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ORIGINAL ARTICLE

A continuum of balance performance between children with developmental coordination disorder, spastic cerebral palsy, and typical development

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ABSTRACT

BACKGROUND: Balance deficits are one of the most common impairments in developmental coordination disorder (DCD) and cerebral palsy (CP), with shared characteristics between both groups. However, balance deficits in DCD are very heterogeneous, but unlike in CP, they are poorly understood.
AIM: To unravel the heterogeneity of balance performance in children with DCD by comparing them with CP and typical development (TD).
DESIGN: Cross-sectional case-control study.
SETTING: Different outpatient settings and the community.
POPULATION: Children aged 5-10.9 years with TD (N.=64, boys: 34, mean [SD] age: 8.1 [1.6]), DCD (N.=39, boys: 32, mean [SD] age: 8.1 [1.5], formal diagnosis [N.=27]), and CP (N.=24, boys: 14, mean [SD] age: 7.5 [1.4], GMFCS level I [N.=14]/II [N.=10], unilateral [N.=13]/bilateral [N.=11]).
METHODS: We evaluated balance performance with the extended version of the Kids-Balance Evaluation Systems Test (Kids-BESTest). Between-group differences in domain and total scores (%) were assessed via ANCOVA (covariate: age), with Tukey *post-hoc* analyses ($P \leq 0.01$).
RESULTS: Children with DCD and CP performed poorer than TD children on total and domain scores with large effects (domains: $\eta^2=0.25$-0.66 [$P<0.001$], total: $\eta^2=0.71$ [$P<0.001$]). Still, post hoc comparisons revealed that DCD children scored significantly better than CP on the total score and four domains ($P \leq 0.009$), while performing similarly on tasks related to stability limits ($P=0.999$) and gait stability ($P=0.012$).
CONCLUSIONS: There is a continuum of balance performance between children with TD, DCD and CP, but with great inter- and intra-individual heterogeneity in DCD and CP. DCD and CP children have difficulties with tasks requiring anticipatory postural adjustments, fast reactive responses, and with tasks that require complex sensory integration, suggesting an internal modeling deficit in both groups. This implies that these children must rely on slow conscious feedback-based control rather than fast feedforward control and fast automatic feedback. The performance of both DCD and CP children on their stability limits/verticality is similarly poor which further emphasizes a potential deficit in their sensory input and/or integration. Future research must focus on unraveling the control mechanisms, to further understand the heterogeneity of these balance deficits.
CLINICAL REHABILITATION IMPACT: The heterogeneous balance performances in both children with DCD and CP underscore the importance of comprehensively evaluating balance deficits in both groups. This comprehensive assessment contributes to a better understanding of individual balance deficits, thereby facilitating more tailored treatment programs.

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KEY WORDS: Postural balance; Motor skills disorders; Cerebral palsy.

Children with developmental coordination disorder (DCD) are characterized by motor difficulties, among which 60-87% experience balance performance levels far below their age-expectations.[1-3] These balance deficits significantly interfere with their daily life activities in school, home and/or leisure.[4, 5] Consequently, improvement of these balance deficits is the most frequent request for help in physiotherapeutic practice.[6] However, balance deficits in DCD are very heterogeneous and still poorly understood.[7]

DCD is a neurodevelopmental condition and persists into adolescence and adulthood, leading in 50% of the cases to psychosocial sequelae, such as anxiety or social isolation, and to physical complaints related to cardiovascular diseases, such as obesity and diabetes.[5] Symptoms emerge in early childhood, but the diagnosis of DCD is mostly made after the age of 5 years as the demand for more complex motor skills rises during school-age.[5] DCD often co-occurs with other neurodevelopmental conditions such as attention-deficit/hyperactivity disorder (ADHD) (22-50%) or autism spectrum disorder (ASD) (4-18%).[5]

It is hypothesized that DCD may lie on a continuum with ambulatory CP children as they can show similar deficits in, for instance, higher-order motor control processes, such as movement planning or clinical presentation, such as balance performance.[8-11] Specifically, in children with DCD or CP who were exposed to pre- and perinatal risk factors such as preterm birth, some evidence exists that the neural structure, at both macro and micro levels, shows similarities.[8] Nevertheless, both DCD and CP are two distinct disorders based on the presence (CP) or absence (DCD) of neurological lesions.[4, 12] Although, CP is a heterogeneous disorder, specific subgroupings based on its clinical presentation exist and are widely accepted.[12, 13] In DCD, however, the underlying neurological mechanisms causing the clinical presentation are less understood.[5, 14]

Balance deficits are evident among all children with DCD.[14, 15] However, the existing studies evaluated balance deficits to a limited extent, focusing on only one or a few aspects of postural control, but failing to address the comprehensive underlying postural control framework described by Horak.[7, 14, 16]

To ensure adequate postural control, proper functioning, and interaction of each individual part of the sensorimotor system is required.[16, 17] The multisystemic framework by Horak (2006)[16] considers task specificity in balance tasks by distinguishing the following postural control systems: movement strategies (anticipatory and reactive postural adjustments), sensory strategies (reweighting somatosensory, visual and vestibular information to stay balanced), orientation in space (orienting the body (parts) in space with respect to gravity, the support surface, visual surround and internal references), and control of dynamics (controlling the body's center of mass while changing from one posture to the next). Depending on the type of postural control task, different systems are required.[16] During typical childhood, school-aged children refine their different postural control systems to eventually acquire healthy adult-like control through learning and experience between 10 and 14 years of age. This development is non-linear and occurs at different rates depending on the specific control system.[18]

Children with DCD show heterogeneous postural control deficits situated within and across the different control systems because they seem to be task specific.[7, 14] Their anticipatory control, movement planning, and fast online monitoring seem to be inefficient, making them rely on slower feedback-based control to execute voluntary movements appropriately. Consequently, in easier tasks, such as standing still with eyes open or exploring their functional stability limits in anterior direction, it seems that children with DCD compensate as they perform similarly to TD children. However, they fail and perform poorer than TD children in complex tasks requiring more anticipatory control or sensory integration.[7] Evidence on the reactive postural responses remains conflicting.[7] The reliance on slow feedback-based control agrees with the internal modeling deficit hypothesis of DCD.[5]

Like in DCD,[6] improvement of balance difficulties is one of the most frequent requests for help in CP.[8, 9, 19] The spastic subtype is the most prevalent and therefore the most investigated group in postural control research.[20] Similar to DCD, children with spastic CP also have difficulties with their anticipatory postural adjustments,[20-23] which they try to compensate for with their slower feedback-based control system in easy conditions, such as quiet standing with eyes open.[24] They have difficulties in finding their limits of stability, also in the anterior direction,[25, 26] with sensory integration, especially when vision and somatosensory input are disturbed, and with tasks requiring fast reactive postural responses in different directions.[21, 24] More difficulties are evident with increased task complexity, which confirms an internal modeling deficit in CP related to postural control in these children.[10]

