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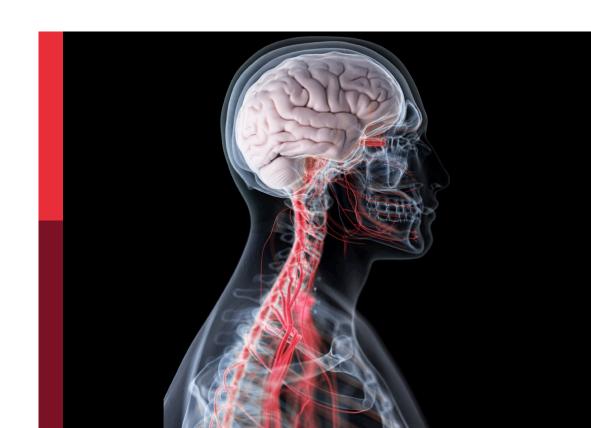
volume II

Edited by

Kate Wilmut, Catherine Purcell and Emily J. Meachon

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Current perspectives on developmental coordination disorder (DCD), volume II

Topic editors

Kate Wilmut — Oxford Brookes University, United Kingdom Catherine Purcell — Cardiff University, United Kingdom Emily J. Meachon — University of Basel, Switzerland

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Table of contents

- 05 Editorial: Current perspectives on Developmental Coordination Disorder (DCD), volume II
 - Kate Wilmut, Catherine Purcell and Emily J. Meachon
- Motor-cognitive coupling is impaired in children with mild or severe forms of developmental coordination disorder

Reza Abdollahipour, Ludvík Valtr, Kamila Banátová, Lucia Bizovská, Tomáš Klein, Zdeněk Svoboda, Bert Steenbergen and Peter Henry Wilson

- 17 Children with developmental coordination disorder are less able to fine-tune muscle activity in anticipation of postural perturbations than typically developing counterparts
 - Carla Harkness-Armstrong, Emma F. Hodson-Tole, Greg Wood and Richard Mills
- 29 Children with developmental coordination disorder have less variable motor unit firing rate characteristics across contractions compared to typically developing children

Maaike Esselaar, Johnny V. V. Parr, Greg Wood and Emma Hodson-Tole

39 Understanding factors that influence physical activity behavior in people with developmental coordination disorder (DCD): a mixed-methods convergent integrated systematic review

Catherine Purcell, Nadja Schott, Victoria Rapos, Jill G. Zwicker and Kate Wilmut

Anthropometric, physical activity, and psychological characteristics of Korean adults with and without developmental coordination disorder (DCD)

Min Joo Kim, Soo Mi Nam, Byeol Kim, Ilhyeok Park, Jaebum Park and Jae Kun Shim

77 Roadside experiences of parents of children with developmental coordination disorder and/or attention deficit hyperactivity disorder

Rayan Falemban, Kate Wilmut, Heather Hurst and Catherine Purcell

- 97 EEG spectral power in developmental coordination disorder and attention-deficit/hyperactivity disorder: a pilot study
 - Emily J. Meachon, Marlene Kundlacz, Kate Wilmut and Georg W. Alpers
- 107 Cortical grey matter volume differences in children with developmental coordination disorder compared to typically developing children

Myrah Malik, Alexander Weber, Donna Lang, Tamara Vanderwal and Jill G. Zwicker



120 Changes in cortical grey matter volume with Cognitive Orientation to daily Occupational Performance intervention in children with developmental coordination disorder

Myrah Anum Malik, Alexander Mark Weber, Donna Lang, Tamara Vanderwal and Jill G. Zwicker

Oculomotor differences in adults with and without probable developmental coordination disorder

Emma Sumner and Elisabeth L. Hill



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Editorial: Current perspectives on Developmental Coordination Disorder (DCD), volume II

Kate Wilmut^{1*}, Catherine Purcell² and Emily J. Meachon³

¹The Centre for Psychological Research, Oxford Brookes University, Oxford, United Kingdom, ²School of Healthcare Sciences, Cardiff University, Cardiff, United Kingdom, ³Faculty of Psychology, University of Basel, Basel, Switzerland

KEYWORDS

Developmental Coordination Disorder, co-occurrence, Attention Deficit and Hyperactivity Disorder (ADHD), social-cultural context, muscular control

