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Impaired postural control of axial segments in children with cerebral palsy

Jonathan Pierret a,b,*, Sébastien Caudron a,c, Jean Paysant b, Christian Beyaert b, Christian Beyaert a,b

- ^a Université de Lorraine, DevAH (EA3450 Développement, Adaptation & Handicap), F-54000, Nancy, France
- ^b Institut Régional de Réadaptation, Centre Louis Pierquin, UGECAM du Nord-Est, Nancy, France
- ^c Univ. Grenoble Alpes, CNRS, LPNC, 38000 Grenoble, France

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ABSTRACT

Background: Sensorimotor control of axial segments, which develops during childhood and is not mature until adolescence, is essential for the development of balance control during motor activities. Children with cerebral palsy (CP) have deficits in postural control when standing or walking, including less stabilization of the head and trunk which could affect postural control.

Research question: Is dynamic stabilization of axial segments during an unstable sitting task deficient in children with CP compared to typically developing children? Is this deficit correlated with the deficit of postural control during standing?

Method: Seventeen children with CP (GMFCS I-II) and 17 typically-developing children from 6 to 12 years old were rated on the Trunk Control Measurement Scale (TCMS). In addition, posturography was evaluated in participants while they maintained their balance in stable sitting, unstable sitting, and quiet standing, under "eyes open" and "eyes closed" conditions. In sitting tasks, the participants had to remain stable while being prevented from using the lower and upper limbs (i.e. to ensure the involvement of axial segments alone).

Results: Children with CP compared to TD children had significantly larger surface area, mean velocity and RMS values of CoP displacements measured during the unstable sitting task and the standing task, under both "eyes open" and "eyes closed" conditions. No significant group effects were observed during the stable sitting task. The TCMS total score was significantly lower, indicating trunk postural deficit, in the CP group than in the TD group and was significantly correlated with postural variables in the sitting and standing tasks.

Significance: Children with CP indeed have a specific impairment in the postural control of axial segments. Since the postural control of axial segments is important for standing and walking, its impairment should be taken into account in rehabilitation programs for children with CP.

1. Introduction

From early childhood onwards, children with cerebral palsy (CP) exhibit impairments in postural control in static and dynamic situations, even after they are able to stand and walk on their own [1,2]. For example, during quiet standing, a greater center of pressure (CoP) sway than in typically developing (TD) children is reported, indicating less accurate and efficient postural control [2,3]. Children with CP have specific difficulty in resolving intersensory conflicts when standing, and

appear to be more affected when somatosensory information is disrupted [2]. With regard to the effect of vision on posture, most previous studies found that children with CP swayed more in an "eyes closed" (EC) condition than in an "eyes open" (EO) condition [1,3] – as did TD children (i.e with the same effect magnitude). At the opposite, few studies evidenced a greater effect of visual deprivation in children with CP compared to TD children [34]. Therefore, the visual deprivation effect on the postural control of children with CP with reference to TD children remains unsolved, especially under challenging condition.

Abbreviations: ANOVA, analysis of variance; CP, cerebral palsy; CoP, center of pressure; DR, dynamic reaching; EC, eyes closed; EO, eyes open; GMFCS, Gross Motor Function Classification System; RMS, root mean square; SMC, selective movement control; SSB, static sitting balance; TCMS, Trunk Control Measurement Scale; TD, typically developing.

^{*} Corresponding author at: EA 3450 DevAH, Université de Lorraine, Campus Brabois – Santé -Bât C – 9 Avenue de la Forêt de Haye, F-54505, Vandoeuvre-les-Nancy, France.

E-mail addresses: jonath.pierret.research@gmail.com (J. Pierret), sebastien.caudron@univ-grenoble-alpes.fr (S. Caudron), jean.paysant@ugecam.assurance-maladie.fr (J. Paysant), christian.beyaert@univ-lorraine.fr (C. Beyaert).

