

kinesitherapie

Masterthesis

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Faculteit Revalidatiewetenschappen

master in de revalidatiewetenschappen en de

Comparing balance deficits between children with Developmental Coordination Disorder, Cerebral Palsy and typically developing children: a case-control study

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

COPROMOTOR : Prof. dr. Evi VERBECQUE

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Comparing balance deficits between children with Developmental Coordination Disorder, Cerebral Palsy and typically developing children: a case-control study

What differences do children with DCD age 5 to 10 years exhibit versus CP and TDC on the total Kids-BESTest score? And what differences do these children with DCD show versus CP and TDC on the different domains of the Kids-BEST test?

Which domains linked to underlying mechanisms are weakest and strongest in the DCD group?

Highlights:

- Children with Developmental Coordination Disorder (DCD) score generally worse on balance tasks compared to typically developing children.
- Children with DCD seem to score better overall than children with CP on the Kids-Best test although this difference in score is not significant.
- Domain VI '*stability in gait'* is the only domain showing a significant score difference between all 3 groups.
- Further research is needed to ensure generalizability to confirm these results, preferably with a larger sample and a different study design such as a cohort study.

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Prof. dr. Evi Verbeque	Yinthe Kaes
Dra. Charlotte Johnson	Karlien Machiels

Research context

This case-control duo thesis is part of an ongoing Ph.D. project of Charlotte Johnson (C.J.) funded by The Research Foundation - Flanders (FWO): 'Understanding the heterogeneity of balance control in children with Developmental Coordination Disorder and its impact on motor performance: a synergistic approach using brain imaging, neuromechanics, and functional assessments'. This study was approved by the medical ethical committee of UZA-UAntwerpen and Uhasselt. (B300201941833).

The main aim of this joint Ph.D. (UAntwerpen – UHasselt) is to understand the heterogeneity of Developmental Coordination Disorder (DCD) by studying postural control and its underlying mechanisms. This project is located within the pediatric domain of REVAL's rehabilitation research center. Our thesis focuses on the first part of this project, understanding the heterogeneity of DCD by investigating postural control. In this thesis, balance is examined in comparison to children with Cerebral Palsy (CP) and typically developing children (TDC) through the Kids Balance Evaluation Systems Test (Kids-BESTest). The Kids-BESTest is a conceptual framework that evaluates all underlying systems of postural adjustments and transitions, reactive postural responses, sensory orientation, and gait stability). In Master 1, we received training on the modified version of the Kids-BESTestest (E. Verbecque) and also practiced with children for another study within the rehabilitation sciences framework. Also, in Master 1 the duo of this thesis co-practiced some test sessions with C.J. within the framework of her Ph.D. project of which our thesis is a part.

The purpose of the research was determined by supervisors E. Verbecque, K. Klingels, and mentor C. Johnson. A research question with sub-questions was then developed in consultation between the students. The implementation of the protocol and data collection was done by Dra. C.J. and written down by the students in the method. Raw data of item, domain, and total scores were obtained by Dra. C.J. These measurements of the Kids- BESTest consisted of a two-hour testing session completed by Dra. C.J. herself with assistance from students. The following assessments were administered during this session: Kids-BESTestest; Test of Gross Motor Development, 3rd edition; Neuromechanical analysis (accelerometers and surface electromyography); Functional Near-Infrared Spectroscopy. The final scoring of

the Kids-BESTest, used in this study, was done by Dra. C.J. who is experienced in this field. Processing of the results was done by the students using the statistical program SPSS.

Balance problems are common in children with CP and DCD. However, little is known about. the underlying cause of balance problems as well as the exact differences and/or similarities within the entire balance construct of typically developing children, children with DCD, and CP. The comparison of these three target groups has not been studied within a research study, therefore this doctoral study is unique in this field of research.

The introduction and method were written by both students Yinthe Kaes (Y.K.) and Karlien Machiels (K.M.). Both students reflected on the statistics and in consultation with Dra. C.J. and E. Verbecque, a conclusion was made about the best-suited statistics choice. Y.K. performed the statistics (with IBM SPSS statistics) and obtained the results. These results were written down in the thesis by both students. The discussion was thought about separately. The discussion was written by Y.K. with help from K.M. Abstract was written by K.M. and the conclusion by Y.K.

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1. Abstract

Background: Developmental Coordination disorder (DCD) is a condition with various motor deficits in which postural control is an important one. We want to compare DCD to other populations because little is known about their postural control as opposed to children with for example Cerebral Palsy (CP). Few comparisons have been made between DCD and other disorders, considering the entire construct of postural control. The purpose of this study is to examine postural control with the Balance Evaluation Systems test for Children (Kids-BESTest) by establishing a comparison between DCD, CP and, typically developing children (TDC).

Participants: The study consisted of 86 participants including 20 children with DCD, 12 children with CP, and 54 TDC, ages 5 to 10 years old. The diagnosis of DCD was verified following DSM-5 criteria. The diagnosis of CP was classified using the Gross Motor Function Classification System level I or II and TDC were screened using the Movement Assessment Battery for Children - Second edition.

Measurements: To examine postural control, the modified version of the Kids-BESTest was used. It consists of 31 items across 6 different domains and each item is given a score between 0-3. These are converted to percentages for domain and total scores. We compared these final scores between the 3 groups with a Multivariate Analysis of Variance (MANOVA).

Results: TDC obtained scores above 80% indicating normal balance. Children with DCD and CP scored below average and significantly weaker than TDC (DCD vs TDC: p < 0.001; CP vs TDC: p <0.001) on total scores. Across domains, children with DCD scored better but not significantly better than children with CP. Exceptions applied to two domains, (II) *limits of stability and verticality* and (V) *sensory orientation* where the CP group scored better but also not significantly better than children with DCD.

Conclusion: DCD and CP children have a performance under the cut-off value of balance performance measured with the Kids-BESTest. DCD differs significantly from TDC children but not from CP children, although this is not the case in all domains. Further research into the existence of a balance continuum is necessary.

Keywords: Developmental Coordination Disorder (DCD); Cerebral Palsy (CP); typical developing children (TDC); postural control; Kids-BESTest

2. Introduction

Developmental Coordination Disorder (DCD) is an idiopathic disorder with a prevalence of 5-6% of school-aged children (American Psychiatric Association & Association, 2013; Blank et al., 2019). Typically, DCD is characterized by a delay in performing and acquiring motor skills severely impacting daily life activities. These problems in motor performance mainly present themselves in gross and fine motor skills, balance control, and coordination (Wilson et al., 2013). Problems are also encountered in sensorimotor aspects, such as proprioceptive functions, visuomotor perception, sensory integration, and motor planning (Wilson et al., 2013). DCD often occurs in association with one or more neurodevelopmental and neurobehavioral disorders (Lingam et al., 2010). Common comorbidities include Attention-Deficit/Hyperactivity-Disorder (ADHD), Autism Spectrum Disorder (ASD), speech disorders, learning disabilities, and language disorders. It should considered that ADHD, ASD or others may negatively affect motor performance on assessments, complicating the interpretation of the findings (Blank et al., 2019).

Up to 60-87% of children with DCD experience balance deficits, measured by the one-leg stance test, compared to typically developing children (TDC) (Geuze, 2005; Van der Linde et al., 2015; Verbecque et al., 2021). Postural control is the ability to control the center of mass to the base of support (BOS). Knowing one's limits of stability is a key component in maintaining balance and being resilient to perturbations and using movement strategies (ankle, hip, or stepping strategy) to overcome them in both static and dynamic situations. However, postural control is much more complex, comprising many underlying mechanisms (Shumway-Cook & Woollacott, 2014). Horak et al., 2009 created a postural control framework, distinguishing six domains of balance. The first domain, constraints on the biomechanical system, refers to weaknesses such as a weak ankle/hip or flexed posture that limits the ability to use compensatory mechanisms in postural control. Limits of stability are defined as how far the center of mass of the body can be moved across the base of support. Verticality is a representation of upright gravity. Anticipatory postural adjustments are active movements of the body's center of mass in anticipation of a voluntary movement caused by feedforward projections. *Reactive postural control* refers to the body's response to a perturbation such as a push, slip, or trip. Short, medium, and long proprioceptive feedback loops should be automatically engaged in this mechanism. The domain of sensory integration, looks at integration of sensory information and converting it into spatial information. Limitations result in disorientation/instability when the support surface/visual environment moves. The ability to catch the 'falling' center of mass of the body during walking (changing foot or changing in the base of support) is referred to as *stability in gait* (Horak et al., 2009).

The systematic review by Verbecque et al. 2021 discusses balance deficits in these domains between typically developing children (TDC) and children with DCD. In *functional stability and verticality*, DCD children score as well as TDC children except in their posterior stability limits. In *Transitions and anticipatory postural adjustments*, deficits were demonstrated during limb movement in the anterior direction. Also, large differences were found in the timed single-leg stance, at a disadvantage for children with DCD. *Reactive postural control* shows conflicting results. Findings in *sensory integration* show that even though the DCD population has reduced balance control and additionally a larger sway area and sway path, they still exhibit a similar movement strategy as TDC while keeping the eyes open in the natural position. Similar findings were reported in standing on a stable surface with visual disturbances. However, differences have been observed, showing greater use of a hip strategy and postural swing during a closed-eye position. No consistent differences were found in step time and mass center parameters for *stability in gait* (Verbecque et al., 2021).