Editorial on the Research Topic

Current perspectives on Developmental Coordination Disorder (DCD), volume II

Introduction

Developmental Coordination Disorder is characterized by a difficulty with motor control and coordination which falls substantially below the level expected given an individual's age and opportunity for learning (American Psychiatric Association, 2022). Individuals with DCD also experience associated secondary consequences which include poorer mental health outcomes (Kirby et al., 2013; Draghi et al., 2020), physical inactivity (as described in this Research Topic by Purcell et al.) and challenges with executive function (Purcell et al., 2015; Sartori et al., 2020; Meachon et al., 2022). Despite a prevalence rate of \sim 5% (Blank et al., 2019), which is significantly higher than for autism, DCD remains under-represented within the academic literature and is often misunderstood among professionals (Meachon et al., 2024). This Research Topic aimed to capture current research focusing on DCD and this editorial draws out three themes from the collection of 10 articles

The social-cultural context

One consideration which is so often ignored within research is the environmental context of the participants, the society in which they exist and how the expectations of that society might change or influence behavior. Interestingly two papers within this Research Topic shed light on this environmental or social-cultural context. Abdollahipour et al. considered this within the context of their findings regarding reaching behavior, stating that the different environmental contexts and engagement in outdoor activities of the participants in their study in comparison to previous studies might explain some of the differences in findings. Furthermore, Kim et al. also considered how distinctive cultural factors in the Republic of Korea might influence the presentation of DCD, especially given the high rates of physical activity within that population. The consideration of DCD within different groups and populations is very important, especially in light of multiple studies drawing on the same sample, as identified by Purcell et al. within the physical activity literature in DCD. This, along with the dominance of western research may somewhat bias our understanding of DCD to one specific social-cultural context.

Wilmut et al. 10.3389/fnhum.2024.1466104

DCD and ADHD

Children with DCD are more likely than children without DCD to have attentional difficulties, in fact it is estimated that 50% of children with DCD have co-occurring ADHD (Fliers et al., 2010). This is represented within this Research Topic with four of the articles making specific reference to this. Three of these considered the biological or neural underpinnings of DCD with an aim to make comparisons between children with DCD only and children with DCD and ADHD. Unfortunately, two of these articles which focused on the structure of gray matter (Malik M. et al.) and changes in gray matter following intervention (Malik M. A. et al.) didn't have a sufficient sample size to compare these groups but instead combined them into one to compare to typically developing individuals. Therefore, although these studies do not help us to understand DCD as separate to ADHD, they do provide important findings with regards to brain structure and re-organization after intervention. In contrast, a third study did make this comparison in adults and included a DCD only sample, an ADHD only sample, a sample with DCD and ADHD and a sample with neither. Meachon et al. used EEG to consider resting state neural differences across these groups. The identification of neural structures or mechanisms which are specific to DCD is vital to further our understanding of this as being separate to ADHD.

A final study considered DCD and ADHD in much more of an activity of daily living. Falemban et al. used a qualitative method to delve into the experiences of parents when crossing the road with their child. This consideration of the lived experience is so incredibly important in order for researchers to fully understand DCD and its co-occurrences.

Considerations of muscular control

Finally, three studies included measures or proxies of muscular control. Two of these focused-on children with DCD and found that children with DCD have unique motor strategies in muscular activity during the experience of perturbations while standing (Harkness-Armstrong et al.) and EMG firing rate during a gripping task (Esselaar et al.). These studies demonstrate the importance of considering task complexity and variance of performance rather than just an overall average when describing movement control in children with DCD. Both of these studies represent research which is aiming to explain or describe behavior in children with DCD such as falling or bumping into objects and dropping objects while carrying. In the third study, Sumner and Hill considered oculomotor control in adults with DCD as an extension to a previous study in children. Despite the wealth of research which focuses on children (very often young children) with DCD, it is important to remember that DCD is a life long condition.