Hence, both children with DCD and CP seem to be at risk for balance deficits across all postural control systems, possibly caused by an internal modeling deficit.[7, 21] Still, the previous results are a combination of individual studies targeting one or two systems comprising different samples

and different outcome measures with a limited variation of task types. Targeting all different systems in the same sample is needed to explain the heterogeneity of postural control deficits and to understand how the different systems are interrelated in DCD and in CP.[17, 27]

Despite the overlapping deficits between DCD and CP, children with CP more consistently show poor results compared with TD, whereas those with DCD perform more variably. Furthermore, in CP, the specific brain lesions and clinical subgroupings, such as uni- *versus* bilateral or Gross Motor Function Classification System (GMFCS) levels, are related to specific postural control deficits.[20, 21] Yet, given the similarities in their neural structure,[8] it seems reasonable to assume that the underlying neural alterations in children with DCD and CP cause a continuum of behavioral outcomes such as balance performance. Nevertheless, to the best of our knowledge, no study has compared postural control between children with DCD and those with CP. This comparison, however, would allow us to better understand and explain the heterogeneity in balance deficits in DCD. This will ultimately guide more tailored physiotherapeutic evaluation and treatment.

Therefore, the aim of this study is to unravel the heterogeneity of balance performance in children with DCD by comparing the performance to those with CP and TD. This resulted in the following research question: what are the differences in balance performance between children with DCD, CP, and TD? First, we hypothesize that there is a continuum of balance performance, which is defined as a spectrum where the potential variability in performances of children with DCD are expected to be situated. Using the continuum, children with DCD are situated between children with CP (severe deficits at the lower end of the continuum) and children with TD (normal performance at the higher end of the continuum). This is hypothesized to result in significantly different averages between groups but with overlapping individual results of DCD with both CP and TD. Second, we hypothesize that the group average of both DCD and CP children is significantly lower than that of TD children. Third, we hypothesize that both CP and DCD will show inter-individual and intra-individual variability considering the deficient postural control systems, with more heterogeneous results in the DCD group.

Materials and methods

Data were collected for this case-control study between August 2021 and July 2023 and approved by the Committee for Medical Ethics UZA-UAntwerp on June 4th, 2021 (ID: B300201941833, chairperson: Prof. P. Michielsen). Five- to ten-year-old children were recruited via regular or specialized schools, private practices, the Antwerp CP referral center, other PhD researchers within the same research group (after the parent's consent), acquaintances and social media. Prior to enrollment, parent(s)/guardian(s) provided written informed consent, and the children provided informed assent after the test procedures were explained.

Participants

An age-matched sample, consisting of children with TD, DCD, and CP was included when they met the predefined eligibility criteria. Regardless their group assignment, all children had to be aged between 5 and age 10 years 11 months, without any signs of cognitive delay. Comorbidities, *i.e.* ASD and ADHD, were allowed in each group in order to make the groups representative of the study populations.

The TD children were included when they were born at term (≥37 weeks of gestation), had no diagnosis of a neuromotor or another medical, behavioral, or intellectual condition potentially impeding typical balance performance (objectified by a parent-reported general questionnaire), and showed typical motor competence (objectified by a total score >percentile 16 on the Movement Assessment Battery for Children, Second Edition (MABC-2).[28]

Children with DCD were included when they met all four Diagnostic and Statistical Manual of Mental Disorders, 5th ed. (DSM-5) criteria, objectified as recommended by the latest international recommendations for DCD:[4, 5] (criterion A) motor skill acquisition and performance is at an age-inappropriate level (objectified by the MABC-2: total score ≤percentile 16/subscale score ≤percentile 5[28]); (criterion B) motor skill deficits significantly and persistently interfere with the activities of daily living (objectified by the parent-reported DCD-Questionnaire (DCD-Q) and/or if a child follows physiotherapy targeting motor problems[29]); (criterion C) onset of symptoms in early childhood (evaluated by the parents with anamnesis/general questionnaire); (criterion D) motor skill deficits are not better explained by another medical (neurological, intellectual, visual, etc.), psychological, social condition, or cultural background (evaluated by a neuromotor examination performed by an acknowledged pediatrician or when confirmed by the parents). Children were officially diagnosed with DCD, registered in a diagnostic trajectory with suspected DCD or detected in the TD group (Supplementary Digital Material 1: Supplementary Figure 1).[5]

Children with a diagnosis of spastic uni- or bilateral CP

were included if they were classified with the GMFCS as level I, II or III. All children had to be able to stand independently for at least 1 min and walk for at least 6 m with(out) walking aid (*e.g.*, key walker).[12, 30, 31] A magnetic resonance image (MRI) must have been taken to confirm the diagnosis.

Since no data in children with DCD were present at the start of the study, the sample size was estimated based on pilot data of the Balance Evaluation Systems Test for Children (Kids-BESTest) in 15 young TD children aged 5 to 7. To achieve an effect size f of 0.63 with a power of 0.8 and a Type I error probability of 0.05, a total sample size of 30 TD children would be needed, *i.e.* 10 children in each age band (age 5, age 6 and age 7). This sample size per subgroup was multiplied by two for the DCD and CP group, to correct for variability within each group (N.=20) and the TD group was considered the size of the DCD and CP group combined (N.=40). *Post-hoc* power calculations based on the collected data revealed a power of 0.93 (η^2=0.71, effect size f=1.56, numerator=2, number of groups=3, number of covariates=1, Type I error probability=0.05).

Assessments

Balance Evaluation Systems Test for Children, Extended version

Balance performance was evaluated using an extended version of the original Kids-BESTest.[32, 33] The Kids-BESTest is a comprehensive criterion-referenced test with six different domains: 1) biomechanical constraints; 2) stability limits/verticality; 3) anticipatory postural adjustments/transitions; 4) reactive postural responses; 5) sensory orientation; and 6) stability in gait. Domain 1 evaluates the prerequisites for balance, whereas the remaining five domains assess balance performance. The test comprises 36 different tasks across the six domains (summary of the tasks in Table I). Each task is scored from 0 (worst performance) to 3 (best performance) on a 4-point rating scale. The tasks are scored either quantitatively, qualitatively, or a combination of both. The extended version considers developmental changes from the age of 5. This version comprises age-specific cut-off values (quantitative scores), age-specific qualitative scores and deleted specific tasks because they are too difficult at that age. Decisions and modifications are all based on previous evidence and were performed in collaboration with the original Kids-BESTest authors. This resulted in the following age-bands: 5 years, 6 years, 7 years, 8-10 years, 11-14 years. Also, we have added additional qualitative movement descriptors to facilitate the observation of the performance of each task. More details are provided in another publication on the development of this extended version. The domain scores are expressed as a percentage calculated from the acquired score (sum of task scores) *versus* the potential maximal score, which varies per domain according to the number of tasks. The total score is the average of the different domain scores, expressed as a percentage. The original Kids-BESTest is reliable in TD and CP children aged 8 to 14 years.[32, 33] Validity has been investigated for specific tasks.[34, 35]