While sitting [5] or standing [6], children with CP from early childhood (2 years old) until adolescence (~ 15 years old) also exhibit abnormal postural reactions to external disturbances consisting of sudden translations of the support. Specifically, children with CP have a descending recruitment pattern of lower limb muscles in the standing position, which are opposite to the ascending and distal-to-proximal recruitment patterns observed in TD children. These atypical muscle recruitment patterns are associated with highly variable muscle responses, long activation latencies, antagonistic co-contractions, and poor adaptation to the intensity of the disturbance [6,7]. Interestingly, in TD children, the development of axial stabilization continues between 6 and 11 years of age and is not yet mature at the beginning of adolescence [17]. Since children with CP show a delay in the development of postural responses in sitting and standing, we could suggest that they would also show a deficit in axial segment stabilization during this period where it is still developing in TD children. Indeed, during middle childhood, in the sitting position, children with CP notably have difficulty keeping their head stable and sway more than TD children [8,9]. When walking, children with CP have also a greater range of head and trunk motion in each plane [10-12]. Given that the trunk is the primary reference frame for postural control [13], the development of postural and kinetic activities (such as locomotion and the acquisition of gross motor skills) is also likely to be altered by impaired control of axial segments [13].

We therefore reasoned that evaluating ability to stabilize axial segments may be of value for the therapeutic management of children with CP. Growing interest in the assessment of trunk control in children with CP has prompted the development of a validated, specific clinical tool the Trunk Control Measurement Scale (TCMS) [14]. This scale provides a functional evaluation of trunk control in a sitting subject during movements of the upper and lower limbs (with the trunk stabilized) or during active movements of body segments (including the trunk). Furthermore, a number of unstable seat devices have been recently developed [15]. The maintenance of balance on these devices requires strong postural regulaton by the upper body. In this way, they allowed the specific evaluation of trunk postural control in various pathological population [15,16]. Recently, the development of axial segment control has been analyzed in TD childrend aged from 6 to 12 by using such an unstable sitting device [17]. This methodology is therefore suitable to pediatric populations. To the best of our knowledge, unstable seat devices have not previously been used to test the postural control of axial segments in children with CP. At this age period (ages 6-12) over which this function is still gradually developing in TD children [17], this approach would highlight the contribution of axial segment impairments to balance disorders in children with CP.

The main objectives were to determine whether (i) children with CP compared to TD children have a deficit in postural control of axial segments, using TCMS and an unstable sitting device; and (ii) whether trunk control assessed with TCMS correlates with postural control assessed by posturography during standing and sitting postural tasks. An additional objective was to determine if visual deprivation has a higher effect on postural stabilization in children with CP compared to TD children.

2. Methods

2.1. Participants

Seventeen children with CP (7 girls, 10 boys, age: 7.9 y \pm 2.4, height: 127 cm \pm 15, weight: 26.7 kg \pm 7.5; 11 diplegic and 6 hemiplegic) and 17 age-matched TD children (8 girls, 9 boys, age: 7.7 y \pm 2, height: 129 cm \pm 15, weight: 25.6kg \pm 6.8) were included in the study. During the medical consultation, it was determined that all the children had a sufficient level of understanding of and cooperation to perform the tasks and were able to walk without walking aids [for the children with CP, Gross Motor Function Classification System (GMFCS) level I: n=9;

GMFCS level II: n=8]. The GMFCS was assessed by the same experienced pediatrician (CB), after interviewing the parents and the child [18]. None of the TD children had a history of neurologic or musculoskeletal disorders. None of the children with CP had undergone surgery to correct orthopaedic disorders in the lower limbs or axial segments or had received botulinum toxin injections in the lower limbs in the 6 months prior to the study. The experimental protocol complied with the tenets of the Declaration of Helsinki and was approved by the local investigational review board "CPP Est-III" (France) n°2015-A000022-47/15.02.03. The participants' parents gave their written, informed consent to their child's participation in the study.