Conclusions

Although small, this Research Topic represents the diverse research which is being undertaken in the field of DCD. Looking across these studies we can see the pertinent issues within the field and also the exciting progress in terms of understanding the underlying biology which is driving the behavior with which we are all so familiar, it also acknowledges the importance of considered DCD within the context of co-occurrence and differing levels of task complexity. However, this is also complemented by research which is considering the voice of individuals with lived experience and research which acknowledges the importance of environment or social-cultural contexts of behavior. This represents a new tone in current DCD research compared to the mechanistic work which dominated the field 20 years ago. As this research field grows it is important to identify differences in findings and put effort into explaining why these might occur. It is only by carefully considering all the underlying and contextual factors that might influence an individual that we can fully understand the behavior that we are attempting to describe. Regardless of the research methods or the task being used the shift in focus from describing a disorder to considering the person is such an important one to ensure research is serving the people on which it focuses.

Author contributions

KW: Writing – original draft, Writing – review & editing. CP: Writing – original draft, Writing – review & editing. EM: Writing – original draft, Writing – review & editing.

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Conflict of interest

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EDITED BY
Catherine Purcell,
Cardiff University, United Kingdom

REVIEWED BY
Johnny V. V. Parr,
Manchester Metropolitan University,
United Kingdom
Vicky McQuillan,
University of Salford, United Kingdom

*CORRESPONDENCE Reza Abdollahipour ☑ reza.abdollahipour@upol.cz

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Motor-cognitive coupling is impaired in children with mild or severe forms of developmental coordination disorder

Reza Abdollahipour^{1*}, Ludvík Valtr¹, Kamila Banátová¹, Lucia Bizovská¹, Tomáš Klein¹, Zdeněk Svoboda¹, Bert Steenbergen² and Peter Henry Wilson^{1,3}

¹Department of Natural Sciences in Kinanthropology, Faculty of Physical Culture, Palacký University Olomouc, Olomouc, Czechia, ²Department of Pedagogical and Educational Sciences, Behavioural Science Institute, Radboud University, Nijmegen, Netherlands, ³Healthy Brain and Mind Research Centre, School of Behavioural and Health Sciences, Faculty of Health Sciences, Australian Catholic University, Melbourne, VIC, Australia

Children with developmental coordination disorder (DCD) show deficits in motorcognitive coupling. However, it remains unclear whether such deficits depend on the severity of DCD. The aim of this study was to examine cognitive-motor coupling under different levels of inhibitory control in children with severe (s-DCD) or moderate DCD (m-DCD), compared with typically-developing children (TDC). The performance of 29 primary-school children aged 6-12 years with s-DCD (Mage=9.12+1.56 years), 53 m-DCD (Mage=8.78±1.67years), and 201 TDC (Mage=9.20±1.50years) was compared on a double jump reaching task (DJRT) paradigm, presented on a large 42-inch touchscreen. The task display had a circular home-base, centred at the bottom of the display, and three target locations at radials of -20°, 0°, and 20°, 40cm above the home-base circle. For the standard double-jump reaching task (DJRT), children moved their index finger from home-base circle to touch the target stimulus as fast as possible; 20% were jump trials where the target shifted left or right at lift-off. For the anti-jump reaching task (AJRT), 20% of trials required an anti-jump movement, touching the contralateral target location. While no group differences were shown on the DJRT, the DCD group were slower to complete reaching movements than the TDC group on AJRT; on the latter, the two DCD sub-groups were not shown to differ. Results confirm the presence of motor inhibition deficits in DCD which may not be dependent on the motor severity of the disorder.

KEYWORDS

developmental coordination disorder (DCD), children, motor control, motor inhibition, goal-directed action

1. Introduction

Developmental coordination disorder (DCD) is one of the most common neurodevelopmental disorders, with converging research showing a core deficit in predictive motor control (aka *internal modeling deficit*—IMD; Ruddock et al., 2016; Subara-Zukic et al., 2022), evident across effector systems including oculomotor (Katschmarsky et al., 2001), manual (Hyde and Wilson, 2011, 2013), and dynamic balance (Jelsma et al., 2016, 2020), as well as the integration of cognitive and motor control. This deficit manifests in slower response times,

especially in response to unexpected changes in the environment, and more movement corrections given the child's reliance on slower forms of feedback-based control (Wilson et al., 2013). However, it remains unclear whether this underlying issue in control depends on the severity of DCD: i.e., severe DCD (s-DCD) compared with mild-to-moderate DCD (m-DCD), and on the cognitive load of different movement tasks, issues that bear on our understanding of motor-cognitive coupling in everyday action (Ruddock et al., 2015, 2016).