TABLE I.—*Kids-BESTest tasks per domain.*

Domain	Tasks
Domain 1 Biomechanical constraints	1. Base of support 2. Center of mass alignment 3. Ankle strategy and range of motion 4. Hip/trunk lateral strength 5. Sit on the floor and stand up
Domain 2 Stability limits/Verticality	1. Lateral lean – left/right 2. Verticality 3. Functional reach forward 4. Functional reach lateral – left/right
Domain 3 Anticipatory postural adjustments/Transitions	1. Sit to stand 2. Rise to toes 3. Stand on one leg – left/right 4. Alternate stair touch 5. Standing arm raise
Domain 4 Reactive postural responses	1. In place response – forward 2. In place response – backward 3. Compensatory stepping – forward 4. Compensatory stepping – backward 5. Compensatory stepping – lateral – left/right
Domain 5 Sensory orientation	1. Clinical test for sensory interaction and balance a. Firm surface eyes open b. Firm surface eyes close c. Foam surface eyes open d. Foam surface eyes closed 2. Incline eyes closed
Domain 6 Stability in gait	1. Gait on level surface 2. Change in gait speed 3. Walk with head turns – horizontal 4. Walk with pivot turn 5. Step over obstacle 6. Timed up and go test 7. Timed up and go test with dual task

Screening and descriptive measures

The MABC-2[28] was used to check eligibility in TD and DCD children. The DCD-Q[29] evaluated motor skill difficulties in daily life to check eligibility of TD and DCD children and to describe all children. Parents filled out a general questionnaire to check eligibility and collect descriptive information related to the children's general and

medical background: pregnancy, birth, any diagnosed developmental disorders, severe visual or hearing impairments, use of aids such as glasses, orthoses, and cochlear implants, medication use, leisure activities, and sports participation. The children's gross motor competence and their psychosocial well-being were described by results on the Test for Gross Motor Development, Third Edition (TGMD-3)[36] and the Strengths and Difficulties Questionnaire (SDQ)[37] scores respectively.

Test procedure

Prior to assessment, parents completed the questionnaires. Children were assessed either in a specialized movement lab (TD, DCD and CP children), in their usual private practice (DCD or CP children), or at school (TD and DCD children), in a quiet separate room with a level floor that was at least eight meters long. The results of this study are part of a larger case-control study that also investigates relationships between balance and motor performance and between balance performance and underlying control mechanisms (brain and muscle activity). For 6- to 10-year-olds, the entire test procedure lasted for approximately 3 hours, starting with the assessment of six Kids-BESTest items in combination with functional Near-Infrared Spectroscopy and electromyography: Lateral lean, Stand on one leg, Alternate stair touch, In place response backward, Compensatory stepping backward, and Step over obstacle. Afterwards, the remaining items of the Kids-BESTest (without sensors) were further assessed, then the MABC-2 (in TD and DCD children) and finally the TGMD-3 was performed. There was at least one break during the entire session. Five-year-olds only performed the functional assessments (total duration of 1-1.5 h). When assessing children at a regular school, we spaced the assessments at intervals of up to two weeks. First, the MABC-2 was evaluated to check eligibility. Afterwards, the Kids-BESTest and TGMD-3 were assessed.

The assessment of the Kids-BESTest lasted for 20-30 min, assessed by an experienced assessor (CJ, SV, NJ) or two master students who were thoroughly trained before assessment. Training comprised online preparation (8 hours) and hands-on sessions (24 hours). The domains of the Kids-BESTest were randomized per child. All assessments were videotaped and reevaluated in case there was doubt about a specific child's performance.

Statistics

Statistical analyses were performed using JMP Pro 17 (SAS Institute Inc., Cary, NC, USA).

Normality was checked visually and through the Shapiro-Wilk test. Continuous descriptive variables were described with the mean, SD, and range (age, body length, body weight, BMI, TGMD-3 scores, MABC-2 scores, DCD-Q score, and SDQ scores). The frequency of categorical variables was determined (sex distribution, number and type of co-occurring diagnoses, whether parents reported an impact score in the SDQ, DCD diagnosis, GMFCS level, uni-/bilateral CP). Between-group differences were assessed using a One-Way ANOVA (continuous) or using a Chi-squared Test (categorical). The MABC-2 scores between DCD and TD were determined using an independent samples *t*-test. The significance level was set at 0.05.

The covariate age and dependent variables were nonnormally distributed but showed equal variances. To evaluate group differences in balance performance, we applied an ANCOVA in which the dependent variables were the domains and total scores expressed as percentages, group was considered as a fixed factor, and age was added as a covariate. We considered both the main and interaction effects. If not significant ($P>0.01$) the interaction term was removed from the model. Eta squared (η^2) was calculated by taking the sum of squares of the group or age-effect divided by the total sum of squares of the model and used as measures for the effect size of group and age per domain and for the total score. The effect size, $\eta 2$, was considered small: 0.01-0.059, medium: 0.06-0.13 or large ≥ 0.14.[38] *Post-hoc* pairwise comparison was performed with the Tukey HSD test comparing all groups to each other accounting for age. To estimate the individual balance performance, each score of the DCD and CP children was evaluated against a criterium of our TD values. The criterium was based on the exploratory cut-off of percentile 15 (p15) of our TD data (N.=69), which was determined for the total and domain Kids-BESTest scores. Next, the results for DCD and CP were dichotomized per domain and for the total score (0 if ≤p15 of the TD children and 1 if >p15). Then, the distribution of 0 and 1 per domain and for the total score was compared between the CP and DCD groups using the Chi-squared Test. The significance level was set at 0.01 to correct for multiple testing (repeated analyses on domain and total scores).