2.2. Tasks and data acquisition

2.2.1. The sitting postural tasks

The children were asked to sit on an unstable seat device (Fig. 1) placed on a three-dimensional force platform (AMTI, Watertown, MA, USA). A cardan joint and four springs arranged around the cardan joint allowed the seat to tilt in a frictionless manner along the mediolateral and anteroposterior axes, with a maximum lowering of 3 cm of the seat edges (tilt of 12°). The participant was instructed to remain as still as possible while sitting in an upright position with the arms crossed on the chest (i.e. suppressing the upper limbs' contribution to postural control), and the feet resting on an adjustable footrest (in height and depth) integral with the unstable seating device. The knee and hip joint angles were kept at 90°, and movements of the lower limbs were also restricted by foam blocks strapped between and around them (i.e. suppressing the lower limbs' contribution to postural control). Hence, only the body's axial segments contributed to balance on the unstable seat device. The device was either locked in a static horizontal position using wooden blocks (the stable sitting postural task; Fig. 1C) or was left unlocked (the unstable sitting postural task). In the latter task, we used the prior calibration procedure developed in 2013 by Larivière et al. (individual adjustment of the distance between the pivot joint and the springs) [15] so that the task's level of difficulty was independent of the participant's anthropometric characteristics.

2.2.2. The standing postural task

The children were required to stand on the three-dimensional force platform with their arms at their sides, feet about hip width apart and to remain as still as possible.

The sitting and standing postural tasks (each lasting 30 s) were carried out three times under both eyes open (EO) and eyes closed (EC) conditions. The order of the sitting tasks stable vs. unstable combined with the EO vs. EC conditions was randomized. The participant rested for 30 s between tasks.

2.2.3. The Trunk Control Measurement Scale

The TCMS is composed of three subscales all completed in sitting posture: the static sitting balance (SSB) subscale (20 points) evaluates the capacity to stabilize the trunk during upper and lower limb movements; the selective movement control (SMC) subscale (28 points) evaluates the selective movement of the trunk within the base of support; and the dynamic reaching (DR) subscale (10 points) measures the ability to make reaching movements outside the base of support [14]. Each item on the scale was scored three times, and the best of the three scores was recorded. The total TCMS score ranged from 0 (worst performance) to 58. The TCMS was administered after postural tasks after a rest period of at least 15 min to avoid measurement bias due to fatigue. TCMS was administered by the same experienced investigator (JP).

2.3. Data recording and processing

During all the postural tasks, CoP trajectories were recorded at a sampling rate of 1000 Hz. Raw data were filtered with a low-pass Butterworth filter (order: 4; cut-off: 12 Hz). Next, the following data were

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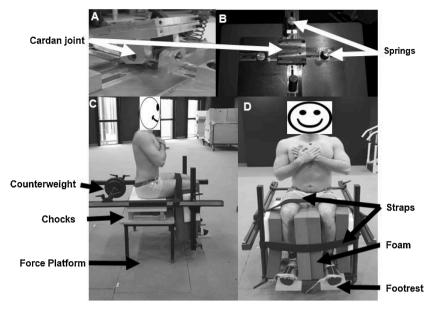


Fig. 1. (A) A side view of the unstable seat device's cardan joint. (B) A view of the underside of the seat, showing the cardan joint and the four springs. The difficulty of the postural task can be homogenized as a function of the participant's anthropometric characteristics by changing the distance between the spring and the cardan joint; the greater the distance, the more stable the seat. (C) Sagittal and (D) frontal views of the unstable seat device.

averaged over three trials: (i) the CoP area (mm²), computed from the 95 % confidence ellipse of the CoP displacement; (ii) the CoP mean velocity (mm.s⁻¹); and (iii) the root mean square (RMS) of the CoP displacements (mm). All the data processing steps were performed using MAT-LAB R2017 software (MathWorks, Natick, MA, USA).

