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The gait is less stable in children with cerebral palsy in normal and dual-task gait compared to typically developed peers

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ABSTRACT

Keywords: Gait variability, Inertial measurement unit, Gait kinematics, Stability, Walking, Attention

1. Introduction

Cerebral palsy (CP) is a permanent disorder affecting the motor development attributed to non-progressive lesion or trauma occurred prenatally or early in life in the developing brain (Bax et al., 2005). The majority of children with CP exhibit impairments in gait and balance control (Beckung and Hagberg, 2002; Hutton and Pharoah, 2002). Although the gait in CP appears less stable than in typically developed (TD) controls, it is not straightforward to objectively quantify the gait stability (Bruijn et al., 2013a). The methods for this are emerging but may require sample sizes > 100 steps when using wearable assessment devices (Riva et al., 2014a), such as inertial measurement units (IMUs), and the derived indices can lack intuitive physical meaning. E.g., multiscale-complexity measures, indicating the total amount of entropy can be challenging to interpret (Riva et al., 2013, 2014a). Thus, ambiguity still exists regarding gait stability in CP, and no general procedure exists to objectively quantify gait stability in clinical practice (Chakraborty et al., 2020).

Gait has shown to be less stable in children with CP compared to TD controls by using optical 3D-gait analysis and “foot-placement estimator” (Bruijn et al., 2013b), “spatiotemporal relationships between center of mass and pressure” (Hsue et al., 2009a, 2009b) and “Floquet analysis of step variability” (Kurz et al., 2012). IMUs have been used to compute a “harmonic ratio” between even and odd harmonics of the stride (body) acceleration to reflect step-to-step asymmetry (Bellantuono et al., 2013). Harmonic ratio has been shown to be higher (i.e. less stable gait) in children with CP than in TD peers (Iosa et al., 2012). Overall, unconstrained gait appears less stable in children with CP than in TD peers.

There is limited evidence how gait stability is altered by concurrent motor and cognitive tasks in CP. Dual tasking is common in everyday life and can have a significant effect on the child’s ability to manage the daily tasks. The gait kinematics indeed differ between CP and TD in unconstrained and motor-task constrained...
gait (Hung and Meredith, 2014), cognitive-task constrained gait (Carcreff et al., 2019; Palluel et al., 2019), and are altered more in CP by concurrent cognitive task (Katz-Leurer et al., 2014). However, it is unclear whether children with CP demonstrate an excessive dual-task cost (i.e. impairment of stability).

There is demand for a standardized, easy-to-use, robust method to quantify and monitor gait stability. To that end, IMUs are wearable, relatively unobstructed, and require low labour cost in recordings, and have potential to follow effects of neurorehabilitation on gait stability in children with CP (Iosa et al., 2018). One promising metric is “refined-compound-multiscale entropy” (RCME), capturing complexity at multiple temporal coarseness scales (Ihlen et al., 2016). The shorter time-scales may be indicative of within step cycle variations, and the longer time-scales of step-to-step or stride-to-stride variations (Ihlen et al., 2016), and has been shown to be an efficient predictor of prospective falls in elderly (Ihlen et al., 2016, 2018), with reasonable week-to-week reliability (Rantalainen et al., 2020). However, RCME as an indicator of gait stability remains untested among children with CP.

Our first aim was to clarify the feasibility of the IMU-RCME method in quantifying gait stability. We hypothesized that IMU-RCME method reveals significant differences between patients and TD, and that IMUs provide similar gait kinematics (step duration) as conventional optical 3D-method. Our second aim was to evaluate the effect of dual-tasking on gait in children with hemiplegic (HP) or diplegic (DP) CP in comparison to TD controls. We hypothesized that the gait complexity is higher (i.e. less stable gait) and is more altered (i.e. higher dual-task cost) by constrained cognitive and motor tasks in children with CP, and that the gait stability would be associated with static standing postural balance.

2. Materials and methods

2.1. Participants

Patients. Thirty children and adolescents (13.3 ± 2.3 years, 18 female) with spastic hemiplegic (HP: 13.5 ± 2.4 years, n = 18, 12 female) or diplegic (DP: 13.0 ± 2.1 years, n = 12, 6 female) CP participated in the study. Their Gross Motor Function Classification System level was I–II (I in 5 patients), meaning the ability to walk independently but limited to minimal ability to perform gross motor skills such as running and jumping (Palisano et al., 1997). They had no known cognitive or co-operative deficiencies, hearing deficit, visual deficit other than refractive error, orthopedic surgery done in the lower extremities, condition (other than CP) or medication known to affect gait and balance. The patients were recruited from the rehabilitation unit of Children’s hospital, Helsinki University Hospital and by advertising via a patient organization.