Results

Participants

A total of 127 children were included, 64 with TD, 39 with DCD, and 24 with CP. The reasons for exclusion are provided in Supplementary Digital Material 2, Supplementary Figure 2. Of the children with DCD, 27 received a

TABLE II.—*Descriptive details of all groups.*

	Typical development		Developmental coordination disorder		Cerebral palsy		P value
	N.	Mean (SD)/min-max	N.	Mean (SD)/min-max	N.	Mean (SD)/min-max	
Age (years)	64	8.05(1.58)/5.00-10.62	39	8.14 (1.54)/5.42-10.95	24	7.52 (1.44)/5.07-10.35	0.271[a]
Body length (cm)	64	131.75 (10.68)/107.50 -156.00	39	130.03 (12.60)/109.50-154.00	22	127.61 (10.08)/113.5-150.5	0.317[a]
Body weight (kg)	64	27.71 (6.74)/18.00-53.70	39	29.03 (8.37)/16.60-49.90	23	26.32 (7.10)/18.10-43.20	0.364[a]
BMI (kg/m^2)	64	15.79 (2.00)/11.89 – 23.55	39	16.86 (2.56)/13.32-23.51	22	16.15 (2.43)/13.19-22.20	0.070[a]
Sex (n) (F:M)	64	30:34	39	7:32		10:14	0.011[c]
TGMD-3 total score – gross motor index (47-158)	59	93.47 (8.59)/76-114	37	67.51 (10.97)/50-88	19	69.74 (8.39)/47/88	<0.001[a]
Locomotor – scaled score (0-20)	59	9.37 (1.53)/6-13	37	4.11 (2.23)/1-10	20	4.10 (2.22)/1-8	<0.001[a]
Ball skills – scaled score (0-20)	59	8.44 (2.09)/5-14	37	4.84 (2.47)/2-10	19	5.53 (1.95)/1-8	<0.001[a]
MABC-2 total score – percentile	64	55.91 (24.05)/25-95	39	2.58 (3.90)/0.1-16			<0.001[b]
Manual dexterity – percentile	64	55.97 (23.62)/5-98	39	12.18 (18.51)/0.1-75			<0.001[b]
Aiming & Catching – percentile	64	44.59 (27.18)/5-95	39	7.84 (10.40)/0.1-37			<0.001[b]
Balance – percentile	64	57.71 (23.68)/5-95	39	6.46 (8.18)/0.1-37			<0.001[b]
DCD-Q total score	64	64.7 (7.67)/43-75	39	33.28 (8.17)/18-54	20	44.8 (11.18)/26-63	<0.001[a]
SDQ total problem score	64	6.97 (4.95)/0-23	39	13.15 (5.53)/3-27	21	10 (5.31)/2-20	<0.001[a]
SDQ Impact (impact:no impact)	64	18:46	39	34:5	21	13:8	
SDQ Impact score	18	1.33 (2.14)/0-8	34	2.94 (2.15)/0-7	13	1.54 (1.33)/0-4	0.013[a]
Formal diagnosis of DCD (yes:no)			39	27:12			
Co-occuring diagnoses (N./%)							
AD(H)D	64	1/1.56%	39	4/10.25%	24	1/4.17%	0.057[c]
ASD	64	1/1.56%	39	8/20.51%	24	3/12.50%	0.005[c]
Dyslexia/speech fluency disorder	64	2/2.89%	39	1/2.56%	24	0/0%	0.506[c]
GMFCS level I:II (N.)					24	14:10	
Unilateral:Bilateral (N.)					24	13:11	

[a]Result of a One-Way ANOVA test; [b]result of an independent samples *t*-test between DCD and TD; [c]result of a Chi-squared Test; N.: total sample; BMI: Body Mass Index; TGMD-3: Test Of Gross Motor Development, 3rd ed.; MABC-2: Movement Assessment Battery for Children, 2nd ed.; DCD-Q: DCD-Questionnaire; SDQ: Strength and Difficulties Questionnaire; GMFCS: Gross Motor Function Classification System.

formal diagnosis, whereas the remaining 12 children met the inclusion criteria, but were not (yet) diagnosed. Among them, three children were included via assessment in their regular school, and the remaining nine children were registered in a diagnostic trajectory with suspected DCD. The sample is further described in Table II.

Balance performance

In the CP group, one 5-year-old could not perform the tasks in domain 4 and one 5-year-old with DCD could not reliably perform domain 5. These scores were not considered in their total Kids-BESTest score.

Group results

The interaction between group and age was not significant in any domain or for the total score (P=0.092-P=0.793) and therefore excluded from the ANCOVA analysis. Performance at older age was significantly higher compared to younger ages in domain 4 (η^2=0.15), in domain 6 (η^2=0.08), and in the total score (η^2=0.05) for all groups with a large, medium, and low effect for age, respectively. When accounting for age, children with TD scored significantly better on the total score and all domains compared with both DCD and CP children, with large effect sizes (η^2) varying from 0.25 (domain 2) to .66 (domain 3). The effect size for the total score was 0.71 (total score) (Table III, Figure 1, 2). Despite a large effect size for group in domains 2 (η^2=0.25) and 6 (η^2=0.54), post hoc testing revealed no significant differences between the DCD and CP group (P=0.999; P=0.012 respectively) (Table III, Figure 2).

Individual results

The p15 values based on our TD Kids-BESTest scores (explorative analysis) varied per domain/total score (Figure 3, 4). These values were used to assess whether the DCD and CP children performed as expected for their age. In the CP group, no child met the age-expected scores considering the total score, domains 1, 3, and 6. In DCD, not all but most children performed at or below expectations (≤p15) for the total score (36/39) and in domains 1 (35/39), 3 (34/39), and 6 (34/39). The distribution (number of children with a score >p15 or ≤p15) was not significantly different between DCD and CP (P>0.01). However, the distribution in domain 4, was significantly different (Chi-squared Test: P=0.002) with 20/39 DCD children performing below the age-expected score *versus* 22/24 children with CP.

TABLE III.—*Scores per domain and total score per group and results of ANCOVA and post-hoc testing.*

	TD (N.=64)		DCD (N.=39)	CP (N.=24)	Age			Group			Tukey HSD-group
	Mean (SD)	p15	Mean (SD)	Mean (SD)	F-value	P value	η^2	F-value	P value	η^2	
Domain 1 Biomechanical constraints	93.85% (6.21%)	89.67%	80.00% (8.52%)	71.39% (6.36%)	4.07	0.046	0.01	104.81	<0.001	0.61	TD-DCD <0.001 TD-CP<0.001 DCD-CP<0.001
Domain 2 Stability limits/ Verticality	77.53% (11.23%)	64.05%	64.32% (11.00%)	63.33% (13.79%)	5.29	0.023	0.03	21.41	<0.001	0.25	TD-DCD <0.001 TD-CP<0.001 DCD-CP=0.999
Domain 3 Anticipatory postural adjustments/ Transitions	88.09% (8.82%)	77.78%	62.96% (12.28%)	52.54% (12.75%)	0.49	0.487	0	120.73	<0.001	0.66	TD-DCD <0.001 TD-CP<0.001 DCD-CP=0.001
Domain 4 Reactive postural responses	84.29% (11.52%)	72.22%	68.55% (15.40%)	55.07% (17.15%)	40.91	<0.001	0.15	50.01	<0.001	0.37	TD-DCD <0.001 TD-CP<0.001 DCD-CP=0.002
Domain 5 Sensory orientation	95.21% (6.77%)	86.67%	84.91% (10.24%)	75.55% (21.23%)	0	0.979	0	26.07	<0.001	0.30	TD-DCD <0.001 TD-CP<0.001 DCD-CP=0.009
Domain 6 Stability in gait	81.28% (13.06%)	66.67%	56.24% (13.59%)	44.77% (13.64%)	26.87	<0.001	0.08	93.65	<0.001	0.54	TD-DCD <0.001 TD-CP<0.001 DCD-CP=0.012
TOTAL SCORE	86.71% (5.85%)	80.35%	69.40% (6.17%)	60.41% (7.84%)*	31.29	<0.001	0.05	216.94	<0.001	0.71	TD-DCD <0.001 TD-CP<0.001 DCD-CP<0.001