Poor inhibitory control is commonly observed in DCD, which impacts the performance of tasks that require coupling of predictive motor and cognitive control. Inhibition is defined here as the ability to withhold or re-direct a motor response, often in the face of a prepotent stimulus (Ruddock et al., 2016). Predictive control (viz the ability to use forward estimates of limb position as a means of correcting an action rapidly in real time) is critical to motor coordination and skill development (McNamee and Wolpert, 2019). Ruddock and colleagues have shown that children with DCD are slower to make online corrections to target perturbations and less accurate than typically developing children (TDC) on both the double jump reaching task (DJRT) and anti-jump reaching task (AJRT). The latter task involves the ability to couple online predictive control and cognitive inhibition (Ruddock et al., 2016): the performer is required to monitor sudden jumps in target location (either to the left or right of fixation), but then inhibit a prepotent response and implement a reach movement to a contralateral location. Deficits in eye-limb coupling in DCD are also linked to poor inhibitory control and its integration with motor control (Michel et al., 2018). Sub-group differences have not been tested, however.

The downstream effects on motor performance are potentially quite profound for children with combined cognitive and motor control deficits. In longitudinal research, our group has shown that children with more severe motor coordination difficulties are likely to experience motor and cognitive issues in later development (Wilson et al., 2020). Children with persistent DCD had much poorer executive function than both typically developing children and those with remitting DCD. In short, the combination of persistent DCD and cognitive deficits is relatively common and predicts poorer developmental outcomes in later childhood.

An important theoretical and clinical question that remains unanswered is whether children with s-DCD (≤5th percentile on standardized tests of motor skills) show more profound difficulties in cognitive and motor control than those with m-DCD (between the 5th and 15th percentile). Combined deficits in predictive and inhibitory control may impede performance of visually-guided motor tasks, especially under time constraints and/or cognitive load (Ruddock et al., 2015). Children with s-DCD do perform worse on manual dexterity tasks which, by their nature, require a high degree of visuomotor integration (McQuillan et al., 2021). As well, in the case of motor imagery, an ability linked to internal modeling, Williams et al. (2008) showed that children with s-DCD had particular difficulty on a complex whole-body rotation task, unlike m-DCD who performed like controls. Such findings raise the question as to whether the severity of DCD constrains the ability to generate and utilize (predictive) internal models for action.

The goal of the study reported here was to compare the performance of large groups of children with s-DCD, m-DCD, and TDC on two versions of the visually-guided pointing task (DJRT and AJRT). Children with DCD were classified according to the level of

motor impairment, measured by MABC-2: s-DCD (\leq 5th percentile) and m-DCD (TTS \leq 16th percentile, but >5th). We predicted that the performance of children with DCD would be worse than TDC on key metrics of DJRT and AJRT performance, and, moreover, s-DCD would perform worse than m-DCD on each task.

2. Materials and methods

2.1. Participants

Eighty-two children with DCD (Mage=8.90 ± 1.63 years, age range 6-12 years, 37 girls/45 boys) were recruited, 29 with s-DCD $(Mage = 9.12 \pm 1.56 \text{ years}, \text{ age range } 6-12 \text{ years } 10 \text{ girls/} 19 \text{ boys}) \text{ and}$ 53 with m-DCD ($Mage = 8.78 \pm 1.67$ years, age range 6–12 years, 27 girls/26 boys), together with 201 TDC (Mage = 9.20 ± 1.50 years, age range 6-12 years, 101 girls/100 boys), aged between 6 and 12 years, the latter forming part of a larger longitudinal study. As previous research has shown a large effect on group differences for both DJRT and AJRT (Hyde and Wilson, 2013; Ruddock et al., 2015), an a priori power analysis with G*Power 3.1 indicated that 28 participants in each group would achieve a desired power $(1 - \beta)$ of 0.90, effect size d = 0.08, and an α level of 0.05 (Faul et al., 2007). We used the MABC-2 test battery to assess the level of motor competency in a large pool of children. In total, 201 TDC completed both the DJRT and AJRT. All children were recruited from four primary schools in the Olomouc region and surrounding communities in Moravia of the Czech Republic. All the schools were from urban areas/cities and the number of inhabitants in each city was over 25,000. Initially, the MABC-2 test battery was administered to all children within the age range of 6-12 years. Then, those children who performed under the 16th percentile were also assessed by teachers using the MABC-2 checklist. The study was approved by the Ethical Committee of the Faculty of Physical Culture, Palacký University Olomouc (FTK 46/2020), and participating schools. Informed consent was signed by the parents or legal guardians of the children, and oral assent was provided by each child before testing.