2.4. Statistical analyses

Quantitative data were reported as the mean \pm standard deviation (SD). For each postural task, the three postural dependent variables were analyzed separately using a mixed-design analysis of variance (ANOVA). For each variable, a 2 \times 2 two-way ANOVA was performed with "group" as the between-subject factor (TD group; CP group) and "vision condition" (EO; EC) as the within-subject factor. Tukey's honestly significant difference test was used for post-hoc comparisons, when necessary. Size effects were reported with partial eta² statistics (η_p^2). The TCMS total score and subscale scores for the CP and TD groups were compared using independent Student's t-tests. Pearson's correlation coefficient was computed in each group, in order to examine putative linear relationships between the TCMS scores and each of the CoP variables during the sitting and standing postural tasks. The threshold for statistical significance was set to $\alpha=0.05$.

3. Results

In children with CP, the demographic variables, postural variables, and TCMS did not differ significantly between GMFCS I and GMFCS II; therefore, the data were pooled for analysis.

3.1. The stable sitting postural task

During the stable sitting postural task, there was no main effect of group or group \times vision interaction both for CoP area, CoP mean velocity and CoP RMS. However, there was a significant main effect of vision, with higher values under EC conditions than under EO conditions (Fig. 2, Table 1).

3.2. The unstable sitting postural task

One child in the CP group failed to complete this task, and so the corresponding data were excluded from this analysis. During the unstable sitting postural task, significant main effects of group and vision were observed for the three dependent variables; the values were higher in the CP group than the TD group, and higher in the EC condition than in the EO condition, respectively (Fig. 2, Table 1). There was also a significant interaction between the group and the vision condition for the three variables (Table 1). More precisely, all the variables had significantly higher values in the CP group than in the TD group under both EC and EO conditions (EC condition: p < 0.001 for all variables, EO condition: p < 0.001 for CoP area, p = 0.02 for CoP velocity, and p = 0.008 for CoP RMS). For each variable, there was a significant difference between the EO and EC conditions in each group (p < 0.001 for each variable in the CP group; p < 0.05 for each variable in the TD group).

3.3. The quiet standing postural task

During the quiet standing postural task, significant main effects of group and vision were observed for the three dependent variables; the values were higher in the CP group than the TD group, and higher in the EC condition than the EO condition (Fig. 2, Table 2). Lastly, there was no significant interaction between the group and the vision condition.

3.4. The TCMS scores

The mean \pm SD total TCMS score was significantly lower in the CP group than the TD group [36.3 ± 6.9 vs. 53.3 ± 3.2 , respectively; t(32)=9.28, p<0.001]. The same was true for the SSB subscale score [CP: 16.9 ± 2.6 ; TD: 19.8 ± 0.6 ; t(32)=4.44, p<0.001], the SMC subscale score [CP: 12.2 ± 4.0 ; TD: 23.8 ± 2.9 ; t(32)=9.66, p<0.001] and the DR subscale score [CP: 7.1 ± 1.2 ; TD: 9.8 ± 0.6 ; t(32)=8.42, p<0.001]. The total TCMS score had significant, moderate-to-strong negative correlations with (i) the CoP area and CoP RMS during the unstable sitting task in the CP group only and during the quiet standing task in both the CP and TD groups, and (ii) the CoP velocity during the stable sitting task in the TD group although a non-significant trend was observed in the CP group (Table 2).

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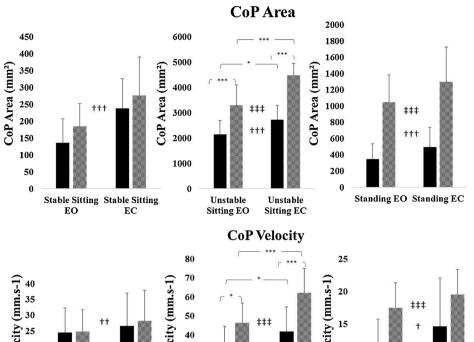