Characteristics of poor postural control in children with DCD are likely to be task dependent. Several factors affect the quality of postural control, particularly the difficulty of the task and the availability of sensory information (Geuze, 2005). Although we know that children with DCD show postural control deficits in different domains, none of the existing studies examined this based on the entire construct of balance (Verbecque et al., 2021). One test comprising all postural control domains is the Balance Evaluation Systems test for Children (Kids-BESTest), which is currently one of the most comprehensive assessment tools to evaluate postural control deficits in children with CP, the kids-BESTest is a reliable, valid, reproducible and feasible test (Dewar et al., 2017; Dewar et al., 2019). CP is a neuromotor disorder caused by lesions or abnormalities in the brain. It is a permanent disorder in which there are motor and/or a variety of other limitations, including balance construct by the Kids-BESTest, it is stated that especially domains III (anticipatory postural adjustments and transitions) and IV (reactive

postural control) are difficult as well as complex tasks such as walking (VI: stability in gait) (Carlberg & Hadders-Algra, 2005; Sílvia Leticia Pavão et al., 2013). Children with CP and DCD both show similar difficulties in motor planning processes, yet the relationship/cause of these similar processes and motor problems between DCD and CP is not known (Williams et al., 2014; Wilson et al., 2017). It is currently argued that DCD belongs somewhere on the continuum between TDC and CP in terms of balance skills. However, this continuum needs to be examined more closely to confirm this statement in more detail. To gain more insight into the heterogeneity of DCD, balance performance of these three groups (DCD, TD & CP) needs to be compared. Therefore, this study aims to compare DCD children with TDC and children with CP on the whole construct of balance, based on the following research question: How do children with DCD aged 5 to 10 years differ from TDC and children with CP on various balancing tasks measured with the Kids BESTest? We hypothesize that TDC show normal postural control. In the total score, we expect that children with DCD will perform slightly better and thus rank higher on the continuum than CP children. Because of poor motor planning in children with DCD and CP, we expect poor scores in the domain of transitions and anticipatory postural adjustments (III). Also, reactive postural control (IV) and stability in gait (VI) are hypothesized to score weak because these involve complex balancing tasks. When these results are known, we can make a clear continuum for each domain and total scores and create more insight into which domain is most affected within each group to create appropriate treatment programs in the clinical setting.

3. Methods

In this case-control study, we assess balance performance in children with DCD, children with CP, and TDC using the Kids-BESTest task, domain, and total scores. This study was approved by the medical ethical committee of UZA-UAntwerpen and Uhasselt (B300201941833).

3.1. Research question

The purpose of this study is to gain more insight into the continuum between DCD, CP, and TDC in terms of postural control. We seek answers to the following questions: 1) What differences do DCD children exhibit versus CP and TDC children on the total Kids-BESTest score? 2) What differences do DCD children show versus CP and TDC on the different domains of the Kids-BEST test? 3) Which domains linked to underlying mechanisms are weakest and strongest in the DCD group?

3.2. Participants

3.2.1. Recruitment and selection criteria

We recruited children between 5 years 0 months to 10 years 1 month old. To minimize the hormonal influence on motor performance seen in children in puberty we exclude children older than 10 years (Brix et al., 2019). Before participation, parents completed the DCD questionnaire (DCD-Q) (Wilson et al., 2009), and all children, except those in the CP group, were evaluated with the Movement Assessment Battery for Children, 2nd edition (MABC-2) (Brown, 2013) to confirm eligibility. The MABC-2 was not performed in CP children because it does not give an accurate picture of the child taking into account the disorder. Two common comorbidities within the DCD group (AD(H)D and ASD) were screened within our study for all groups (Green et al., 2006; Kadesjö & Gillberg, 1998; Wisdom et al., 2007). If other comorbidities were present, this was also mentioned in a non-specific group. The informed written consent and a general questionnaire were obtained from all participants and parents prior to participation.

Children were recruited in Flanders (Belgium) by handing out flyers in schools, private practices, and hospitals, or through a network of researchers and internships. Thus, the recruitment was done by voluntary participation.

Children in the **DCD group** were eligible for inclusion if (1) they had a confirmed diagnosis of DCD according to the DSM-5 criteria (American Psychiatric Association & Association, 2013): (criterion A) objectified by the MABC-2, total percentile score of $\leq 16^{\text{th}}$; (criterion B) confirmed when referred to a pediatric physical therapist for motor training, in which the DCD Questionnaire (DCD-Q) will be used to further assess interference of the motor impairment with daily activities and/or academic achievement); (criterion C) onset of symptoms in the early developmental period as evidenced by their referral to a center for motor training and (criterion D) absence of any medical condition that could cause the motor impairment and an IQ \geq 70. (2) Comorbidities such as ADHD, ASD, dyslexia, etc. were allowed. Children were excluded when there were: (1) medical conditions such as intellectual delays (IQ<70), visual or vestibular impairments, neurological conditions (e.g. CP, muscular dystrophy...); (2) signs of puberty.

Children in the **CP group** were included when they (1) have predominant spastic CP; (2) Gross Motor Function Classification Scale (GMFCS) level I or II (ambulant, can walk >6 meters independently); (3) minimal intellectual level to understand verbal instructions. Children were excluded if (1) they showed an ataxic or dyskinetic type of CP; (2) GMFCS level 3-5 (cannot walk 6 meters independently); (3) intellectual delay judged by the parents; (4) signs of puberty.

The inclusion criteria for the **TDC group** were: (1) born >37 weeks of gestation; (2) score >p25 on the MABC-2 (a standardized score confirming typical motor performance). Children were excluded if there were (1) any medical conditions such as intellectual delays (IQ<70), visual or vestibular impairments, neurological conditions (CP, muscular dystrophy,...), diagnosis of DCD, or other medical conditions that might impede balance control; (2) signs of puberty.

3.2.2. Power and sample size

To aim for a power of 80%, we used the program G*power to calculate how large our sample should be. We used an "a priori" type of power analysis (Faul et al., 2007) with means of TDC vs DCD, TDC vs CP, and DCD vs CP. To measure differences between TDC vs DCD and TDC vs CP, there should only be four participants in each group, which we thought was far too few. We opted for the largest sample size obtained (DCD vs CP), which was 12 participants per group, resulting in a sample of at least 36 participants.

3.3. Procedure

3.3.1. The Kids-BESTest

Postural control of children with DCD was assessed using the Kids-BESTest according to the protocol published by Dewar et al., 2017 (Dewar et al., 2017). We used a version of this Kids-BESTest that was recently adapted by our research group in collaboration with the research group of Prof. L Johnstone (Appendix 2). This version takes age-specific needs into account, comprising five different versions per age band: 5, 6, 7, 8-10 years, and 11-14 years. These age-adjusted versions made the test more sensitive for detecting balance deficits. Also, specific qualitative descriptors have been added in this version, giving a more detailed observation of the item performed. Unpublished results tell us that the age-adjusted version is reliable but validity has yet to be proven. As in the original kids-BESTest, each item (considering age-specific needs) was scored from 0 (worst performance) to 3 (best performance). All item scores were added together, making a total score of 0-108. A 2nd total score was also calculated, this time without including domain I (biomechanical constraints) in the calculation. By adding up all domain items, a domain score was calculated for each of the six domains. Then these total and domain scores were converted to percentages. A cutoff value of 80% is applied in the assessment of the Kids-BESTest. If the total score is below this value, it is useful to look at specific domain scores where scores are also below 80%. The domains of the Kids-BEST test were randomized in advance without changing the item order within a domain. The randomization was blinded by a sealed envelope for the principal investigator.

The first domain of the test is *biomechanical constraints* (domain I), which includes items such as the base of support (BOS), center of mass (COM) alignment, ankle strategy and range of motion (ROM), hip/trunk lateral strength, sit on the floor and stand up. The domain of *limits of stability and verticality* (domain II) includes lateral lean (R+L) and verticality, also functional reach forward and lateral will be tested. Items such as sit-to-stance, rise-to-toes, standing arm raise, one-leg stance, and alternate stair touch are included in the domain of *transitions and anticipatory postural adjustments* (domain III). For the *reactive postural response* (domain IV) domain, in-place responses for both forward and backward were tested. The item compensatory stepping strategy was tested forward, backward, and lateral. *Sensory integration* (domain V) was tested by incline eyes closed and a clinical test of sensory interaction on balance (CT-SIB). CT-SIB was performed on a firm surface and a foam surface, both with eyes opened and closed. Lastly, for the domain of *gait stability* (domain VI), gait on level surface, change in gait speed, walking with head turns, walking with pivot turns, stepping over an obstacle, and Timed up and go are tested. All the items with their domains are listed in Table 1.