2.1.1. Inclusion criteria

Children with DCD fulfilled four criteria of the Diagnostic and Statistical Manual of Mental Disorders, 5th edition (DSM-5; American Psychiatriac Association, 2013). As recommended by the European Academy of Childhood Disability (EACD; Blank et al., 2019), the Movement Assessment Battery for Children, 2nd edition (MABC-2 Test) (Henderson et al., 2007) was used to assess the level of children's motor competence (criterion A). To evaluate the persistence of motor impairments in activity of daily living (criterion B), the MABC-2 checklist (Henderson et al., 2007) was completed by classroom teachers. The Checklist has excellent internal consistency, Cronbach's α>0.92 (Schoemaker et al., 2012; Kita et al., 2016), good-to-excellent inter-rater reliability, ICC=0.78-0.91 (Ramalho et al., 2013) and proven discriminant validity as a predictor of motor impairment (Schoemaker et al., 2012). The adapted Czech version of the Checklist is also sensitive to DCD and correlates significantly with the MABC-2 Test (rs = -0.31) (Banátová et al., 2022). School psychologists reviewed the medical and behavioral records of each child to assess criteria C and D. Twenty-one children were identified as having ADHD and excluded. Of these, 1 had comorbid autism spectrum disorder (ASD),

and 9 a learning disorder. It should be acknowledged that even though this screening by school psychologists will help to exclude children with diagnosed conditions (e.g., ADHD, ASD), this will not identify children who are undiagnosed at that point as the rate of co-occurring conditions is high for children with DCD. Children who scored ≤ 16 th percentile on the MABC-2 Test and MABC-2 checklist, and met DSM criteria C and D were classified as DCD. DCD subgroups were as follows: m-DCD if MABC-2 total score between the 6th to 15th percentile; s-DCD if ≤ 5 th percentile. The TDC group all performed above the 20th percentile.

2.1.2. Exclusion criteria

Children with an intellectual, physical, or sensory disability, or symptoms of a medical condition affecting movement were excluded.

2.2. Measurement instruments

The MABC-2 Test was administrated in the school gym by a group of examiners who underwent the training and they were certificated and experienced experts according to standardized guidelines in the Examiner's Manual (Henderson et al., 2007). Standardized norms of the MABC-2 Test for the Czech population were used (Psotta, 2014). The MABC-2 Test has good validity and reliability across different age bands (Henderson et al., 2007; Schulz et al., 2011).

2.3. Apparatus and experimental task

The Double-Jump Reaching Task (DJRT) was used to assess online motor control. The DJRT paradigm was programmed using the VIRTOOLS Software Package (3DVIA, 2010), launched on a PC laptop, and displayed on a black Iiyama 43-in touchscreen monitor (Iiyama, Tokyo, Japan). The television was placed horizontally on a height-adjustable table in portrait orientation. All stimuli were displayed against a black background to reduce contrast interference.

The display consisted of a green "home base" circle and three yellow "target" circles, each of 25 mm in diameter. The home base circle was positioned in the middle bottom of the screen, 50 mm from the bottom edge of the display, and three yellow target circles at a distance of 40 mm above the home base, positioned at -20° , 0° , 20° . The home base was lit green when touched with the index finger and switched off at the point when the index finger was lifted from the surface. The child returned their index finger to the home base for each successive trial. To prevent the impact of anticipation, a random delay of 500-1,500 ms was used for target illumination. A successful trial occurred when the child touched the illuminated yellow target location within its circular boundary with the index finger; at the point of contact, the yellow light was extinguished, and an auditory tone emitted, indicating that the trial had ended. 80% of all trials were non-jump: the middle yellow target circle remained lit until touched by the index finger. The remaining 20% were jump trials: the yellow target location switched (or jumped) to either the left or right peripheral location at lift-off (or movement onset).