Controls. Thirty-one healthy TD controls (13.5 ± 2.2 years, 18 female) participated in the study. They had no known cognitive or neurological deficits or medication known to affect gait and balance. The controls were recruited from school visits, the university and hospital staff’s family members and acquaintances.

The study was approved by the ethics committee of Helsinki University hospital (HUS/2318/2016). The study was conducted in accordance with the Helsinki Declaration. All the volunteered patients and healthy controls gave informed verbal assent, and their guardians gave written informed consent to participate in the study.

2.2. Experimental setup

The participants walked at preferred speed up-and-back a 10-m walkway in the gait laboratory of Helsinki University Hospital, Finland. Fig. 1 illustrates the experimental setup. There were three different tasks: (1) normal unconstrained gait, (2) motor dual task where the participant carried a tray with an empty mug on top of it, and (3) cognitive dual task where the participant listed aloud semantic (animals or foods and drinks) or phoneme (words starting with s or k) class words as fast as possible in accordance with the NEPSY-II assessment (Korkman et al., 2007). All three tasks were performed in randomized order in sets of ten 10-m-walks within four blocks. In total, there were 40 sets for each task (~400 steps/task).

2.3. Measurements

We used a lightweight IMU (NGIMU, x-io Technologies Limited, Bristol, UK) placed on mid-back at L3–L5 level with an elastic Velcro-strap to record kinematics of the body during gait. 3-axis of acceleration (range ± 16 times gravitational acceleration) and 3-plane gyroscopic signals (range ± 2000°/s) were sampled at 100 Hz with 16-bit analog-to-digital conversion and were stored to PC via Wi-Fi connection. Optical 3D-gait data (~100 steps/task) was collected simultaneously at 100 Hz using 16-camera Vicon system (Vicon, Oxford, UK).

To assess whether dynamic and static stability were associated, we recorded postural sway (center-of-force velocity) using plantar-pressure plate (Hi-End Footscan®, RSscan, Paal, Belgium) at 33 samples/s. Participants stood eyes open as still as possible for 30 s.

2.4. Data processing

Accelerations and gyrations were estimated in the laboratory coordinate system by utilizing the gradient descent algorithm (Madgwick et al., 2011) calculated by the IMU-device, and were used in subsequent calculations. Each pass through the 10-m walkway was identified based on resultant acceleration intensity, and gyration around the vertical axis. Changes of direction at either end of the walkway and initial and final portions of the gait were removed from the analysis based on the mean vertical acceleration below 0.05 g using sliding 0.5 s window. The data 0.5 s before and after crossing the threshold were omitted. RCME (Ihlen et al., 2016; https://github.com/tjrantal/javaMSE) indices were computed separately for vertical and resultant horizontal accelerations from each pass. Briefly, template length (m) of four was used, 20 temporal coarseness scales (τ) were considered, and 0.3 of the standard deviation of the pass was used as the tolerance (Rantalainen et al., 2020). The mean of all passes for each temporal coarseness scale was computed, and were further summarized with varimax principal-component (PC) analysis. The first PC was used for the further analysis to represent overall gait complexity.

Steps were identified from each pass based on 2-Hz zero-lag low-pass-filtered vertical acceleration at least 0.1 s lasting continuous peaks above the median of the filtered vertical acceleration. Heel-strike events were then defined as the instant of the highest peak of the product of resultant acceleration and gyration within the step peak event. Step durations were calculated as the differential between the subsequent heel strike events. The step durations were filtered to include only steps within 0.5 of the median of the step durations to account for any heel-strikes that were not identified. The mean and the standard deviation (SD) of pooled (across the 40 sets in each task) step durations are reported.