Table 1

Biomechanical constraints	Limits of stability and verticality	Transitions and anticipatory postural adjustments	Reactive postural responses	Sensory integration	Stability in gait
Base of support	Lateral lean	Sit to stand	In place response – forward	Clinical test of sensory interaction on balance (CT-SIB)	Gait on level surface
Center of mass alignment	Verticality	Rise to toes	In place response – backward	Incline with eyes closed	Change in gait speed
Ankle strength and range of motion	Functional reach — forward	One leg stance	Compensatory stepping – forward		Walking with head turns – horizontal
Hip/trunk lateral strength	Functional reach — lateral	Alternate stair touch	Compensatory stepping – backward		Walking with pivot turns
Sit on the floor and stand up		Standing arm raise	Compensatory stepping – lateral		Step over obstacle
					Timed up and go test (TUG)

Overview Items of each Domain of The Kids-BESTest

Note. Kids-BESTest: Balance Evaluation System Test for Children

3.3.2. Procedure

Testing took place in standardized testing rooms such as labs, and schools,... with no or limited distractions. The testing space should be large enough to accommodate walking tasks of 6 meters. Parents could opt for a test administration on the University campus (UAntwerp or UHasselt) or at their home/school if it meets space requirements as stated above. On campus, the evaluation of the Kids-BESTest is part of an approximately two- to three-hour testing session. At the beginning of the examination, the child's height and weight were documented. The examination had to be completed barefoot; no aids/orthotics were allowed. Short breaks were taken if necessary to ensure attention, motivation, and cooperation. Only one child was

tested at a time and the child's parents were asked to not be present. All tests were administered by an experienced physical therapist within the research team. One or more additional investigators were present to help set up the equipment and ensure the child's safety. However, the scoring of the tests was only conducted by one experienced researcher.

3.4. Data analysis

The domain and total scores were used for statistical analysis. By using the formula "achieved score / maximal score possible * 100", percentages were calculated for each domain separately as well as the total percentage for all domains. A correction was made for item scores if they did not occur in a particular age band by removing them from the domain score.

A one-way ANOVA was chosen for the comparison of characteristics such as age, MABC-2 score, and gestational age between the 3 groups with a significance level of p < 0.05. The chi-square test was used to measure the distribution between the groups for gender, prematurity, and comorbidities where the significance level was also set at p < 0.05.

Differences between groups were analyzed with A multivariate analysis of variance (MANOVA) was chosen to see if there is a significant difference between two or more groups with additionally a posthoc Bonferroni test to clarify which groups differ significantly from each other. A significance level of p < 0.05 was used. The assumptions and statistics for a one-way MANOVA were checked through IBM SPSS Statistics (version 28).

The following assumptions were met; multivariate normality, outliers, and multicollinearity. In MANOVA, the normality of each of the dependent variables for each of the groups of the independent variable (univariate normality) was used as the best "estimate" of multivariate normality. The Shapiro-Wilk test was used for this purpose. To check for univariate outliers box plots were used (Figure 1). Multivariate outliers were checked using Mahalanobis distance. Some univariate outliers were present but no multivariate outliers. There was no multicollinearity because all Pearson correlations were between 0.2 and 0.9.

The assumption for homogeneity of variance-covariance matrices was checked by looking at Box's M-test for equality of covariance but was not met. By performing Levene's test we obtained that the cause of this was the domain of *sensory orientation* (V) because it has a ceiling effect. The rest of the dependent variables did meet Levene's test. The assumption regarding the linear relationship of each pair of dependent variables for each group of the independent variable is unclear. Thus, we considered that if the variables are not linearly related, the power of the test decreases.

Because several assumptions were not met, we followed Pillai's trace in the multivariate test of MANOVA.

4. Results

4.1. Participants

In the TDC group, nine children were excluded from our analysis because they obtained too low MABC-2 percentile scores (<p16). One child from the DCD group was also excluded because he obtained a percentile score on the MABC-2 test above 16. After excluding these children, we included a total of 88 participants, 20 of whom were children with DCD, 12 were children with CP, and 54 were TD children. Overall more boys (56) than girls (32) were included in the study as well as in each group. Age has an equal distribution among the three groups (p = 0.360) and ranges in each group from 5 to 10 years old. MABC-2 scores (*M TDC* = 51.26; *M DCD* = 1.99) differed significantly between the TDC and DCD groups (p < 0.001). The number of additional comorbidities is especially more present in the DCD (53.85%) followed by the CP (50%) group but the sample size is also smaller compared with TDC (6.38%). For CP, equal numbers of children with GMFCS I (4) and GMFCS II (4) were included.

More details about the demographic and characteristics of the participants can be found in Table 1.

4.2. Balance performance

The mean domain and total scores along with standard deviations, and minimum and maximum scores are listed in Table 2. Boxplots of each group are also presented in Figure 1.

A MANOVA showed a significant difference between the groups (TDC, DCD, and CP) (F = 7.305; p < 0.001). An additional Bonferroni posthoc test shows that both the DCD and CP group show deviant total scores (below 80%) and scored significantly lower compared to the TDC (DCD vs. TDC: p < 0.001; CP vs. TDC: p < 0.001). However, no significant difference was found between the DCD and CP group (p = 0.102). The TDC and DCD groups show a fairly small standard deviation.

We also looked at the total score without including domain I "biomechanical constraints". A significant difference was yet again found between TDC and DCD (p < 0.001) as well as between TDC and CP (p < 0.001). No differences were found between DCD and CP (p = 0.261).

As expected the mean scores of all domains show that TDC score best compared to children with DCD and CP. For domain, (I) *biomechanical constraints*, (III) *transitions and anticipatory postural adjustments*, (IV) *reactive postural responses* and *stability in gait* (VI) the means of children with DCD show to be better than CP. For the following domains, a significant difference was found between TDC and DCD children and between TDC and CP children but no significant difference was found between DCD and CP children: (I) *biomechanical constraints* (TDC vs. DCD: p <0.001; TDC vs. CP: p < 0.001; DCD vs. CP: p = 0.082), (III) *transitions and anticipatory postural adjustments* (TDC vs. DCD: p < 0.001; TDC vs. CP: p < 0.001; TDC vs. CP: p = 0.001; TDC vs. CP: p < 0.001; DCD vs. CP: p = 0.001; TDC vs. CP: p < 0.001; DCD vs. CP: p = 0.001; TDC vs. CP: p < 0.001; DCD vs. CP: p = 0.375).

In the domain (II) *limits of stability and verticality* there is only a significant difference between TDC and DCD (p < 0.001). The comparison of TDC vs. CP (p = 0.108) gave no significant difference as did DCD vs CP (p = 1.000). The difference in this domain versus the previous ones is in the comparison between children with DCD and CP where children with DCD scored lower than children with CP.

Domain V, *sensory orientation*, was the only domain in which all groups scored above age expectancy (>80%) with typically developing children scoring best, followed by children with DCD and CP. A significant difference was found between TD and DCD children (p <0.001) and between TD and CP children (p <0.001). But no significant difference between DCD and CP children (p=1.000).

Lastly, in the domain of (VI) *stability in gait,* all groups differed significantly from each other (TDC vs. DCD: p <0.001; TDC vs. CP: p <0.001; DCD vs. CP: p = 0.020).

Table 2

	TDC group (N=54)						DCD group (N=20)					CP group (N=12)				
	Ν	Mean	SD	Min	Max	N	Mean	SD	Min	Max	N	Mean	SD	Min	Max	
Age (years)	53	8.12		5.00	10.97	20	8.23		5.52	10.95	12	7.44		5.07	10.35	0.360*
MABC-2 (percentiles)	54	51.26	25.11	16.00	95.00	19	1.99	16.92	0.10	9.00						< 0.001*
Gestational age (weeks)	47	39.81	1.77	37.00	48.00	19	38.68	2.11	34.00	42.00	12	36.58	3.26	32.00	40.00	< 0.001*
Gender																0.055**
Boys	31					17					8					
Girls	25					3					4					
Prematurity																< 0.001**
>37 weeks	47					16					6					
<37 weeks	0					4					6					
Comorbidities																0.048**
None	47					13					8					
AD(H)D	1					3					2					
ASD	1					3					2					
Other	1					1					0					
GMFCS 1											4					
GMFCS 2											4					
AFO's											7					

Summary of Participant Characteristics

Note. * one-way ANOVA; ** chi-square; TDC : Typically developing children; DCD: Developmental Coordination Disorder; CP: Cerebral Palsy; MABC-2: Movement Assessment Battery for children, 2nd edition; AD(H)D: Attention-Deficit/Hyperactivity-Disorder; ASD: Autism Spectrum Disorder; GMFCS: Gross motor function classification system; AFO: Ankle-foot orthosis; SD: standard deviation; Min: minimum; Max: maximum