The AJRT task was administered in a separate block and was identical to the DJRT task, but with the exception that children were required to touch the contralateral target location on jump trials—referred to here as an anti-jump trial. 20% of all trials were anti-jump.

2.4. Procedure

The study was conducted at the university, in a quiet lab with normal fluorescent ceiling light and with no windows to avoid environmental distractions. Hand dominance was determined by observing the child's preferred hand on manual dexterity items of the MABC-2 and self-report. Both typical DJRT and AJRT tasks were performed in two separate 10-15-min sessions, with the DJRT performed first. For the DJRT, each child was asked to stand behind the table adjusted to child waist and to hold their index finger on the green home base circle and then to reach and touch the center of one of the peripheral yellow target circles as quickly as possible, when illuminated. For AJRT task, each child was asked to reach and touch the (outlined) circle on the side opposite the lit circle (or stimulus) defined as anti-jump trial. Each task was demonstrated prior to each experimental session to confirm that children understood the goal of each task and required actions for non-jump, jump, and anti-jump trials. Twenty practice trials were administered in each session. Each test session consisted of 80 trials divided into two blocks of 40 trials including 32 non-jump and 8 DJRT/AJRT, presented in a pseudorandom order (four each side) within 40 trials over the left- and rightside target locations.

2.5. Measures and statistical analysis

For each task, reaction time (RT) and movement time (MT) of each trial were recorded. MT was measured as the time interval between lift-off of the index finger of the dominant hand from the green "home base" to finger touch on the display. A successful trial was defined when the index finger touched within the circular boundary of the designated target location, both extinguishing the target and emitting an auditory signal which indicated successful completion of the trial (Ruddock et al., 2016). Unsuccessful trials (in which no response was initiated) or errors were excluded. A minimum of eight successfully completed jump/anti-jump trials per block was required (Ruddock et al., 2014). Average MT was calculated for jump, anti-jump, and non-jump trials. Next, outliers with values of ± 1.5 SD from the average (Tukey, 1977) were removed from each group for both DJRT and AJRT. That is, if a child scored >1.5 SD in either the DIRT or AJRT, his/her data was removed for further statistical analysis. Consequently, for both DJRT and AJRT three children with s-DCD, three with m-DCD, and 14 TDC were excluded from further data analysis. For the DJRT, online control was measured by the movement time difference between jump and no-jump trials (MTdiff), while for the AJRT, the coupling of online and inhibitory control was measured by the movement time difference between jump and antijump trials (AJMTdiff; Ruddock et al., 2014).

Response errors were also recorded for each task. For both DJRT and AJRT, four error types were as follows: touch-down error (TDE), identified when the index finger touches the areas outside the yellow target spot; anticipatory error (AE) occurred when the index finger was lifted from the green "home base" circle before the yellow target was presented, or within 150 ms of stimulus display (Wilson et al., 1997); center touch error (CTE) occurred when the central target spot was touched instead of one of the peripheral target spots within the jump trial; and wrong-touch error (WTE) occurred when an incorrect (or cued target spot) was touched within anti-jump trial.

For each task, normality assumptions were tested using the Shapiro–Wilk test (p > 0.05). For each task, planned contrasts were conducted to compare MTdiff scores between groups, the first comparing TDC with a weighted average of m-DCD and s-DCD groups, and the second comparing the two DCD sub-groups. Error scores were compared between groups using Mann–Whitney *U* tests, conducted on AE, TDE, WTE, and CTE scores for DJRT and AJRT tasks. The magnitude of group differences was indexed using Cohen's d and interpreted using standard benchmarks: low (d = 0.2), medium (d = 0.5), and large (d = 0.8) effect (Cohen, 1988). To estimate the effect sizes in the non-parametric Mann–Whitney *U* test, the *r* effect size was calculated by dividing the obtained z score by the square root of the sample size number (Fritz et al., 2012). These r values were then transformed into the equivalent of Cohen's d values using the formula $d = [(\sqrt{h})^*r]/[\sqrt{1-r^2}]$ where h = [(n1+n2-2)/n1] + [(n1+n2-2)/n2](Cohen, 1988; Borenstein et al., 2009; Lenhard and Lenhard, 2016). A Spearman's rho correlation test was used to estimate the correlations between MT^{diff}, AJMT^{diff}, and AE, TDE, WTE, and CTE errors, respectively.

