPc tend to use a slower walking speed when approaching and crossing an obstacle in comparison with TDC. Kurz et al. (2012) found that CPc use a wider step width and similar results were found for arm swing characteristics by Meyns et al. (2011). They described that CPc have an asymmetric amount of arm swing in comparison with TDC. A possible explanation for these gait stability problems can be found in the parameters described by Chakraborty et al. (2020). This review was able to distinguish which gait parameters were more affected by CP. Double-limb support time, step length, and step width at preferred walking speed and velocity at both preferred and increased walking speed were found to be the most affected spatiotemporal parameters.

Reactive postural control assessment also showed that CPc encounter more problems than TDc. This finding is in line with previous research on reactive balance control in CPc, showing that CPc use their stepping strategy earlier than TDc when confronted with low-amplitude force plate perturbations (Chen & Woollacott, 2007; Burtner et al., 2007). These preceding studies mainly focused on in-place responses in the AP direction, whereas our study also included compensatory responses to larger perturbations in the mediolateral (ML) direction as well. This inefficient and unstable stepping strategy could be explained by difficulties in proprioceptive processing (Zarkou et al., 2020) as proprioceptive feedback plays a crucial role in restoring balance in response to unexpected external perturbations (MacKinnon, 2018).

The most important domain for this study 'Transitions and anticipatory postural adjustments' also showed that CPc scored lower than TDc, with a relatively high variability. This study further investigated item 5 of this domain (alternate stair touches) and found that CPc showed more frequent signs of instability (e.g., inadequate weight shift to stand leg and foot placement variability) and took longer to complete the task. Previous studies have primarily focussed on the APA-phase of different movements and found that CPc can generate APAs although these are delayed in onset in the lower limb (m. hamstrings, m. rectus femoris, m. gastrocnemius) and smaller in magnitude in the lower limb (m. hamstrings, m. rectus femoris, m. gastrocnemius) and trunk (m. rectus abdominis, m. erector spinae) (Liu et al., 2007; Shiratori et al., 2016; Tomita et al., 2010; Tomita et al., 2011). Rapson et al. (2023) recently reported that CPc, when stepping, have reduced APAs and a higher stepping inaccuracy in the ML direction. Therefore, the question remains how these dysfunctional APAs affect the dynamic balance control during the stepping task itself. CPc show a lot of variability in placing their foot on the step and the floor during the stepping task and are not able to adequately shift their weight toward the standing leg. Whereas the Kids-BESTest only takes into account the frequency of these instability signs as criteria for the scoring, the MoS gives more information about their balance control and strategy.

4.2 Anticipatory balance control during stepping

The hypothesis for the second research question was that CPc have a smaller and more variable MoS during the stepping task compared to TDc and that this is different depending on which leg (affected vs non-affected) is the standing leg. This study found that CPc have a smaller MoS AP, showing that they are indeed less stable. They also had a higher variance of this MoS AP data, which shows that they have more variability between each step. Interestingly, no within-group differences were found for leg dominance, which displays that both legs are affected, although a large part (40%) of the sample consisted of unilateral CP. During the stepping task, the children constantly need to transfer their weight onto the standing foot and correctly tap the step in front of them. This means their BOS is constantly changing and they need to adapt before (APA) and after (RPA) lifting their foot, which indicates that a step also influences the next step. They have to use feedback and -forward loops to execute the appropriate motor plan. In feedforward loops, APA are necessary before lifting the foot, however, these APA are affected in CPc as explained in 4.1 which means that the weight shift to the standing leg is inadequately performed. A way to compensate for this is using more trunk flexion, this can be seen in the data since their MoS is smaller and their CoM is more forward compared to TDc. This limitation implies that CPc need to rely more on their reactive system (RPA), however, they also exhibit problems in this system partly due to deficits in lower limb proprioception ([Damiano et al., 2013](#); [Zarkou et al., 2020](#)) and difficulties with agonist voluntary muscle activation ([Stackhouse et al., 2005](#)). These limitations help explain why CPc repeatedly show instability signs and may explain the higher variability in MoS AP. The fact no difference was found in MoS AP between the affected and non-affected legs may be attributed to the majority (60%) of the sample being children with bilateral CP. However, in unilateral CP when standing on the dominant leg, these children may have difficulties touching the step in a controlled manner, influencing balance of the dominant/non-affected leg.

It is important to take into account the individual differences in CPc during the stepping task. Firstly, two subgroups can be identified according to the speed-accuracy trade-off ([Heitz, 2014](#)). Some children might prioritize task performance and finish the task as fast as possible while exhibiting a lot of errors in the process. Others might prioritize balance performance with fewer errors and a slower time to complete the task. Secondly, CPc show different

balance strategies which influences the MoS greatly. For example, a child may have very variable foot placement on the step and back on the ground but exhibit relatively little trunk flexion. This has a big impact on their BoS and therefore on their MoS. The MoS can fall outside of their BoS, but at the next step, it can fall back inside. On average this cancels out and is exhibited as a normal mean MoS, yet with more variability. On the contrary, a child might have continuous trunk flexion bringing the CoM forward and resulting in a smaller MoS AP, but with normal variability since their foot placement is less variable. These are examples of how the MoS can identify differences in strategies concerning balance problems and therefore provide more information than the Kids-BESTest item score. The majority (60%) of CPc in this study scored 1 on the Kids-BESTest score, yet exhibited very different mean MoS AP. Further research into these subgroups of CPc is needed.

When interpreting the MoS it should be noted that it is measured at one point during the task (foot touchdown), this means that the MoS in this study does not reflect their global balance throughout the stepping task. Therefore, future studies could look at the MoS trajectory throughout the task using Statistical Parametric Mapping (SPM) analyses.

4.3. Further recommendations

Limitations of this study include the generalizability of these findings since this study included unilateral and bilateral CPc. Future research should assess if differences are found between these subgroups. Other subgroups, like their GMFCS level or as already mentioned, their difference in balance strategies, should also be examined. When taking into account the sample size calculations, this study needed a minimum of 5 children in each group to find a difference in the Kids-BESTest score yet included 15 CPc and 21 TDc. This means the study is overpowered, making it able to detect smaller differences and be statistically significant yet it also can be seen as a waste of resources, time and even unethical for participants (Case & Ambrosius, 2007; Hochster, 2008). Since this study used a cross-sectional design to examine balance problems in children, it is not known whether the problems are aberrant or delayed, and thus if these children can improve with age. The ages of the groups in this study were matched on average, however, age was not included in the statistical analysis.

This study found that CPc show the largest difference in 'stability in gait' and 'limits of stability and verticality' compared to TDc on the Kids-BESTest. Future research could focus on these domains, zooming in on the items. Recently, the MoS has been used to compare gait in CPc and TDc ([Tracy et al., 2019](#)) and could be used further to investigate control mechanisms of dual tasks during gait (e.g., head movements, obstacle crossing, etc.) as the Kids-BESTest does not provide this information.

Since this study found that CPc can exhibit balance problems in all systems of postural control, it is important to use a comprehensive assessment when examining these children. This also has implications for their rehab, and therefore, in the future, randomized controlled trials can be used to investigate whether an individualized training program based on a comprehensive assessment yields superior results compared to a global program without a comprehensive assessment.

5. Conclusion

This study revealed that children with CP show impairments in all systems of postural control, compared to TDC. Further analysis of the alternate stair touch task revealed differences in clinical Kids-BESTest item score, time to complete the task, and MoS AP. Group differences indicated that they are less stable during the stepping task and have a higher step variability. This study implicates the need for comprehensive assessments in the clinical setting, however, further research is needed.

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