Fig. 2. Mean confidence ellipse area (in mm²), CoP velocity (in mm.s⁻¹) and CoP RMS (mm) recorded during the stable sitting postural task, the unstable sitting postural task, and the standing postural task in the TD group (dark bars) and the CP group (grey bars) and under eyes-open (EO) and eyes-closed (EC) conditions. The error bars correspond to 1 SD. The symbol † indicates a significant difference between EO and EC (††† p < 0.001), and the symbol ‡ indicates a significant difference between the TD and CP groups ($\ddagger\ddagger p < 0.001$). The asterisk indicates significant post-hoc test result for the vision x group interaction (*p < 0.05; **p <0.01; ***p < 0.001) – notably the difference between EO and EC conditions within a group, and an intergroup difference within a condition.

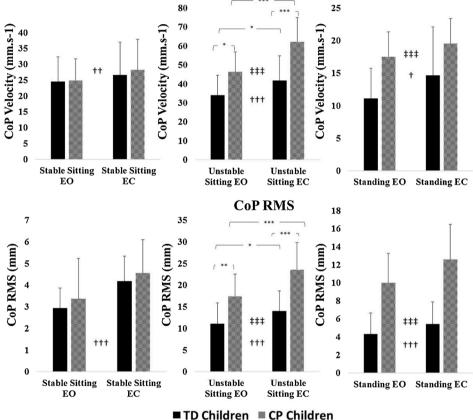


Table 1 Summary of statistical results concerning the ANOVA performed on the three CoP variables (Area, mean velocity and RMS) during each postural task (stable sitting, unstable sitting, quiet standing). Main Group effect and vision effect and group \times vision interaction are reported with F, p and η^2 values.

Postural task	Postural variable	Group			Vision condition			$Group \times vision \ interaction$		
		F Value	p	η^2	F Value	p	η^2	F Value	p	η^2
Stable sitting	CoP Area	$F_{(1,32)} = 2.81$.10		$F_{(1,32)} = 41.0$	<.001	.56	$F_{(1,32)} = 0.13$.71	
	CoP Velocity	$F_{(1,32)} = 0.10$.74		$F_{(1,32)} = 10.5$	<.01	.24	$F_{(1,32)} = 0.56$.45	
	CoP RMS	$F_{(1,32)} = 0.8$.37		$F_{(1,32)} = 37.9$	<.001	.54	$F_{(1,32)} = 0.03$.85	
Unstable sitting	CoP Area	$F_{(1,31)} = 79.4$	<.001	.71	$F_{(1,31)} = 41.7$	<.001	.71	$F_{(1,31)} = 5.10$.03	.14
	CoP Velocity	$F_{(1,31)} = 20.9$	<.001	.40	$F_{(1,31)} = 34.2$	<.001	.52	$F_{(1,31)} = 4.17$.04	.11
	CoP RMS	$F_{(1,31)} = 22.2$	<.001	.41	$F_{(1,31)} = 39.1$	<.001	.55	$F_{(1,31)} = 4.98$.03	.13
Quiet standing	CoP Area	$F_{(1,32)} = 58.4$	<.001	.64	$F_{(1,32)} = 21.2$	<.001	.39	$F_{(1,32)} = 1.40$.24	
	CoP Velocity	$F_{(1,32)} = 15.7$	<.001	.32	$F_{(1,32)} = 6.95$	<.01	.17	$F_{(1,32)} = 0.51$.47	
	CoP RMS	$F_{(1,32)} = 46.4$	<.001	.59	$F_{(1,32)} = 16.5$	<.001	.33	$F_{(1,32)} = 2.60$.11	

Table 2Pearson's r for the correlations between the total TCMS score and the dependent postural variables

Postural task	Postural variable	TD group	CP group	
	CoP area	-0.26	-0.2	
Stable sitting	CoP velocity	-0.53*	-0.43^{t}	
	CoP RMS	-0.26	-0.1	
	CoP area	0.13	-0.52*	
Unstable sitting	CoP velocity	0.15	-0.20	
	CoP RMS	0.13	-0.52*	
	CoP area	-0.63**	-0.68**	
Quiet standing	CoP velocity	-0.45^{t}	-0.2	
	CoP RMS	-0.63**	-0.68**	

t indicates a trend (p = 0.05 to 0.08).