Table 3

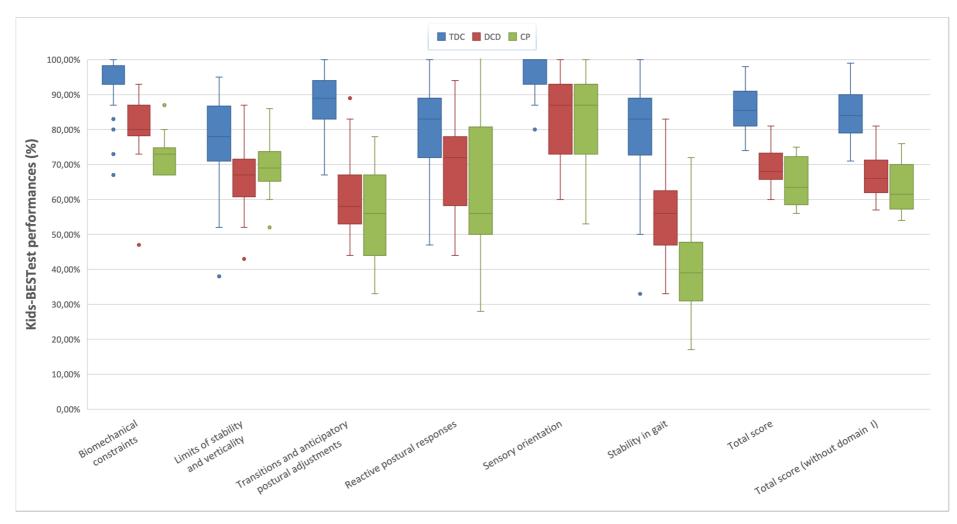
Overview Performances (%) Kids-BESTest

		TDC grou	up (N=53)			DCD gro	up (N=20)		CP group (N=12)				
			P-v	alue	P-value						P-value		
	Mean	SD	TDC vs.DCD	TDC vs.CP	Mean	SD	DCD vs. TDC	DCD vs. CP	Mean	SD	CP vs. TDC	CP vs. DCD	
Biomechanical constraints	92.32	7.04	<0.001*	<0.001*	80.00	9.92	<0.001*	0.082	73.36	6.61	<0.001*	0.082	
Limits of stability and verticality	76.39*	12.15	<0.001*	0.108	66.40	11.03	<0.001*	1.000	69.55	10.51	0.108	1.000	
Transitions and anticipatory postural adjustments	87.74	10.32	<0.001*	<0.001*	60.40	12.10	<0.001*	0.746	55.91	14.45	<0.001*	0.746	
Reactive postural responses	81.70	13.62	0.001*	<0.001*	67.75	14.43	0.001*	0.375	59.18	19.00	<0.001*	0.375	
Sensory orientation	96.74	4.62	<0.001*	<0.001*	84.55	11.84	<0.001*	1.000	86.00	9.24	<0.001*	1.000	
Stability in gait	79.37	14.28	<0.001*	<0.001*	56.00	12.59	<0.001*	0.020*	41.91	14.87	<0.001*	0.020*	
Total score	85.79	6.65	<*0.001	<0.001*	69.25	4.97	<0.001*	0.102	64.36	10.89	<0.001*	0.102	
Total score (without biomechanical constraints)	84.44	7.32	<0.001*	<0.001*	67.00	5.77	<0.001*	0.261	62.73	7.62	<0.001*	0.261	

Note. * p < 0.05; Kids-BESTest: Kids Balance Evaluation Systems Test; TDC :Typically Developing Children; DCD: Developmental Coordination Disorder; CP: Cerebral Palsy; SD: Standard Deviation

Figure 1

Boxplot of Kids-BEST test performances (%)



Note. Kids-BESTest: Kids Balance Evaluation Systems Test; TDC: typically developing children; DCD: developmental coordination disorder; CP: cerebral palsy

5. Discussion

5.1. Reflection on our research question(s)

The purpose of this study was to compare and define clear differences between children with DCD versus TDC and CP children on the entire construct of balance that can be measured by the kids-BESTest. All six domains together, linked to their underlying mechanisms, form total mean scores which show that children with DCD score significantly poorer than TDC. DCD children score better but not significantly better than CP. In particular, domain I (*biomechanical constraints*), domain V (*reactive postural control*), and domain VI (*stability in gait*) can be causes of the overall poorer performance of CP children versus DCD. On the other domains, they score in a similar range.

Within the DCD group, we see that these children perform particularly poorly on domain III (*transitions and anticipatory postural adjustments*) and domain VI (*stability in gait*). Domain II (*limits of stability and verticality*) and IV (*reactive postural responses*) also score below average but better than the previous domains. Only domain I (*biomechanical constraints*) and V (*sensory orientation*) score normal (>80%).

5.2. Reflection on balance performances

It is currently argued that DCD in terms of balance skills belongs somewhere on the continuum between TDC and CP. Some studies claim that children with DCD exhibit similar balance strategies compared to TDC, while other studies argue the opposite (Verbecque et al., 2021). A child's total score gives an overall picture of their postural control. The Kids-BESTest is based on the typical development of balance by age for TDC and we also see in the results that these children perform normally (>80%) on this test (M = 85.79; SD = 6.65). Since we know that DCD and CP children have difficulty with postural control, it was also hypothesized that these two groups would score below the cut-off value (Blank et al., 2019; Carlberg & Hadders-Algra, 2005; Geuze, 2005; S. L. Pavão et al., 2013). Despite knowing that these 2 groups have balance deficits, they have never been compared with each other in terms of postural control. As expected, DCD children have better results (M = 69.25; SD = 4.97) (but not significantly better) than CP children (M = 64.36; SD = 10.89). The DCD group has a low standard deviation, in the CP group it is higher, and in TDC in between. Minimum and maximum scores are also higher in DCD compared with CP. As mentioned earlier, none of the groups have yet been compared

for the entire balance construct and thus we cannot compare these total scores with results from other studies.

We included the total score without the domain of (I) biomechanical constraints in the analysis to see if this shows a big difference from the total score with all six domains and thus could give a better indication of pure balance performance. This domain does not test true balance skills but is rather an objective observation of feet deformities, BOS, postural alignment, ankle/hip strength, and ROM (Horak et al., 2009). Especially for children with mild conditions, the scores of domain I (biomechanical limitations) influence the total scores because children who do not have biomechanical limitations receive a maximum score. This can result in an overestimation of balance performance. On the other hand, because children with CP often experience problems at the functional level (e.g., spastic plantar flexors, malalignment,...), including domain I (biomechanics constraints) in the total score may put them at a disadvantage, causing an underestimation (Abd El-Nabie & Saleh, 2019; Krarup et al., 2021). These hypotheses can be rejected based on our results. In contrast to what we thought, children with DCD show a small positive difference in the mean score when comparing the total score (M = 69.25; SD = 4.97) with the total score without domain I (M = 67.00; SD = 5.77). In the CP group there is a small difference with a lower mean score when comparing total score with total score (M = 64.36; SD = 10.89) without domain I (M = 62.73; SD = 7.62). Probably because only CP children with GMFCS levels I and II were included and they may show fewer biomechanical problems than children with higher GMFCS levels. Yet, the difference between total scores is very small in all groups and gives us little information whether it effectively gives an over- or underestimation as mentioned above.

As expected, children with CP scored lowest (M = 73.36; SD = 6.61) on (I) biomechanical constraints because postural malalignment, pain, and contractures are frequent problems in the population of CP (Abd El-Nabie & Saleh, 2019; Krarup et al., 2021; Ostojic et al., 2020; Vitrikas et al., 2020). The fact that DCD children still score much lower (M = 80.00; SD = 9.92) than TDC children (M = 92.32; SD = 7.04) and thus also differ significantly from each other in this domain may be caused by the fact that DCD children show reduced strength gains in their development versus TDC. However, anthropometric and body composition factors were not included in this study (Demers et al., 2020). Additionally, a study that did consider

anthropometric parameters, shows that DCD children can generate lower maximum force, and are less powerful in general compared to typically developed children (Raynor, 2001).

DCD children present weakest (M = 66.40; SD = 11.03) in domain II (*limits of stability and* verticality) in comparison to TDC (M = 76.39; SD = 12.15) and CP children (M = 69.55; SD =10.51). According to the systematic review by Verbecque et al., 2021, DCD children score the same as TDC on verticality (Verbecque et al., 2021). Thus, one hypothesis might be that DCD children have particular difficulty with limits of stability (LOS). The study by Fong et al., 2016 shows that children with DCD have directional disturbances but mainly in the backward direction (Fong et al., 2016). The backward direction is not included in the Kids-BESTest, only lateral and forward LOS are evaluated allowing children with DCD to give a better result compared to their true performance on this domain. We conclude from other studies that multi-sensory integration problems along with self-movement and atypical postural control strategies (excessive hip strategy) could be important causes of LOS-related directional balance dysfunction in children with DCD (Bair et al., 2011; Bair et al., 2012; Fong et al., 2011). An overlap in deficits between DCD and CP can be seen in the evidence of less-well-developed internal models of body orientation (Bair et al., 2011; Di Vita et al., 2020) which is needed in leaning in their LOS and returning to vertical. What must also be considered is that although TDC score best among the groups in this domain, they also do not reach the 80% mark which could explain why there is no significant difference between TDC and CP. One hypothesis might be that the Functional Reach Test, forward and lateral, are difficult items even for TDC but some ambiguity remains in this domain.