TD: typical development; DCD: developmental coordination disorder; CP: cerebral palsy; significant if P<0.01,
*performance for the unilateral group: 63.73% (7.29%), for the bilateral group: 57.10% (7.16%).

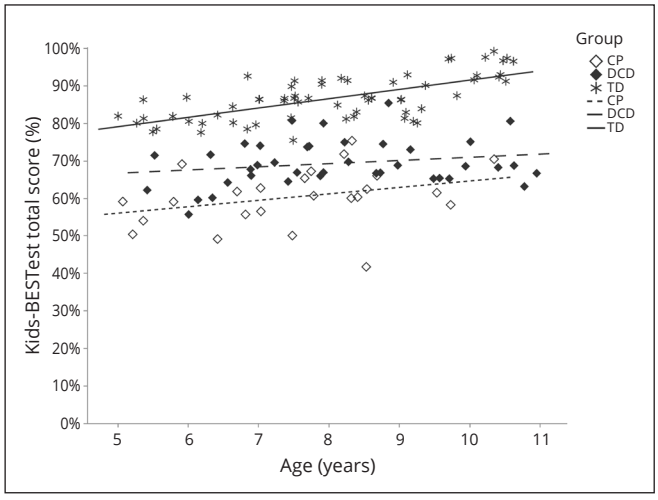

Figure 1.—Total kids-BESTest score *versus* age.
CP: cerebral palsy; DCD: developmental coordination disorder; TD: typical development.

Discussion

The aim of this study was to unravel the heterogeneity of balance performance among DCD, CP, and TD children. We formulated three hypotheses. First, we hypothesized that there is a continuum of balance performance shown by significantly different averages between groups but with overlapping individual results of DCD with both CP and TD. Second, we hypothesized that the group average of both DCD and CP children is significantly lower than that of TD children. Third, we hypothesized that both CP and DCD will show inter-individual and intra-individual variability considering the deficient postural control systems, with more heterogeneous results in the DCD group.

A continuum of balance performance

Children with DCD and CP performed below the TD scores on the total score and all test domains, confirming our second hypothesis. On the total score and the remaining test domains: biomechanical constraints, anticipatory postural adjustments, reactive postural responses, and sensory orientation, DCD children showed an intermediate score between TD and CP children, with individual results overlapping with those of both the TD and CP children. This confirms our first hypothesis of a continuum between DCD, CP and TD on their balance performance. Contrary, for scores on the domains stability limits/verticality and stability in gait, children with DCD and CP performed similarly, but both groups still showed heterogeneous results with some overlap with the TD group in these two domains. Children with DCD have poor balance performance but are generally less affected than children with CP, except for the latter mentioned domains.

The poor performance of children with DCD and CP on the tasks requiring anticipatory postural adjustments is in line with previous evidence.[7, 20-23] This suggests an inefficient use of the internal models in both groups, thereby

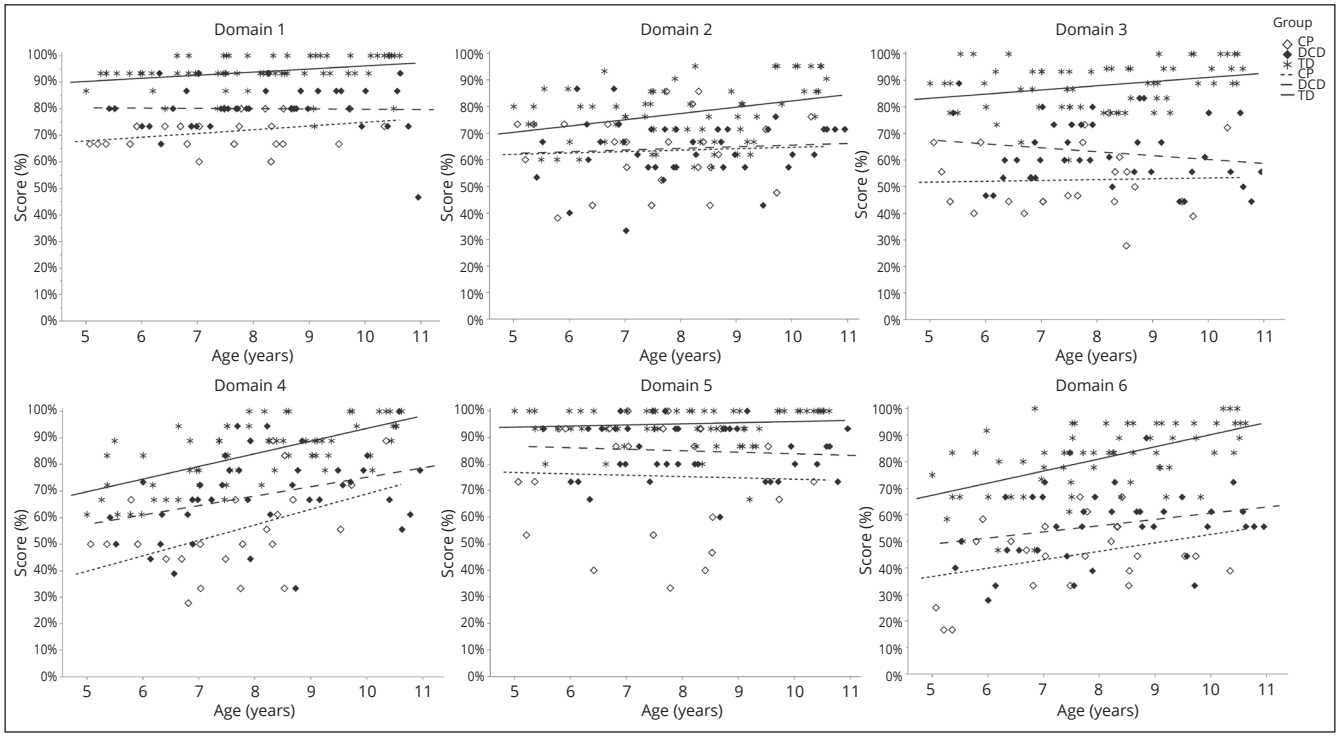

Figure 2.—Scatterplots of the Kids-BESTest domain scores *versus* age per group.
CP: cerebral palsy; DCD: developmental coordination disorder; TD: typical development.