4. Discussion

Children with CP had more difficulty than TD children in stabilizing their axial segments during unstable sitting and quiet standing tasks. In children with CP, the total TCMS score was lower than in TD children and was negatively correlated with postural variables in both the unstable sitting and quiet standing tasks. Visual deprivation was associated with decreased stability in both groups and in all the sitting and standing postural tasks.

4.1. Impaired postural control of axial segments in children with CP

Relative to TD children, children with CP displayed impairment in the postural control of axial segments in the unstable sitting task but not in the stable sitting task. This suggests that control of axial segments was sufficiently well developed in children with CP to stabilize on a stable seat. Furthermore, the children with CP included in this study (GMFCS I or II) had moderate impairments of gross motor function. Liao et al. [8] reported that CoP sway distances did not differ significantly when comparing children with CP and TD children during static sitting but were significantly higher in children with CP during dynamic sitting on a support tilting backward before being back to horizontal.

Both static and dynamic aspects of postural control of the trunk were impaired in the CP group according to our present results for the three TCMS subscale scores, as previously reported by Heyrman et al. [14]. Interestingly, the total TCMS score was moderately and negatively correlated with both CoP area and CoP RMS during the unstable sitting task. This finding shows that the unstable sitting task is a reliably challenging task that specifically reveals and quantifies impairments in the postural control of axial segments. Furthermore, the unstable sitting task is not based on an externally induced movement of the support; hence, it contrasts with earlier reports of dynamic postural tasks involving either an expected movement (such as support oscillations [8] or unexpected translations [5]). On the contrary, the seat movements during unstable sitting are internally induced in relation to the movements of the axial segments because none of the four limbs can move independently. As a result, the ability to maintain balance on the unstable sitting device is suggested to rely to some extent on a proactive control of the posture, requiring the anticipation of axial segment oscillations that will induce inclinations of the seat. This provides a possible explanation for the difficulty of children with CP to stabilize themselves during the dynamic tasks. In TD children, the improvement in postural control is partially explained by an improvement in proactive control with age [19], which is based on the progressive construction of internal model of action [20]. In children with CP, the development of internal models of action is disrupted, as well as the proactive control [22,23], specifically in the trunk musculature [21].

Furthermore, the self-balancing task becomes even more difficult when the seat angle is high, due to poor stabilization of the axial segments and altered postural reactions [5,7]. In addition, muscle weakness — especially at the level of the *gluteus maximus* and *medius* — may accentuate the difficulties that children with CP have in balancing on the unstable sitting device [25,26,35]. Indeed, while the thighs are immobilized on the unstable sitting device, hip muscles which exert a force on the pelvis and indirectly on the seat, may contribute to the sitting posture and its stability [24].

Impaired postural control of axial segments may be a key target for rehabilitation in children with CP. Enhanced axial rehabilitation, based on a variety of exercises in intermediate postures that strongly involve the trunk to cope with balance, may be of great interest if it affects the standing postural control [27].

4.2. Does the impaired control of the trunk affect the standing postural control in CP children?

The total TCMS score is negatively correlated with both CoP Area and RMS in CP and TD children. Thus, a lower TCMS score is associated with larger postural sways during the quiet standing task. The trunk, which accounts for about 60 % of body mass in children [28] and thus contributes to the elevated position of the body's center of mass in the standing position, plays a crucial role in postural control [29]. Indeed, even small deviations of the trunk have a strong impact on the CoP displacements. It is worth noting that TD children in middle childhood develop in parallel their ability to stabilize themselves on an unstable sitting device involving only the axial segments [17] as well as during quiet standing [30]. The fact that postural sways during quiet standing do not decrease with age in children with CP [1] may be related to delayed postural development [31] which also affects control of the axial segments. A longitudinal study of axial stabilization in children with CP would be required to confirm this hypothesis.