In domain III (*transitions and anticipatory postural adjustments*), TDC show good results (M = 87.74; SD = 10.32). DCD children score low in this domain (M = 60.40; SD = 12.10) compared to other domains within the DCD group. CP then scores lowest (M = 55.91; SD = 14.45). Tasks addressing anticipatory control such as "alternate stair touching" and "standing arm raise" are items in this domain that contain forward movements of the limbs, with which DCD children exhibit deficits (Johnston et al., 2002; Kane & Barden, 2014; Yam & Fong, 2019). Also, during fast arm movements forward, slower contractions of the abdominal muscles and faster contractions of the erector spinae were found. These may contribute to the disadvantage of children with DCD in this domain. Multiple studies in CP also found that these children have problems with postural muscle pre-tensioning (Bigongiari et al., 2011; Girolami et al., 2011;

Liu et al., 2007; S. L. Pavão et al., 2013). Another item performed is the "one leg stance test" (OLS). Previous research highlighted that the OLS (in the MABC-2 test) induced large differences between TDC and DCD when expressed as a standard score in favor of TDC (Cherng et al., 2007). In both the DCD and CP groups, we hypothesize a deficit in motor planning, although studies show that there is still a lack of supporting neurophysiological data (Williams et al., 2014; Wilson et al., 2013). Motor planning is accompanied by efficient feedback loops and feedforward mechanisms to anticipate prior to a disruption or change in posture that will occur. These are important aspects in the items of this domain (Horak et al., 2009; Shumway-Cook & Woollacott, 2014). Therefore, it was hypothesized that these two groups would score below average in this domain.

CP children score lowest in domain IV (reactive postural control) (M = 59.18; SD = 19.00) followed by the DCD group (M = 67.75; SD = 14.43). Even though no statistically significant difference was present, children with DCD seemed to perform better than the CP children (Table 1). Standard deviations are high in this domain for all groups compared with other domains. An important mechanism for reactive control is to automatically elicit appropriate muscle responses to maintain position and prevent a fall. There are still conflicting results on the activation of muscular response to maintain the upright position. This is either elicited by a moving platform (Neurocom) or a push. Both methods elicit the same muscular response to prevent a fall but with a push, significantly later in DCD children compared to TDC but not with a moving platform (Cheng et al., 2018; Fong et al., 2015). Cheng et al., 2018 found some delay in the activation of lower limb postural muscles (backward platform displacement: medial hamstrings and gastrocnemius; forward platform displacement: rectus femoris and tibialis anterior) in children with DCD compared to TDC, although these findings are not consistent. This study also states that children with DCD had a slower response to the backward perturbations and a faster response to forward perturbations (Cheng et al., 2018). To elaborate further on this, more in-depth research is needed. Cheng et al., 2018 also suggests that inattention (measured with an EEG) in children with DCD (with and without ASD) may affect how they respond to the external disturbances (Cheng et al., 2018). Studies (with moving platform) in CP classified as GMFCS levels I, II and III with spastic hemiplegia/diplegia have found that they need to use a step strategy more often at lower platform movement speeds than TDC. Recovery from instability is also significantly longer

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(Chen & Woollacott, 2007). A similar study such as Chen & Woollacott, 2007 would be recommended for children with DCD to compare strategy use at large and small perturbations with TDC or children with CP. Our findings suggest that the postural reactive system is inadequate in both DCD and CP and may explain why falls are more common in these children.

For domain V (sensory orientation) excellent results were found for TDC (M range between groups = 84.55 - 96.74) and only for this domain, did all groups score normal (>80%). Yet DCD children score lowest, but mean scores indicate only a very small difference between CP (M = 86.00; SD = 9.24) and DCD (M = 84.55; SD = 11.84). This contrasts with the fact that both children with DCD and CP experience difficulties with sensorimotor aspects (such as proprioceptive functions, visuomotor perception, and sensory integration) (Wilson et al., 2013). Several studies verify that children with DCD have difficulty maintaining static balance when relying solely on visual or vestibular information which is why they show greater sway and greater use of hip strategy (Fong et al., 2012; Grove & Lazarus, 2007). In other studies, it was found that in a natural posture (standing) with eyes open, children with DCD and CP exhibit a larger swing area and swing path measured by force plates, indicating poorer balance (S. L. Pavão et al., 2013; Przysucha & Taylor, 2004; Tsai & Wu, 2008; Tsai et al., 2008). However, DCD children use a similar movement strategy as TDC in a natural position with eyes open but make greater use of a hip strategy and greater postural sway during a closed-eye position (Fong et al., 2012; Fong et al., 2013). These differences in sway and strategy use in DCD and CP children may lead to more signs of instability during test items, which can explain the significantly lower score versus TDC. Because a ceiling effect exists for this domain, we expect this domain to be fairly easy for these populations. Another explanation may be that the test is not sensitive enough and that measuring sway (for example by using the center of pressure) is a more sensitive measure to detect differences in sensory orientation. The difficulty of the perturbation within the item (e.g., the thickness of the Airex cushion) may also be a factor positively affecting the results. Preferably, visual information is used in children with CP to compensate for musculoskeletal and neuromotor dysfunctions (S. L. Pavão et al., 2013). Similar findings show that DCD children also benefit from visual support to counteract sway, due to less effective use of tactile information. The study of Blair et al. thereby suggests that this may also contribute to deficits in multisensory

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integration which is the cause of less well-developed internal models of body orientation in children with DCD (Bair et al., 2011). From this, we conclude that these groups thus clearly have greater difficulty with closed-eye items.

For both DCD and CP, stability in gait (VI) is the weakest domain. We see an extremely low mean score for CP (M = 41.91; SD = 14.87) followed by also a very low score for DCD (M = 56.00; SD = 12.59). All groups are significantly different from each other which means that there is a large range between the groups for this domain. Standard deviations are also fairly high in all groups. It is well established that CP exhibits much more and more severe abnormal gait patterns (Rethlefsen et al., 2017) than TDC and DCD children which may explain their poor results. If we then look at DCD children in terms of gait pattern or parameters, there are few significant differences in temporospatial, kinetic, and kinematic parameters of gait compared to TDC (Smith et al., 2021). Nevertheless, we see a large difference in stability mean scores (Table 1). Wilmut et al. did find some differences in gait pattern in the DCD population such as increased step width, increased variability double support time and step time, greater mediolateral speed and acceleration. Differences were also found in the control of the center of mass, this may be due to an integration problem of sensory information for the control of walking. This can indicate why these children show a higher incidence of stumbling or falling and thus score poorly on this domain (Wilmut et al., 2016). Walking is a complex task that incorporates many of the previous domains such as anticipatory postural adjustments and reactive postural control. Therefore, through training programs in these domains with their underlying mechanisms, improvement could also be seen in functional tasks such as walking.

5.3. Clinical implications

Knowledge about the balance profile of DCD children on the kids-BESTest could improve adequate physical therapy assessment and treatment planning. Improving balance performance will result in the better acquisition of motor skills, allowing children with this condition to perform better in daily activities. Poor postural control is also associated with more falls and injuries (Shumway-Cook & Woollacott, 2014). If it is known in which domain a child fails and thus which underlying mechanisms may be deficient, a therapist knows which type of tasks is the main focus in a clinical setting. In general, for DCD especially domain VI (*stability in gait*) and domain III (*transitions and anticipatory postural adjustments*) score weakly, but with intermediate scores between TDC and CP. That would imply that the focus in a clinical setting should be on these domains linked to their underlying mechanisms and that these can be further explored in subsequent studies within the DCD group. However, other studies with a larger sample should confirm this. It is also important to keep in mind that each child still has its individual deficits even though this study uses an average score for each group. Because the sensorimotor dysfunctions in children with DCD recur in multiple domains, this should be addressed by emphasizing the appropriate use of ankle and hip strategies and the use of visual and vestibular input in individualized postural control training (Fong et al., 2012; Fong et al., 2013).

Only a significant difference was found between DCD and CP for stability in gait (domain VI). Thus, training programs can be compared for the other domains between DCD and CP and how they may overlap. For domains II (*limits of stability and verticality*) and V (*sensory orientation*) the mean difference is small, so especially for these domains similar training programs can be compared and possibly set up. Since there is much more research on CP about balance deficits and their training programs, this could be very interesting to investigate in future studies.

5.4. Strengths and limitations of the study

A strength of the study is that with the recent extension of the age-specific version of the Kids-BESTest (Appendix 1), the factor of age is automatically taken into account during the assessment and considers the normal development of balance in a normally developing child. For example, five-year-old children do not have to perform certain items of domain III (*transitions and anticipatory postural adjustments*) because these items should not yet have been acquired and are therefore not age-appropriate. These "age-specific adjustments" were taken into account in the calculation of the domain and total scores by excluding them from the calculation. It is also assumed that if a child in general has problems with an underlying mechanism of balance, this is reflected in all the item scores of the domain and thus of the domain itself.

Only children with GMFCS levels I and II were included in the CP group because it is required, for a presentative outcome of the Kids-BESTest, to be able to stand and walk without assistance from others. In the general questionnaire completed prior to the study, parents of children with CP were asked if assistive devices such as AFO's or other orthotics were worn. However, they were not allowed to use these during the testing session because they help the child with support and, consequently, could enhance their balance. Therefore, this could affect the measurement.

Some demographic data were missing within our dataset, especially within the CP group for GMFCS level and within the TDC group for gestational age. In the CP group, there was one participant who could not perform domain V (*reactive postural control*). Therefore, the domain and total score of this participant were not included in the analysis and assumed as missing data.

Due to the difference in participants in the groups, with a much larger sample in the TDC group than in the DCD and CP group sample, this may produce biased results due to selection bias. In follow-up studies, it is recommended to use equal participants among groups and possibly matched pairing. Due to the voluntary enrollment of participants, self-selection bias may be present, making the study less representative of the population making generalizability difficult. Because the researcher who administered the tests knew which group the child being tested was in, an interpretation bias can occur. Because the researcher may unconsciously have certain expectations within certain groups and thus steer the scoring in the direction expected. This could be countered by blinding the group to the researcher. No studies have yet been done on the psychometric properties of the Kids-BESTest within the DCD population so it is still unclear whether this test is reliable and valid so measurement bias may be present for the DCD group.