To determine walking speed and step duration, we computed center of mass motion using the 3D-data and Plugin gait full-body model. First, marker trajectories were filtered using Woltring filter (mean-square-error value of 10 mm²). Step-duration analysis was based on foot strike events, which were defined using a combination of force plate and kinematic event-detection method (Bruening and Ridge, 2014), detecting 10-N-vertical force (4th order
Butterworth filtered) thresholds from force-plate contacts (four AMTI HPS464508HF, Watertown, MA, USA). The corresponding resultant velocity of the heel marker was then used to identify the foot strike events for the off-plate steps.

2.5. Statistical analyses

IBM SPSS Statistics software (ver. 26) was used. Non-parametric tests with Holm-Bonferroni correction for the multiple comparisons were used due to low sample size and thus non-normal distribution in some of the variables. Kruskal-Wallis Test was used to test main effects in between-groups comparisons (TD, DP and HP), and Mann-Whitney Test indicated differences between the specific groups. Friedman Test was used to test main effects in between-tasks comparisons (normal, motor and cognitive) within each group separately, and Wilcoxon Signed-Ranks Test indicated differences between the specific tasks. Spearman correlation coefficient was used to test the consistency between IMU and 3D-data based step-duration estimations, and association between static and dynamic stability.

3. Results

The IMU recordings proved feasible. Two patients had missing set (1 out of 4) in some of the three tasks due to problems in Wi-Fi connection. All participants had minimum of 123 s (mean ± SD: 386 ± 180 s) of data per task for RCME analysis. The number of words listed during the cognitive task did not differ between the TD (17.3 ± 5.5 words/min), DP (14.1 ± 4.1 words/min) and HP (13.7 ± 4.9 words/min) groups (p = 0.058). Postural sway during static standing task was correlated with gait complexity both in vertical (r = 0.42, p = 0.001) and horizontal (r = 0.51, p < 0.001) directions (Fig. 2).

3.1. Between-group gait differences

Walking speed. The preferred walking speed (Fig. 2) was slower for DP in all tasks (normal 1.04 ± 0.19 m/s, motor 0.99 ± 0.18 m/s and cognitive 0.93 ± 0.14 m/s) compared to TD (p < 0.001, normal 1.28 ± 0.14 m/s, motor 1.23 ± 0.15 m/s and cognitive 1.15 ± 0.14 m/s) and HP (p < 0.05, normal 1.21 ± 0.15 m/s, motor 1.13 ± 0.11 m/s and cognitive 1.08 ± 0.09 m/s). No difference was observed between TD and HP.

Step duration. Step durations from 3D and IMU-data correlated strongly for all tasks (p < 0.001, normal: r = 0.99, motor: r = 0.91 and cognitive: r = 0.98; p < 0.001). The step duration was similar between the groups (p > 0.586), but its variation was higher in HP for all tasks (p < 0.001–0.01) and DP for normal (p = 0.013) and motor (p = 0.007) tasks when compared to TD group (Fig. 2).

Gait complexity. The gait complexity was greater in CP than TD for all tasks both in vertical and horizontal directions (p < 0.001–0.01; Fig. 3). The overall gait complexity (first PC) was greater in DP (p < 0.001–0.01) and HP (p < 0.01–0.05) for all tasks and directions compared to TD, apart from the vertical direction during normal gait in HP (p = 0.059). No difference was observed between the DP and HP group (p > 0.075). The majority of the 20 coarseness scales above the first scale were greater in HP (p < 0.001–0.05) and DP (p < 0.001–0.05) compared to TD for all tasks and directions. DP showed more complex gait than HP for some coarseness scales in the vertical direction (p < 0.05–0.001) predominantly during the cognitive task.

3.2. Between-task gait differences

The preferred walking speed was slower during cognitive task than normal gait (Fig. 2; TD p < 0.001, HP and DP p < 0.01) and motor task (only in TD p < 0.001 and HP p < 0.01). Furthermore,
TD showed slower walking speed during motor task than normal gait (p < 0.001).

The step duration was longer for the dual-tasks than normal gait only in TD (p < 0.001; Fig. 2), but its variation was higher for the dual-tasks in all groups (p < 0.01–0.001). The gait complexity did not differ significantly between the tasks within each group (Fig. 3b).