4.3. The effect of visual deprivation on postural control is more pronounced in CP children when the task is difficult

As already reported elsewhere [1,3], visual deprivation was associated with higher values of all the CoP variables in all tasks and in both groups. This finding underlines the importance of visual feedback for postural control, as extensively highlighted in the literature. The interaction observed between vision and group in the unstable sitting task suggests that when the postural challenge was accentuated (by either increasing the level of difficulty or modifying the sensory context), the difference between CP and TD children in the control capacity of axial segments increased. Earlier researches have suggested that children with CP have impairments in the organization of sensory inputs. This would probably lead to difficulties when inputs are disrupted during task performance, in particular when the support is unstable and vision is absent [2]. Although the unstable seat task assessed in the present study did not involve sensory disturbance per se, reliance solely on somatosensory and vestibular information prevented children with CP from balancing as effectively as TD children. It should be noted, however, that during the stable sitting task the difference between the mean values of the CoP variables in the EO and the EC condition, although significant, is relatively small (especially for the CoP velocity) and therefore may not be clinically relevant.

4.4. Study limitations

Both for postural variables and for TCMS, we failed to note significant differences between children with CP at the GMFCS I level and children with CP GMFCS II. By the way, some other studies — more specifically designed for that purpose — also did not report significant difference between GMFCS I and II groups, during quiet standing analyzed with conventional posturography [32]. However, the analysis of the dynamic structure of CoP trajectories during quiet standing [32] or the functional reach test (testing dynamic standing balance) [33] are

 $_{**}^{*}p < 0.05.$

^{**}p < 0.01.

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other methods more sensitive to differentiate children GMFCS I and II. One might expect that dynamic postural control of axial segments evaluated with the unstable sitting postural task and TCMS could lead to observe differences between GMFCS I vs. II children. However, the recruitment of children with CP in the present study, not initially designed and quantitatively too low to assess differential effects from groups well-balanced, does not allow us to conclude about this point, as well as about a potential influence of the affected topography (hemiplegic or diplegic). Indeed, this latter factor could also affect the severity of axial control disorders.

Since this study was designed to analyze postural control of axial segments of children with CP over a period when it is still developing in TD children, it would have been appropriate for the study to be a cross-sectional study involving subgroups of age, which would require a much larger number of subjects. Neuromuscular abnormalities are reported to explain disorders of postural control in children with CP [5,25].

It would have been informative, in our study of axial stabilization, to clinically assess the strength of trunk stabilizing muscles such as gluteal muscles, abdominal muscles and trunk extensors. Electromyographic recording of these muscles would have allowed to identify their involvement in the feedforward and feedback mechanisms of trunk control.

Dynamic stabilization on the unstable sitting device could be influenced by a learning effect: this effect was reduced by randomizing the conditions (stable and unstable, eyes open and eyes closed). However, the randomization between the different tasks could have been improved if the TCMS had been included in this randomization. For practical reasons, the TCMS was performed after the postural tasks.

5. Conclusion

To the best of our knowledge, the present study is the first to have used an unstable seat device in a dynamic postural task that specifically highlighted the impairment of axial segment postural control in children with CP. This impaired ability to self-stabilize by using axial segments was correlated with a more general impairment in trunk control under various static and dynamic conditions, as evidenced by the TCMS score. Since axial segments have an important role in motor activities such as standing, we suggest that the impaired postural control of axial segments is likely to contribute to the self-stabilizing difficulties observed among children with CP in quiet standing. Since the postural control of axial segments is important for standing and walking, its impairment should be taken into account in rehabilitation programs for children with CP.

Declaration of Competing Interest

The authors report no declarations of interest.

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