subject	sex	age (years)	type	GMCFS	domain 1	domain 2	domain 3	domain 4	domain 5	domain 6	total score
cp 01	M	5.21	bilateral	2	⊗	⊗	⊗	⊘	⊗	⊗	⊗
cp 02	M	5.36	bilateral	2	⊗	●	⊗	⊗	⊗	⊗	⊗
cp 03	F	6.69	bilateral	2	⊗	●	⊗	⊗	●	⊗	⊗
cp 04	F	7.48	bilateral	2	⊗	⊗	⊗	⊗	⊗	⊗	⊗
cp 05	M	7.66	bilateral	2	⊗	⊗	⊗	⊗	●	⊗	⊗
cp 06	F	8.53	bilateral	2	⊗	⊗	⊗	⊗	⊗	⊗	⊗
cp 07	F	5.79	bilateral	1	⊗	⊗	⊗	⊗	●	⊗	⊗
cp 08	F	6.81	bilateral	1	⊗	●	⊗	⊗	⊗	⊗	⊗
cp 09	F	7.03	bilateral	1	⊗	⊗	⊗	⊗	⊗	⊗	⊗
cp 10	M	8.54	bilateral	1	⊗	⊗	⊗	⊗	⊗	⊗	⊗
cp 11	M	8.68	bilateral	1	⊗	⊗	⊗	⊗	●	⊗	⊗
cp 12	F	9.73	unilateral	2	⊗	⊗	⊗	⊗	⊗	⊗	⊗
cp 13	M	6.42	unilateral	2	⊗	⊗	⊗	⊗	⊗	⊗	⊗
cp 14	M	7.78	unilateral	2	⊗	●	⊗	⊗	⊗	⊗	⊗
cp 15	M	8.32	unilateral	2	⊗	⊗	⊗	⊗	●	⊗	⊗
cp 16	F	8.41	unilateral	2	⊗	●	⊗	⊗	⊗	⊗	⊗
cp 17	M	5.07	unilateral	1	⊗	●	⊗	⊗	⊗	⊗	⊗
cp 18	F	5.91	unilateral	1	⊗	●	⊗	⊗	●	⊗	⊗
cp 19	M	7.03	unilateral	1	⊗	●	⊗	⊗	●	⊗	⊗
cp 20	F	7.75	unilateral	1	⊗	●	⊗	⊗	●	⊗	⊗
cp 21	F	8.21	unilateral	1	⊗	●	⊗	⊗	⊗	⊗	⊗
cp 22	M	8.33	unilateral	1	⊗	●	⊗	●	●	⊗	⊗
cp 23	M	9.53	unilateral	1	⊗	●	⊗	⊗	⊗	⊗	⊗
cp 24	M	10.35	unilateral	1	⊗	●	⊗	●	⊗	⊗	⊗
				p15	89.67%	64.05%	77.78%	72.22%	86.67%	66.67%	80.35%

Figure 3.—Individual results of children with cerebral palsy per domain tested against the criterion of percentile 15 of the typically developing group.
M: male; F: female; GMFCS: Gross Motor Function.
p15: percentile 15 of the typically developing group; ⊗: test score ≤p15; ●: test score >p15; ⊘: test score not considered.

Figure 4.—Individual results of children with developmental coordination disorder per domain tested against the criterion of percentile 15 of the typically developing group
AD(H)D: attention-deficit/hyperactivity disorder; ASD: autism spectrum disorder.
M: male; F: female.
p15: percentile 15 of the typically developing group; ⊗: test score ≤p15; ●: test score >p15; ⊘: test score not considered.

subject	sex	age (years)	DCD diag.	other diag.	domain 1	domain 2	domain 3	domain 4	domain 5	domain 6	total score
dcd 01	M	5.42	yes	ASD	⊗	⊗	⊗	⊗	⊘	⊗	⊗
dcd 02	M	5.52	no	0	⊗	●	●	⊗	●	⊗	⊗
dcd 03	M	6.01	yes	0	⊗	⊗	⊗	●	⊗	⊗	⊗
dcd 04	F	6.14	yes	0	⊗	●	⊗	⊗	⊗	⊗	⊗
dcd 05	F	6.32	yes	0	●	●	⊗	⊗	●	⊗	⊗
dcd 06	M	6.34	yes	0	⊗	⊗	⊗	⊗	⊗	⊗	⊗
dcd 07	M	6.56	yes	ASD, ADHD	⊗	●	⊗	⊗	●	⊗	⊗
dcd 08	M	6.80	no	0	⊗	●	⊗	⊗	●	⊗	⊗
dcd 09	M	6.89	yes	0	⊗	●	⊗	⊗	⊗	⊗	⊗
dcd 10	M	6.89	no	0	⊗	●	⊗	⊗	●	⊗	⊗
dcd 11	F	6.98	no	0	⊗	●	⊗	⊗	⊗	⊗	⊗
dcd 12	M	7.02	no	0	●	⊗	●	●	●	●	⊗
dcd 13	M	7.23	no	0	⊗	⊗	⊗	⊗	⊗	⊗	⊗
dcd 14	M	7.42	no	0	⊗	⊗	⊗	●	⊗	⊗	⊗
dcd 15	M	7.48	no	ASD	⊗	●	⊗	●	●	●	●
dcd 16	M	7.55	yes	0	⊗	⊗	⊗	●	⊗	⊗	⊗
dcd 17	M	7.69	yes	ASD	⊗	⊗	⊗	●	●	⊗	⊗
dcd 18	M	7.72	no	0	⊗	●	⊗	●	⊗	⊗	⊗
dcd 19	M	7.88	no	0	⊗	⊗	⊗	⊗	●	⊗	⊗
dcd 20	M	7.92	yes	ADD	⊗	⊗	⊗	⊗	⊗	⊗	⊗
dcd 21	M	7.92	yes	ADD	⊗	●	●	●	●	⊗	⊗
dcd 22	F	8.22	yes	0	⊗	●	⊗	●	⊗	⊗	⊗
dcd 23	M	8.28	yes	0	●	⊗	⊗	⊗	⊗	●	⊗
dcd 24	M	8.67	yes	0	⊗	●	⊗	●	⊗	⊗	⊗
dcd 25	M	8.73	yes	ASD	⊗	●	⊗	⊗	●	⊗	⊗
dcd 26	M	8.77	yes	ADHD	⊗	⊗	●	●	●	⊗	⊗
dcd 27	M	8.85	no	0	⊗	●	●	●	●	●	●
dcd 28	F	8.98	yes	0	⊗	⊗	⊗	⊗	●	⊗	⊗
dcd 29	M	9.16	yes	ASD	⊗	⊗	⊗	⊗	●	⊗	⊗
dcd 30	M	9.49	yes	0	⊗	⊗	⊗	●	⊗	⊗	⊗
dcd 31	F	9.57	yes	0	⊗	●	⊗	●	⊗	⊗	⊗
dcd 32	M	9.71	yes	0	⊗	●	⊗	●	⊗	⊗	⊗
dcd 33	M	9.94	yes	dyslexia	⊗	⊗	⊗	●	⊗	⊗	⊗
dcd 34	M	10.01	yes	0	⊗	⊗	⊗	●	⊗	⊗	⊗
dcd 35	M	10.41	no	0	⊗	⊗	⊗	⊗	⊗	●	⊗
dcd 36	M	10.58	yes	ASD	⊗	●	⊗	●	⊗	⊗	●
dcd 37	M	10.63	no	0	●	●	⊗	⊗	⊗	⊗	⊗
dcd 38	M	10.78	yes	0	⊗	●	⊗	⊗	⊗	⊗	⊗
dcd 39	M	10.95	yes	ASD	⊗	●	⊗	●	●	⊗	⊗
				p15	89.67%	64.05%	77.78%	72.22%	86.67%	66.67%	80.35%