3.3. Dual-task cost

Fig. 4 illustrates the dual-task cost (i.e. difference to normal gait) for all groups. No significant dual-task cost effect was observed for step duration (p > 0.361) or its variation (p > 0.385). However, gait complexity (first PC) increased more from the nor-

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**Fig. 2.** Step duration and its standard deviation, walking speed, and association between standing postural balance and gait stability (RCME). The step duration values are from IMU based analysis, and the walking speed values are from 3D data. N = normal gait, M = motor-task constrained gait, C = cognitive-task constrained gait. *, **, *** = different from TD group at p < 0.05, p < 0.01, + = different between DP and HD groups at p < 0.05, p < 0.001. ||, ||| = different from normal gait at p < 0.01, p < 0.001.

**Fig. 3.** Gait complexity. (a) Gait complexity at 20 different coarseness scales of the refined-compound-multiscale entropy (RCME). (b) 1st principal component of the 20 coarseness scales. N = normal gait, M = motor-task constrained gait, C = cognitive-task constrained gait. # = different between HP and TD groups at p < 0.05. ^ = difference between DP and TD groups at p < 0.05. + = difference between the DP and HP groups at p < 0.05. *, **, *** = different from TD group at p < 0.05, p < 0.01, p < 0.001.
mal to dual-task gait in HP and DP groups compared to TD group (p < 0.05). The dual-task gait cost did not differ between HP and DP.

4. Discussion

We showed that the IMU-RCME is feasible to quantify gait stability that was more complex (indicating lower stability) in children with hemiplegic and diplegic CP compared to TD peers, and that the cognitive and motor dual tasks altered the gait complexity more in patients with CP (i.e. higher dual-task cost). The effects were emphasized in the attentionally more demanding cognitive task and vertical direction of the body acceleration, and were stronger in DP compared to HP patients.

4.1. Between-group differences in the gait stability

The step duration was similar between the patients and controls, and thus does not explain the differences in the gait stability. However, especially the healthy individuals slowed down their gait during the dual-tasks who also had the fastest and most stable gait. This may indicate that CP patients have less “reserve” to adapt their gait for the increasing task demands. Interestingly, patients with DP had more stable gait than HP, likely because both of their limbs are bilaterally affected and thus DP patients cannot rely on the less affected limb to support their gait stability (Bohm and Doderlein, 2012).

The impaired gait stability in CP was further pronounced in the dual-task conditions. Our findings are aligned with the literature, which have shown that gait kinematics differ between CP and TD both in unconstrained gait and motor-task constrained gait (Hung and Meredith, 2014), and are altered more in CP by concurrent cognitive task (Katz-Leurer et al., 2014). In contrast to our findings, Tracy et al. (Tracy et al., 2019) using “margin of stability” method (Hof et al., 2005) did not find differences in anterior gait stability between CP and TD, but elevated stability in the mediolateral direction when bearing weight on their non-dominant limb compared to TD. Furthermore, their participants were more stable during a cognitively constrained gait than unconstrained one. This contradiction could reflect different methodology used to assess gait stability. Indeed, the “gold standard” method and protocols are still needed for consistent quantification of gait stability at different sites and patient populations.

Less stable gait is less efficient and may expose the person to injuries and falls (Ihlen et al., 2018; Rispens et al., 2015). Elevated risk of falling is a generic problem in CP and is not specific to certain gait pattern abnormality (Boyer and Patterson, 2018). Our results indicated that the gait stability is impaired in CP, which lead to elevated risk of falling, especially in dual-task situations. However, this should be confirmed with prospective studies by following how well the IMU based gait stability index predicts prospective falls at the individual level. Importantly, worse gait and standing stabilities were associated, indicating that the impaired static postural stability is transferred to dynamic stability in children with CP.

4.2. Dual-task cost and effects

The children with CP showed higher dual-task cost than TD in some of the tasks, primarily in vertical direction of the body acceleration during cognitive task in diplegia. We hypothesise that the more the gait is impaired the less automated it is, and thus requires additional attention and cortical contribution. Therefore, we postulate that the elevated attentional demands by the concurrent motor and especially cognitive task impair gait more in the individuals with the most affected gait.