For a power of 80%, we should have at least 12 participants in each group resulting that our sample size being just large enough (TDC: N = 54; DCD: N = 20; CP: N = 12). Nevertheless, to ensure generalizability, we could do a future similar study with a larger group for DCD and CP.

5.5. Recommendations for further research

Where exactly the position of TDC, DCD, and CP is on the continuum of postural control has been partly clarified with this study. However, more research is still needed to create delineated balance profiles for children with DCD, so these profiles can be compared with TDC and CP. These profiles could be made even clearer by doing a longitudinal follow-up to better understand the underlying mechanisms of DCD. Adding measurements for brain activity and neuromechanics to a functional test such as the Kids-BESTest test could also add value to these profiles. This thesis focused mainly on total and domain scores. Analyses focusing on the item level of the Kids-BESTest may provide more in-depth information. This way, it would be possible to find out in even more detail which tasks experience difficulties. This could indicate why certain domains score worse in certain groups.

It would add value to the Kids-BESTest to know for the percentages of total scores whether the test is typical, below average, or deviant by incorporating even more cutoff values (for example >80% = typical; 70-80% = below average; <70% = deviant). Thus, clearer statements can be made about the children's performance. No studies have yet been published on the psychometric properties of the Kids-BESTest for the target DCD group. This would be recommended, especially since this test could be a good tool for detecting specific balance problems and including them in therapy for these children.

Because DCD is such a heterogeneous group, it could be useful to make certain subgroups in this diagnosis to better address and clarify this heterogeneity. Then also these subgroups could be compared within the Kids-BESTest. However, this will require much more research on DCD and its characteristics under which we would best make these subgroups.

6. Conclusion

We conclude that DCD children have a balance performance below the cut-off value, measured with the Kids-BESTest. they differ significantly from TD children but not from CP children in their total score. DCD children score significantly worse than TDC in all domains. Only on domain VI (stability in gait), DCD and CP score significantly different, in the disadvantage of CP children. Of all the domains within DCD, children with DCD scored weakest in the domain of (III) *transitions and anticipatory postural adjustments*, and (VI) *stability of gait*. Further research into the existence of a balance continuum between these groups is necessary. Similar studies with a larger sample size and equal groups are recommended to ensure generalizability of these results. Also, a longitudinal follow-up through a cohort study would add value to further understand the underlying mechanisms of DCD.

7. References

- Abd El-Nabie, W. A. E., & Saleh, M. S. M. (2019). Trunk and pelvic alignment in relation to postural control in children with cerebral palsy. *J Back Musculoskelet Rehabil*, *32*(1), 125-130. <u>https://doi.org/10.3233/bmr-181212</u>
- American Psychiatric Association, D., & Association, A. P. (2013). *Diagnostic and statistical manual of mental disorders: DSM-5* (Vol. 5). American psychiatric association Washington, DC.
- Bair, W. N., Barela, J. A., Whitall, J., Jeka, J. J., & Clark, J. E. (2011). Children with developmental coordination disorder benefit from using vision in combination with touch information for quiet standing. *Gait Posture*, 34(2), 183-190. <u>https://doi.org/10.1016/j.gaitpost.2011.04.007</u>
- Bair, W. N., Kiemel, T., Jeka, J. J., & Clark, J. E. (2012). Development of multisensory reweighting is impaired for quiet stance control in children with developmental coordination disorder (DCD). *PLoS One*, 7(7), e40932. <u>https://doi.org/10.1371/journal.pone.0040932</u>
- Bax, M., Goldstein, M., Rosenbaum, P., Leviton, A., Paneth, N., Dan, B., Jacobsson, B., & Damiano, D. (2005). Proposed definition and classification of cerebral palsy, April 2005. *Developmental Medicine & Child Neurology*, 47(8), 571-576. <u>https://doi.org/10.1017/S001216220500112X</u>
- Bigongiari, A., de Andrade e Souza, F., Franciulli, P. M., Neto, S. E. R., Araujo, R. C., & Mochizuki, L. (2011). Anticipatory and compensatory postural adjustments in sitting in children with cerebral palsy. *Human Movement Science*, *30*(3), 648-657.
 https://doi.org/https://doi.org/10.1016/j.humov.2010.11.006
- Blank, R., Barnett, A. L., Cairney, J., Green, D., Kirby, A., Polatajko, H., Rosenblum, S., Smits-Engelsman, B., Sugden, D., Wilson, P., & Vincon, S. (2019). International clinical practice recommendations on the definition, diagnosis, assessment, intervention, and psychosocial aspects of developmental coordination disorder. *Dev Med Child Neurol*, 61(3), 242-285. <u>https://doi.org/10.1111/dmcn.14132</u>
- Brix, N., Ernst, A., Lauridsen, L. L. B., Parner, E., Støvring, H., Olsen, J., Henriksen, T. B., & Ramlau-Hansen, C. H. (2019). Timing of puberty in boys and girls: A population-based study. *Paediatr Perinat Epidemiol*, 33(1), 70-78. <u>https://doi.org/10.1111/ppe.12507</u>
- Brown, T. (2013). Movement Assessment Battery for Children: 2nd Edition (MABC-2). In F. R. Volkmar (Ed.), *Encyclopedia of Autism Spectrum Disorders* (pp. 1925-1939). Springer New York. <u>https://doi.org/10.1007/978-1-4419-1698-3_1922</u>
- Carlberg, E. B., & Hadders-Algra, M. (2005). Postural dysfunction in children with cerebral palsy: some implications for therapeutic guidance. *Neural Plast*, *12*(2-3), 221-228; discussion 263-272. <u>https://doi.org/10.1155/np.2005.221</u>
- Chen, J., & Woollacott, M. H. (2007). Lower extremity kinetics for balance control in children with cerebral palsy. *J Mot Behav*, *39*(4), 306-316. <u>https://doi.org/10.3200/jmbr.39.4.306-316</u>
- Cheng, Y. T. Y., Tsang, W. W. N., Schooling, C. M., & Fong, S. S. M. (2018). Reactive balance performance and neuromuscular and cognitive responses to unpredictable balance perturbations in children with developmental coordination disorder. *Gait Posture*, *62*, 20-26. <u>https://doi.org/10.1016/j.gaitpost.2018.02.025</u>
- Cherng, R.-J., Hsu, Y.-W., Chen, Y.-J., & Chen, J.-Y. (2007). Standing balance of children with developmental coordination disorder under altered sensory conditions. *Human Movement Science*, *26*(6), 913-926. <u>https://doi.org/https://doi.org/10.1016/j.humov.2007.05.006</u>
- Demers, I., Moffet, H., Hébert, L., & Maltais, D. B. (2020). Growth and muscle strength development in children with developmental coordination disorder. *Developmental Medicine & Child Neurology*, 62(9), 1082-1088. <u>https://doi.org/https://doi.org/10.1111/dmcn.14507</u>
- Dewar, R., Claus, A. P., Tucker, K., Ware, R., & Johnston, L. M. (2017). Reproducibility of the Balance Evaluation Systems Test (BESTest) and the Mini-BESTest in school-aged children. *Gait Posture*, 55, 68-74. <u>https://doi.org/10.1016/j.gaitpost.2017.04.010</u>