confirming the internal modeling deficit hypothesis deficit in DCD.[7, 39] This hypothesis suggests that children with DCD are compromised in using their internal models for adequate motor control, possibly because of their learning deficits.[39] Internal models are crucial for proper sensorymotor control. They predict the movement outcome by feedforward control before slower feedback-based control becomes available. Next, movements are monitored online by comparing the initial predicted outcome (feedforward control) with the actual sensory feedback of the environment (fast and slow feedback loops). In case of a mismatch, fast online adaptations are made, thereby fine-tuning the internal models. These internal models are developed and fine-tuned based on experience and learning.[39]

Appropriate preparation and monitoring of movement are particularly evaluated in the domain of anticipatory postural adjustments. Although we distinguished different domains evaluating a specific dominant postural control system, each test domain relies on the interaction of different postural control systems as the Kids-BESTest evaluates functional balance tasks.[16, 17] Hence, anticipatory postural adjustments are not only required to perform the tasks in the homonymous test domain but are also required for all voluntarily induced tasks.[18, 27] Children with DCD and CP indeed fail in performing the other test domains

requiring proper anticipatory postural adjustments, such as stability limits/verticality (*e.g.*, reaching forward), sensory orientation (*e.g.*, standing as stable as possible), and stability in gait (*e.g.* stepping over an obstacle). Besides anticipation, these domains require complex monitoring of ongoing movements in different situations by changing sensory information.

To further confirm the internal modeling deficit hypothesis, we expected that in more complex conditions, requiring more online monitoring or sensory integration in complex environments, these children would rely highly on compensatory mechanisms based on slow feedback-based control, as argued in the introduction.[39] Indeed, consistent with prior evidence in CP,[7, 21, 24] tasks with high end-point precision and high sensory demands, were the most difficult to perform for children with DCD and CP. For instance, more complex tasks in the test domain of anticipatory postural adjustments, such as alternate stair touching, which required precision, timing, and interlimb coordination, were more demanding than the sit to stand task, which required less precision and interlimb coordination. In addition, in the domains of sensory orientation and stability limits/verticality, both children with DCD and CP performed worse in high-demanding sensory conditions, when vision was occluded and/or somatosensory input was disturbed, such as standing on a foam surface with eyes closed, or leaning with eyes closed.[17]

While feedback (either fast or slow) plays a role in all tasks to some extent, the function of fast proprioceptive feedback was evaluated more isolated in the test domain of reactive postural responses.[17] Both, children with DCD and CP displayed impaired responses, even in these structured test circumstances, suggesting that these children cannot reliably use their fast proprioceptive feedback system. Two of the five tasks in this domain were performed in the backward direction, which did not allow visual input (in-place response and compensatory stepping – backward, Table I). These two tasks were the most challenging and were performed with greater difficulty compared with the tasks in the forward and lateral direction. Although previous research has shown contradictory findings, some studies confirmed that perturbations in the backward direction showed differences between DCD and TD children because of more inconsistent muscle activation timing, abnormal recruitment order, and longer onset latencies.[7, 40] Perhaps some children failed in these tasks because of a deficient proprioceptive system. We know that most children with CP have difficulties in their lower-limb proprioception, *e.g.* joint-position sense,[41] but in DCD evidence is conflicting.[42]

Children with DCD appear to exhibit more pronounced difficulties in the domains of stability limits/verticality and stability in gait, performing similarly to those with CP. Both domains are complex, requiring the integration of multiple postural control systems and constant online monitoring of movement and environment.[16, 17]

Specifically, the tasks in the domain of stability limits/verticality involve good feedforward mechanisms and well-developed orientation in space.[16, 17] The development of spatial orientation highly depends upon the complex multisensory integration of visual, vestibular, and somatosensory input, including proprioception.[43] Malfunctioning sensory input or sensory integration can lead to inadequate postural control.[16, 18] The vestibular system plays a key-role in tasks in this domain, especially when vision is obstructed, *e.g.*, during the leaning task.[16] In DCD, we found no study that directly measured vestibular function, but studies estimated its function indirectly, for instance, based on balance measures (*i.e.*, domain of sensory orientation).[7, 16] Consistent with previous evidence,[7, 44] we observed poor balance in children with DCD when standing still on a foam surface with their eyes closed, forcing them to rely more on their vestibular system (domain of sensory orientation). However, based on our results, we cannot distinguish whether difficulties in this domain can be attributed to peripheral (sensor organs and nerves) and/or central sensory integration (temporal and parietal cortex) problems. In DCD, we expect difficulties in the integration and reweighting of the sensory input.[5, 39, 44] However, in other developmental conditions, vestibular dysfunction is often overlooked.[45] Because of the large overlap between DCD and other neurodevelopmental conditions,[5] it is plausible that some children with DCD experienced undiagnosed vestibular dysfunctions in out sample. Vestibular dysfunction can also occur in children with CP.[24] Although we excluded children with known sensory disorders (vestibular, visual, or somatosensory) from our sample, vestibular function and proprioception were not systematically assessed. This may have led to overlooked vestibular or proprioceptive dysfunctions.