The neurophysiological mechanism for the less automated gait in CP is unclear and most likely varies from patient to patient. In general, gait impairments in CP are believed to be of neuronal origin, being associated with simplified motor control strategy (i.e. impaired efference), muscle weakness and spasticity (Beckung and Hagberg, 2002). Another possible explanation for the less automated gait is impaired somatosensory afference from the periphery to the brain in CP, due to consequences of the early brain lesion to development of the sensorimotor cortices (Rossignol et al., 2006). Consequently, the cortical motor control may be hindered due to inaccurate internal model of the locomotor system in the brain (due to impaired afference), provided by the proprioceptive system (Prosko and Gandevia, 2012). This hypothesis is supported by evidence that both joint-position sense (Wingert et al., 2009) and cortical proprioceptive processing (Piitulainen et al., 2020) are impaired in CP, which are compensated by use of other senses, e.g., vision in CP (Liao and Hwang, 2003) to accomplish motor tasks. This compensatory strategy requires additional attention to the ongoing motor task that could partly explain our results, but both impaired efference and afference mechanisms may be valid in CP. Therefore future studies should attempt to quantify the proprioceptive abilities objectively to identify the specific mechanisms of impaired gait stability.

Cognitive competence (listed words/min) during walking did not differ significantly between the groups. It thus appears, that children with diplegia put greater emphasis on the cognitive task at the cost of gait stability compared to TD with presumably more automated gait. Therefore, the effect of the cognitive dual-task was pronounced in DP.

The dual-task cost was emphasized in the vertical direction of the body acceleration, that reflects the redirection of the centre of the mass velocity during step-to-step transitions (Adamczyk and Kuo, 2009). This finding further suggest that sub-optimal transitions during dual-task gait may be a mechanism that reduces the overall robustness and reactivity of the locomotor system to sudden external perturbations (Mazaheri et al., 2015).

4.3. Benefits and limitations of IMU based gait stability

IMU recordings are simple to implement, and therefore a suitable option for future screening of gait disorders, and e.g. to monitor the effects of neurorehabilitation on gait stability in children with CP (Josà et al., 2018). IMUs are becoming more miniaturised, ubiquitous and with lower energy demands, which would enable long-term free-living monitoring and potentially gait self-
assessments (Benson et al., 2018). Wearable technology can be utilised in non-laboratory environments, could potentially increase the ecological validity. However, this would need to be investigated in further research. The most important factor to ensure high-quality IMU data pertains to preventing the whipping caused by a loose bonding of the device onto the body. IMU needs to be firmly attached to a suitable location, e.g., to lower back. Therefore, smartphones etc. are not always the best options.

Several methods to compute indicators of gait stability from IMU data have been suggested, e.g., harmonic ratio (Bellanca et al., 2013), Floquet multipliers (Riva et al., 2014a), complexity estimates (Ihlen et al., 2016). It remains to be shown which of these methods are best associated with gait impairments and would provide the most beneficial clinical information. We chose to test RCME in the present study as the implementation was shared openly, and it had already been shown to have prognostic ability for falls among older adults (Ihlen et al., 2016; Ihlen et al., 2018). There is arguably room for software development in this field to provide the clinicians with easy-to-use software tools for interpreting and reporting gait dynamics based on laboratory- and free-living IMU-recordings.

5. Study limitations

Gait entropy analyses were conducted only for the parts of constant velocity between changes of directions at ends of the 10-m walkway, and subsequently calculated the mean of all steps, which is standard approach for spatiotemporal gait variability assessments. Future studies could aim to collect continuous gait data, although this choice does not appear to be a major issue (Riva et al., 2014b).

When assessing gait in children and adolescents their leg length varies, and may affects the gait parameters (Hof, 1996). In the current study, TD controls were age-matched to patients with CP, thus there were no significant difference in the age (p = 0.577) nor leg length (p = 0.113) between them. In addition, the leg length did not correlate to any of the used gait stability measures (ps > 0.491).

Although consistent results were observed based on IMU assessed step duration and its variation we have previously reported poor criterion validity for variation (Rantalainen et al., 2020). When compared to the present 3D-motion capture step duration SD the IMU indicated a significant 26 ms bias (p < 0.001) and essentially no agreement (ICC = −0.25, 95% and CI −0.44 to −0.03) between the methods. However, the differences between the groups and conditions were detectable despite the imprecision of the IMU assessment.

6. Conclusions

The gait in children with CP was less stable (i.e. higher complexity) and the dual-task cost was higher primarily for children with diplegic CP than TD in the vertical direction of the body acceleration, indicating that attentional load hinders more the gait stability in children who potentially have less automated gait. This means that more cortical resources are needed to compensate the impaired gait especially in diplegic patients.

Declaration of Competing Interest

The authors declared that there is no conflict of interest.

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