3. Results

3.1. Age group

An independent *t*-test showed that there was no significant difference in the mean age of DCD and TDC groups, t(280) = 1.496, p = 0.136 as well as between m-DCD and s-DCD groups, t(80) = -0.915, p = 0.363.

3.2. MTdiff and MT

The results of planned contrasts for $\mathrm{MT}^{\mathrm{liff}}$ and MT in each group are presented in Table 1.

3.3. Errors

For both DJRT and AJRT, the results of the non-parametric Mann–Whitney *U* tests on AE, CTE, WTE, and TDE errors and effect sizes comparing TDC and DCD are presented in Table 2 and Figure 1A, and between m-DCD and s-DCD are presented in Table 2 and Figure 1B, respectively.

3.4. Correlations between MTdiff and errors

Spearman correlations between MT^{diff} and Error scores on each task (DJRT and AJRT) are presented for each participant group in Table 3

4. Discussion

The primary goal of our study was to compare the ability of children with m-DCD, s-DCD, and TDC on two versions of a visual perturbation task: the DJRT that requires (automatic) online

adjustments to a new target location, and the AJRT that requires coupling of rapid online control and response inhibition. Performance of the DJRT was shown to be comparable between TDC, m-DCD and s-DCD groups, while for the AJRT, the DCD group at large performed worse than TDC, while no difference was shown between m-DCD and s-DCD. In addition, correlational data suggested a link between MT^{diff} scores and TDEs across all groups. These findings have important implications for our understanding of cognitive-motor coupling in DCD, discussed below.

Contrary to previous studies (Hyde and Wilson, 2013; Ruddock et al., 2015), our results failed to show performance differences between DCD and TDC on the DJRT, a task that requires rapid online corrections based on a forward estimate of limb trajectory. The earlier Australian studies showed significantly larger MTdiff scores for DCD groups, as well as longer response times to change reach trajectory on jump trials. More specifically, using a target distance of 30 cm, Hyde and Wilson (2011) reported mean scores of 338 ms for DCD compared with 260 ms for TD. As well, in a comparison of DCD, age-matched control (AMC), and younger controls (YC), Hyde and Wilson (2013) showed a similar performance pattern between DCD and YC, suggesting a developmental immaturity in rapid online control: MTdiff for these two groups was 344 and 388 ms, respectively, compared with 275 ms for AMC. In the current study, corresponding MT^{tiff} values were slightly faster overall: 227 ms for DCD and 220 for controls. It is possible that the absence of a group effect here compared with earlier studies is due to differences in participant demographics and contextual factors (i.e., regional vs. large cities), which influence the physical activity levels of the respective DCD samples, discussed below.

For the AJRT, results confirmed deficits in DCD when coupling rapid online (motor) control with inhibitory control (Ruddock et al., 2015). The earlier study by Ruddock et al. (2015) compared DCD and TD groups at three different ages: younger (6-7 years), mid-aged (8-9 years), and older (10-12 years). Younger and mid-aged children with DCD were disadvantaged on anti-jump trials relative to their age-matched controls; e.g., for younger children, AJMTdiff scores were 499 ms versus 352 ms, respectively, and for mid-aged children, 359 ms versus 248 ms. For older children, the difference was not significant between motor groups: 207 ms versus 210 ms. In the current study, AJMT^{diff} was higher for the total DCD group (597 ms) than TD (533 ms). Taken together, as cognitive control develops steadily over the childhood period and beyond (Friedman et al., 2009; Luna, 2009), it is likely that the reduced level of performance on the AJRT in children with DCD may reflect delayed development of cognitive control and its coupling to feedforward/predictive motor control. This hypothesis is supported by data showing that group differences are largely confined to younger cohorts, consistent with our study reported here and earlier work (Hyde and Wilson, 2011; Ruddock et al., 2015, 2016).