In the domain stability in gait, a combination of anticipatory postural adjustments, reactive responses, and fast online integration of the available sensory information is necessary to perform complex walking activities.[46, 47] The submaximal scores of both DCD and CP imply that these children cannot adequately adapt their gait to different challenging conditions, such as when avoiding an obstacle, walking with head turns, or performing the Timed Up and Go test. Evidence in CP is quite consistent with large

effects showing differences with TD children, such as longer double support times, shorter step-lengths, or slower walking speed,[48] but in DCD, evidence is much less conclusive.[7] For instance, although stride length was not different between DCD and TD during level walking on a ground surface, the stride length was significantly shorter on a treadmill.[7] In our study, we observed differences between the DCD and CP groups and the TD group already for level walking. This increased with increasing task type difficulty (more anticipation, reaction and complex online monitoring), such as during obstacle crossing or when performing horizontal head-turns.

Age-effects

Interestingly, although the interaction effect of age*group was not significant, visually (Figure 1, 2), both the CP and TD children performed better at older ages compared with the younger children. However, this trend was not observed in the DCD group. At older age, the differences between children with TD and DCD seem to become larger (and become smaller with CP). These findings contrast the hypothesis of DCD showing a developmental delay, where performances of older children with DCD are similar to younger TD children instead of age-peers.[14, 44, 49] This hypothesis suggests that children with DCD may catch up with their age-peers over time when longitudinally followed up. Rather, our results are in line with the hypothesis that DCD shows a deviant development, meaning that there is a fundamental neurological basis in DCD explaining their postural control difficulties.[44, 50] However, these hypotheses are based merely on cross-sectional data, instead of longitudinal data, and the underlying reasons remain unexplored.[14, 44, 49, 50]

Individual balance performance

We can confirm our third hypothesis (Figure 3, 4) that both inter- and intra-individual variability existed within both the DCD and CP group. In both groups, inter-individual variability was observed. Most children performed below expectations in the domains of biomechanical constraints, anticipatory postural adjustments, and stability in gait. There was a larger inter-individual variability in the other domains. Intra-individual variability between the domain scores was also present in both groups. For instance, in one domain, a child can perform similarly to TD peers, while in another domain, the child's performance falls significantly below TD expectations. With our results, we cannot explain this heterogeneity (*i.e.*, inter- and intra-individual variability). The heterogeneity is potentially related to the severity of the disorder. In CP, for instance, postural control is usually related to the GMFCS level or the type of CP (uni-/bilateral),[20, 21] but this could not be confirmed visually and was not the scope of this study. In DCD, however, there is no consensus on the definition of the severity, but the severity may be related to specific deficits in neurological mechanisms, underlying the balance domains.[17, 44] Furthermore, heterogeneity may be induced by the amount and/or type of co-occurring developmental conditions,[14] such as AD(H)D and ASD. We could not statistically confirm this but visually our results do not confirm this. All hypotheses warrant further research.

Clinical implications

Since each postural control system is required for adequate balance performance, the observed inter- and intra-individual heterogeneity implies that comprehensive evaluation is needed for individualized approaches in pediatric physiotherapeutic rehabilitation, which is possible with the Kids-BESTest. Based on the Kids-BESTest results, therapists can identify the individual deficient underlying postural control systems and use these estimates to establish a physiotherapeutic diagnosis facilitating therapy planning. Subsequently, during therapy, the individual deficient systems can be improved by targeting these systems in a variety of functional activities.[5, 51, 52] For instance, a child performs poorly in the test domain of stability limits/verticality. Therefore, during therapy, the physiotherapists can then stimulate the child to perform reaching and leaning activities in different directions (toward their limits of stability) with and without eyes closed during activities the child likes, such as playing soccer, cycling, or gaming. These findings may open new horizons for group therapy interventions focusing on the request for help (*i.e.* balance deficits), regardless of the underlying deficit, thereby allowing for a mix of children with different types of pathologies.

Limitations of the study

This study had some limitations. The children with DCD and CP could not be age-matched, we therefore created three groups that were balanced for age (*i.e.*, comparable based on the mean (no statistical difference) and age range). We partly corrected for this potential bias by adding age as a covariate, but there was an underrepresentation of 10-year-old CP children (N.=1). We included children with spastic CP based on their GMFCS scores. However, the group consisted of a variety of specific brain lesions and a combination of bilateral and unilateral types. Future

studies with larger samples may investigate postural control differences related to clinical types and the specific underlying lesions. Children with GMFCS III were allowed if they were able to stand independently and walk a short distance without aids. However, none were included. Since the original BESTest was developed from the knowledge of basic research on postural control and despite it being a proposal for a revolutionary balance assessment scale, some researchers have questioned the structure of the scale in adult populations. Hence, further research into the structural validity of the Kids-BESTest is necessary.

Future research

Future research must unravel the specific relationship between balance performance and underlying control mechanisms (*i.e.* internal modelling deficit hypothesis or involvement of the mirror neuron system and the extent to which compensatory processes and strategies are employed) to understand the heterogeneity of balance performance in DCD. This can be achieved by using techniques measuring real-time brain-body interactions by combining electromyography with real-time brain imaging techniques such as electroencephalography, functional Near Infrared Spectroscopy, or a combination of both while performing balance tasks. To unravel whether our findings imply that the DCD group shows a different balance development compared to children with TD and CP, and if so, which mechanisms are at play, must be confirmed in longitudinal designs, preferably with age-matched groups.

Conclusions

This study supports a continuum of balance performance between children with TD, DCD and CP. Children with DCD show an intermediate score between TD and CP on their overall balance performance and on most balance domains. The poor performances on anticipatory postural adjustments, stability in gait, sensory orientation, stability limits/verticality, and reactive postural responses confirm an internal modeling deficit in DCD and CP. Still, there is a large heterogeneity of balance performance within and across domains, leading to inter- and intra-individual variability. This implies that comprehensive balance assessment is warranted to guide physiotherapeutic therapy. Future research should unravel the underlying control mechanisms that may explain the heterogeneity of the results using combined techniques that measure behavior, movement strategies, and real-time brain imaging to understand the body-brain interactions.

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Conflicts of interest

The authors certify that there is no conflict of interest with any financial organization regarding the material discussed in the manuscript.

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Authors' contributions

Evi Verbecque and Katrijn Klingels join last authorship. All authors meet the following 4 criteria: 1) substantial contributions to the conception or design of the work; or the acquisition, analysis, or interpretation of data for the work; 2) drafting the work or revising it critically for important intellectual content; 3) final approval of the version to be published; 4) agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All authors read and approved the final version of the manuscript.

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