- Dewar, R., Claus, A. P., Tucker, K., Ware, R. S., & Johnston, L. M. (2019). Reproducibility of the Kids-BESTest and the Kids-Mini-BESTest for Children With Cerebral Palsy. Arch Phys Med Rehabil, 100(4), 695-702. <u>https://doi.org/10.1016/j.apmr.2018.12.021</u>
- Di Vita, A., Cinelli, M. C., Raimo, S., Boccia, M., Buratin, S., Gentili, P., Inzitari, M. T., Iona, T., Iosa, M., Morelli, D., Ruggeri, F., Russo, G., Guariglia, C., & Palermo, L. (2020). Body Representations in Children with Cerebral Palsy. *Brain Sci*, *10*(8). <u>https://doi.org/10.3390/brainsci10080490</u>
- Faul, F., Erdfelder, E., Lang, A.-G., & Buchner, A. (2007). G*Power 3: A flexible statistical power analysis program for the social, behavioral, and biomedical sciences. *Behavior Research Methods*, 39(2), 175-191. <u>https://doi.org/10.3758/BF03193146</u>
- Fong, S. S., Lee, V. Y., & Pang, M. Y. (2011). Sensory organization of balance control in children with developmental coordination disorder. *Res Dev Disabil*, 32(6), 2376-2382. <u>https://doi.org/10.1016/j.ridd.2011.07.025</u>
- Fong, S. S., Ng, S. S., Chung, L. M., Ki, W. Y., Chow, L. P., & Macfarlane, D. J. (2016). Direction-specific impairment of stability limits and falls in children with developmental coordination disorder: Implications for rehabilitation. *Gait Posture*, 43, 60-64. <u>https://doi.org/10.1016/j.gaitpost.2015.10.026</u>
- Fong, S. S., Tsang, W. W., & Ng, G. Y. (2012). Altered postural control strategies and sensory organization in children with developmental coordination disorder. *Hum Mov Sci*, 31(5), 1317-1327. <u>https://doi.org/10.1016/j.humov.2011.11.003</u>
- Fong, S. S. M., Ng, S. S. M., Guo, X., Wang, Y., Chung, R. C. K., Stat, G., Ki, W. Y., & Macfarlane, D. J. (2015). Deficits in Lower Limb Muscle Reflex Contraction Latency and Peak Force Are Associated With Impairments in Postural Control and Gross Motor Skills of Children With Developmental Coordination Disorder: A Cross-Sectional Study. *Medicine (Baltimore)*, 94(41), e1785. <u>https://doi.org/10.1097/md.00000000001785</u>
- Fong, S. S. M., Ng, S. S. M., & Yiu, B. P. H. L. (2013). Slowed muscle force production and sensory organization deficits contribute to altered postural control strategies in children with developmental coordination disorder. *Research in Developmental Disabilities*, 34(9), 3040-3048. <u>https://doi.org/https://doi.org/10.1016/j.ridd.2013.05.035</u>
- Geuze, R. H. (2005). Postural control in children with developmental coordination disorder. *Neural Plast*, *12*(2-3), 183-196; discussion 263-172. <u>https://doi.org/10.1155/NP.2005.183</u>
- Girolami, G. L., Shiratori, T., & Aruin, A. S. (2011). Anticipatory postural adjustments in children with hemiplegia and diplegia. *J Electromyogr Kinesiol*, 21(6), 988-997. <u>https://doi.org/10.1016/j.jelekin.2011.08.013</u>
- Green, D., Baird, G., & Sugden, D. (2006). A pilot study of psychopathology in Developmental Coordination Disorder. *Child Care Health Dev*, *32*(6), 741-750. <u>https://doi.org/10.1111/j.1365-2214.2006.00684.x</u>
- Grove, C. R., & Lazarus, J. A. (2007). Impaired re-weighting of sensory feedback for maintenance of postural control in children with developmental coordination disorder. *Hum Mov Sci*, 26(3), 457-476. <u>https://doi.org/10.1016/j.humov.2007.01.014</u>
- Horak, F. B., Wrisley, D. M., & Frank, J. (2009). The Balance Evaluation Systems Test (BESTest) to differentiate balance deficits. *Phys Ther*, *89*(5), 484-498. <u>https://doi.org/10.2522/ptj.20080071</u>
- Johnson, C., Hallemans, A., Goetschalckx, M., Meyns, P., Rameckers, E., Klingels, K., & Verbecque, E. (2023). Psychometric properties of functional postural control tests in children: A systematic review. Ann Phys Rehabil Med, 66(4), 101729. <u>https://doi.org/10.1016/j.rehab.2022.101729</u>
- Johnston, L. M., Burns, Y. R., Brauer, S. G., & Richardson, C. A. (2002). Differences in postural control and movement performance during goal directed reaching in children with developmental coordination disorder. *Hum Mov Sci*, 21(5-6), 583-601. <u>https://doi.org/10.1016/s0167-9457(02)00153-7</u>
- Kadesjö, B., & Gillberg, C. (1998). Attention deficits and clumsiness in Swedish 7-year-old children. Dev Med Child Neurol, 40(12), 796-804. <u>https://doi.org/10.1111/j.1469-8749.1998.tb12356.x</u>

- Kane, K., & Barden, J. (2014). Frequency of anticipatory trunk muscle onsets in children with and without developmental coordination disorder. *Phys Occup Ther Pediatr*, 34(1), 75-89. <u>https://doi.org/10.3109/01942638.2012.757574</u>
- Krarup, L. H., Kristensen, P. K., Strand, L., Bredtoft, S. L., Mechlenburg, I., & Nordbye-Nielsen, K. (2021). Ankle contractures are frequent among children with cerebral palsy and associated with lower gross motor function and degree of spasticity. *Acta Paediatr*, 110(7), 2171-2178. <u>https://doi.org/10.1111/apa.15804</u>
- Lingam, R., Golding, J., Jongmans, M. J., Hunt, L. P., Ellis, M., & Emond, A. (2010). The Association Between Developmental Coordination Disorder and Other Developmental Traits. *Pediatrics*, 126(5), e1109-e1118. <u>https://doi.org/10.1542/peds.2009-2789</u>
- Liu, W. Y., Zaino, C. A., & McCoy, S. W. (2007). Anticipatory postural adjustments in children with cerebral palsy and children with typical development. *Pediatr Phys Ther*, *19*(3), 188-195. https://doi.org/10.1097/PEP.0b013e31812574a9
- Ostojic, K., Paget, S., Kyriagis, M., & Morrow, A. (2020). Acute and Chronic Pain in Children and Adolescents With Cerebral Palsy: Prevalence, Interference, and Management. *Arch Phys Med Rehabil*, 101(2), 213-219. <u>https://doi.org/10.1016/j.apmr.2019.08.475</u>
- Pavão, S. L., dos Santos, A. N., Woollacott, M. H., & Rocha, N. A. (2013). Assessment of postural control in children with cerebral palsy: a review. *Res Dev Disabil*, 34(5), 1367-1375. <u>https://doi.org/10.1016/j.ridd.2013.01.034</u>
- Pavão, S. L., dos Santos, A. N., Woollacott, M. H., & Rocha, N. A. C. F. (2013). Assessment of postural control in children with cerebral palsy: A review. *Research in Developmental Disabilities*, 34(5), 1367-1375. <u>https://doi.org/https://doi.org/10.1016/j.ridd.2013.01.034</u>
- Przysucha, E. P., & Taylor, M. J. (2004). Control of Stance and Developmental Coordination Disorder: The Role of Visual Information. *Adapted Physical Activity Quarterly*, *21*(1), 19-33. <u>https://doi.org/10.1123/apaq.21.1.19</u>
- Raynor, A. J. (2001). Strength, power, and coactivation in children with developmental coordination disorder. *Developmental Medicine and Child Neurology*, *43*(10), 676-684. <u>https://doi.org/10.1017/S0012162201001220</u>
- Rethlefsen, S. A., Blumstein, G., Kay, R. M., Dorey, F., & Wren, T. A. (2017). Prevalence of specific gait abnormalities in children with cerebral palsy revisited: influence of age, prior surgery, and Gross Motor Function Classification System level. *Dev Med Child Neurol*, 59(1), 79-88. <u>https://doi.org/10.1111/dmcn.13205</u>
- Shumway-Cook, A., & Woollacott, M. (2014). *Motor control: Translating research into clinical practice: Fourth edition*.
- Smith, M., Ward, E., Williams, C. M., & Banwell, H. A. (2021). Differences in walking and running gait in children with and without developmental coordination disorder: A systematic review and meta-analysis. *Gait Posture*, 83, 177-184. <u>https://doi.org/10.1016/j.gaitpost.2020.10.013</u>
- Tsai, C. L., & Wu, S. K. (2008). Relationship of visual perceptual deficit and motor impairment in children with developmental coordination disorder. *Percept Mot Skills*, 107(2), 457-472. https://doi.org/10.2466/pms.107.2.457-472
- Tsai, C. L., Wu, S. K., & Huang, C. H. (2008). Static balance in children with developmental coordination disorder. *Hum Mov Sci*, 27(1), 142-153. <u>https://doi.org/10.1016/j.humov.2007.08.002</u>
- Van der Linde, B. W., van Netten, J. J., Otten, B., Postema, K., Geuze, R. H., & Schoemaker, M. M. (2015). Activities of Daily Living in Children With Developmental Coordination Disorder: Performance, Learning, and Participation. *Physical Therapy*, *95*(11), 1496-1506. https://doi.org/10.2522/ptj.20140211
- Verbecque, E., Johnson, C., Rameckers, E., Thijs, A., van der Veer, I., Meyns, P., Smits-Engelsman, B., & Klingels, K. (2021). Balance control in individuals with developmental coordination disorder: A systematic review and meta-analysis. *Gait Posture*, *83*, 268-279. <u>https://doi.org/10.1016/j.gaitpost.2020.10.009</u>

- Vitrikas, K., Dalton, H., & Breish, D. (2020). Cerebral Palsy: An Overview. Am Fam Physician, 101(4), 213-220.
- Williams, J., Hyde, C., & Spittle, A. (2014). Developmental Coordination Disorder and Cerebral Palsy: Is There a Continuum? *Current Developmental Disorders Reports*, 1(2), 118-124. <u>https://doi.org/10.1007/s40474-014-0009-3</u>
- Wilmut, K., Du, W., & Barnett, A. L. (2016). Gait patterns in children with Developmental Coordination Disorder. *Experimental Brain Research*, 234(6), 1747-1755. <u>https://doi.org/10.1007/s00221-016-4592-x</u>
- Wilson, B. N., Crawford, S. G., Green, D., Roberts, G., Aylott, A., & Kaplan, B. J. (2009). Psychometric properties of the revised Developmental Coordination Disorder Questionnaire. *Phys Occup Ther Pediatr*, 29(2), 182-202. <u>https://doi.org/10.1080/01942630902784761</u>
- Wilson, P. H., RUDDOCK, S., SMITS-ENGELSMAN, B., POLATAJKO, H., & BLANK, R. (2013). Understanding performance deficits in developmental coordination disorder: a meta-analysis of recent research. *Developmental Medicine & Child Neurology*, 55(3), 217-228. <u>https://doi.org/https://doi.org/10.1111/j.1469-8749.2012.04436.x</u>
- Wilson, P. H., Smits-Engelsman, B., Caeyenberghs, K., Steenbergen, B., Sugden, D., Clark, J., Mumford, N., & Blank, R. (2017). Cognitive and neuroimaging findings in developmental coordination disorder: new insights from a systematic review of recent research. *Developmental Medicine & Child Neurology*, 59(11), 1117-1129. <u>https://doi.org/https://doi.org/10.1111/dmcn.13530</u>
- Wisdom, S. N., Dyck, M. J., Piek, J. P., Hay, D., & Hallmayer, J. (2007). Can autism, language and coordination disorders be differentiated based on ability profiles? *Eur Child Adolesc Psychiatry*, 16(3), 178-186. <u>https://doi.org/10.1007/s00787-006-0586-8</u>
- Yam, T. T. T., & Fong, S. S. M. (2019). Y-Balance Test Performance and Leg Muscle Activations of Children with Developmental Coordination Disorder. J Mot Behav, 51(4), 385-393. <u>https://doi.org/10.1080/00222895.2018.1485011</u>

8. Appendices

Appendix 1: Informed Consent



Onderzoek evenwicht - Toestemming en vragenlijsten

Beste ouder, voogd, grootouder, tante, oom

Gelieve volgende vragen te beantwoorden over uw kind: in totaal neemt dit 15 minuten in beslag en zijn

de vragen gesplitst in 4 delen over 4 pagina's.

De pagina's bevatten:

- In deel 1: vragen we om uw toestemming dat uw kind deelneemt aan dit onderzoek rond het evenwicht.
- In deel 2, 3 en 4 stellen we enkele vragen over uw kind.

Meer informatie over het ondezoek? klik hier

Alle gegevens worden vertrouwelijk behandeld.

Hartelijk dank voor uw interesse uit naam van het hele onderzoeksteam,

Charlotte Johnson,

doctoraatsstudente

DEEL 1: INFORMATIE- & TOESTEMMINGSFORMULIER

Hebt u gemist waar u het volledige formulier kan lezen? Het volledige document vindt u hier: klik hier

Hierbij bevestig ik, ondergetekende [vul hieronder je naam en voornaam in], dat ik over het

onderzoek ben ingelicht en een kopie van het informatiedocument ontvangen heb.

Ik heb de informatie gelezen en begrepen. De onderzoeker heeft mij voldoende informatie gegeven met betrekking tot het doel en het opzet van het onderzoek, de voorwaarden en de duur ervan, en de mogelijke bekende voor- of nadelen die dit onderzoek voor mij kunnen inhouden. Bovendien werd mij voldoende tijd gegeven om de informatie te overwegen en om vragen te stellen, waarop ik bevredigende antwoorden gekregen heb.

Naam + voornaam:

1. Ik ben ervan op de hoogte dat ik de toestemming tot deelname van mijn kind aan het onderzoek en tot verwerking van de gegevens van mijn kind kan weigeren en dat ik de toestemming om de data van mijn kind te gebruiken op elk moment kan intrekken, nadat ik de onderzoeker hierover heb ingelicht, zonder dat dit mij/mijn kind enig nadeel kan berokkenen

🔿 Ja

2. Mijn oorspronkelijke toestemming tot deelname aan het onderzoek en gebruik van de persoonsgegevens van mijn kind zal het gebruik van de gegevens van mijn kind toelaten met betrekking tot de periode dat ik in het onderzoek ingesloten was.

🔿 Ja

3. Ik ben mij bewust van het doel waarvoor de gegevens van mijn kind verzameld, verwerkt en gebruikt worden in het kader van dit onderzoek. Ik ben ervan op de hoogte gesteld dat de gegevens van mijn kind gedurende 20 jaar zullen bewaard worden voor wetenschappelijk onderzoek.

🔿 Ja

 Ik ben ervan op de hoogte gesteld dat de gegevens van mijn kind gedurende 20 jaar zullen bewaard worden voor wetenschappelijk onderzoek.



5. Ik weet dat ik het recht heb om de gegevens van mijn kind in te zien, te verbeteren of te wissen. Als ik klachten heb over het beheer van deze gegevens weet ik dat ik me kan richten tot de onderzoekers of de functionaris voor de gegevensbescherming van de Universiteit Antwerpen (privacy@uantwerpen.be).

6. Ik geef toestemming aan de onderzoeker om de gegevens van mijn kind te verzamelen, te verwerken en te gebruiken zoals beschreven in het informatiedocument dat ik ontving en conform de geldende wetgeving.
Deze gegevens zullen strikt vertrouwelijk behandeld worden. Wanneer de studieresultaten gepubliceerd worden, zal de identiteit van mijn kind geheim blijven.

🔿 Ja

7. Ik ga ermee akkoord dat er standaard videobeelden van mijn kind worden gemaakt tijdens het uitvoeren van de Kids-BESTest en dat deze videobeelden bewaard en gebruikt worden uitsluitend voor dit onderzoek.

🔿 Ja

8. Ik ga eveneens akkoord met het gebruik door de opdrachtgever van het onderzoek van de gecodeerde medische gegevens van mijn kind voor andere onderzoeksdoeleinden. Elk nieuw onderzoek vereist een beoordeling en goedkeuring door het Ethisch Comité.

9. Ik stem geheel vrijwillig toe om mijn kind deel te laten nemen aan dit onderzoek en om mee te werken aan alle gevraagde onderzoekshandelingen (bv. invullen van vragenlijsten, deelname aan een interview, ...).

🔿 Ja

10. Ik ben bereid informatie te verstrekken i.v.m. de medische geschiedenis van mijn kind, zijn/haar geneesmiddelengebruik en eventuele deelname aan andere studies.

🔿 Ja

11. Ik ga ermee akkoord dat de therapeuten, betrokken bij de zorg van mijn kind, op de hoogte worden gebracht van de resultaten van dit onderzoek.

🔿 Ja

O Neen

O NVT

12. Ik ga er eveneens mee akkoord dat u als onderzoeker de nodige informatie krijgt over mijn kind van de therapeuten, betrokken bij de zorg van mijn kind, die nodig zijn voor uw onderzoek (bv. de resultaten van de M-ABC-2, scores op vragenlijsten...)

🔿 Ja		
0		
O Neen		
O NVT		

13. Ik wens op de hoogte gebracht te worden over de resultaten van mijn kind in het kader van dit

onderzoek.

🔿 Ja		
O Neen		

Voor akkoord,

Handtekening,



Appendix 2: The age-adjusted scoring criteria of the Kids-BESTest (example)

DOMAIN IV: REACTIVE POSTURAL RESPONSES – <u>AGE 8-10</u>					
OBSERVATION	SCORING CRITERIA				
	0	1	2	3	Score

Item 1. IN PLACE RESPONSE - FORWARD

Look for the applied movement strategies (ankle and or hip) and whether the child needs Check the boxes that correspond with your observation during test administration. to use the arms or take a step to recover balance. One practice trial may be allowed if **Lowest score counts for overall judgement**. needed. Check the boxes that correspond with your observation.

Movement strategy and/or signs of instability

ankle strategy (feet in place)

trunk aligned with legs with minimal/no arm movement

hip strategy (feet in place)

- Correct: counteracts with mild hip extension
- Incorrect: uses hip flexion instead of extension (flexes forward)
- Excessive: large amplitude trunk, hip or arm movement

stepping strategy

- □ small step (≤ foot length)
- □ large step (> foot length)
- □ multiple steps (≥ 2 steps)

requires assist/ would fall if not caught/will not attempt

Item 2. IN PLACE RESPONSE - BACKWARD

to use the arms or take a step to recover balance. One practice trial may be allowed if Lowest score counts for overall judgement (considering quantitative and qualitative performance). needed.

Check the boxes that correspond with your observation.

Movement strategy and/or signs of instability

ankle strategy (feet in place)

trunk aligned with legs with minimal/no arm movement

hip strategy (feet in place)

Correct: counteracts with mild hip flexion

Incorrect: uses hip extension instead of flexion (extends backward)

- Excessive: large amplitude trunk, hip or arm movement
- stepping strategy
 - □ small step (≤ foot length)
 - □ large step (> foot length)
 - □ multiple steps (≥ 2 steps)

requires assist/ would fall if not caught/will not attempt

□Would fall if	□Uses a <i>stepping</i>	Recovers stability	□Recovers stability	
not caught	strategy with a large	with feet in place	with feet in place	
OR <i>requires</i>	step or multiple steps	using a correct hip	using an ankle strategy	
assist	OR use incorrect hip	strategy		
OR will not	strategy	OR feet in place with		
attempt	OR use excessive	excessive arm motion		
	correction in	OR uses a stepping		
	arms/trunk	strategy with a small		
		step		

Interference: □refuses □distracted /does not follow instructions correctly □inconsistent/ highly variable performance

Look for the applied movement strategies (ankle and or hip) and whether the child needs Check the boxes that correspond with your observation during test administration.



Interference: refuses distracted /does not follow instructions correctly inconsistent/ highly variable performance

Appendix 3: Data analysis decision tree