This deficit in cognitive-motor coupling may explain the performance difficulty that these children display on more complex motor tasks that present cognitive planning, sequencing, or dualtask components (Wilson et al., 2013). This is shown, for example, by motor planning difficulties in DCD when the complexity of the task increases (Krajenbrink et al., 2020, 2021). Immaturity in reciprocal connectivity between frontal and posterior control systems may impair the integration of cognitive control with realtime adjustments to movement trajectory (Ruddock et al., 2015). While deficits in cognitive inhibition appear less pronounced with

TABLE 1 Descriptive statistics for the double jump reaching task (DJRT) and anti-jump reaching task (AJRT): movement time (MT) and movement time difference (MT^{diff}), expressed for each group [typically-developing children (TDC) versus developmental coordination disorder (DCD)], as a function of motor severity [moderate DCD (m-DCD) versus severe-DCD (s-DCD)], and age (younger: 6–8 years; older: 9–12 years).

DJRT								
	Groups	N	Mean	Std. deviation	t	р	d	
	TDC	201	220.20	79.66	0.425		0.08	
$\mathrm{MT}^{\mathrm{diff}}$	DCD	82	227.06	97.58	-0.435	0.664	0.08	
M I	m-DCD	53	231.51	92.25	0.638	0.524	0.12	
	s-DCD	29	218.93	107.87	0.638	0.524	0.12	
	TDC	68	236.50	95.2	0.227	0.813	0.15	
MT ^{diff} (6–8 years	DCD	37	251.86	105.49	-0.237	0.813	0.15	
old)	m-DCD	27	263.85	89.89	0.214	0.228	0.42	
	s-DCD	10	219.50	139.89	0.214	0.228	0.42	
	TDC	133	211.87	69.33	-0.279	0.781	0.07	
MT ^{diff} (9–12 years	DCD	45	206.67	86.5	-0.279	0.781	0.07	
old)	m-DCD	26	197.92	83.62	-0.927	0.355	0.23	
	s-DCD	19	218.63	91.19	-0.927		0.23	
	TDC	201	623.01	160.01	1.767	0.078	0.22	
MT (no imme)	DCD	82	660.60	188.43	1.767		0.22	
MT (no-jump)	m-DCD	53	653.81	172.15	-0.492	0.623	0.10	
	s-DCD	29	673.00	217.77	-0.492	0.623	0.10	
	TDC	201	843.21	150.31	-2.167	0.021	0.20	
MT (i)	DCD	82	887.66	164.66	-2.10/	0.031	0.28	
MT (jump)	m-DCD	53	885.32	155.15	0.105	0.054	0.04	
	s-DCD	29	891.93	183.69	-0.185	0.854	0.04	

AJRT								
	Groups	N	Mean	Std. deviation	t	р	d	
	TDC	201	532.92	164.11	-2.819	0.005	0.37	
MT ^{diff}	DCD	82	596.54	187.31	-2.819	0.005	0.37	
MI	m-DCD	53	592.25	185.27	-0.306	0.759	0.06	
	s-DCD	29	604.38	194.03	-0.306	0.739	0.00	
	TDC	68	634.63	156.81	-2.81	0.006	0.44	
MT ^{diff} (6–8 years	DCD	37	708.70	183.01	-2.81	0.006	0.44	
old)	m-DCD	27	675.81	180.7	-2.004	0.048	0.79	
	s-DCD	10	797.50	166.27	-2.004	0.048	0.79	
	TDC	133	480.92	142.38	-0.947	0.345	0.16	
MT ^{diff} (9–12 years	DCD	45	504.31	133.81	-0.947		0.16	
old)	m-DCD	26	505.46	148.54	0.064	0.949	0.02	
	s-DCD	19	502.74	114.52	0.064		0.02	
	TDC	201	711.16	184.32	-1.616	0.107	0.81	
MT (no imme)	DCD	82	757.20	203.01	-1.616		0.81	
MT (no-jump)	m-DCD	53	768.00	200.31	0.696	0.487	0.15	
	s-DCD	29	737.45	209.96	0.696	0.487	0.15	
	TDC	201	1224.08	229.12	2 227	0.001	0.54	
MT (i	DCD	82	1353.73	255.99	-3.327	0.001	0.54	
MT (jump)	m-DCD	53	1360.25	319.03	0.226	0.727	0.06	
	s-DCD	29	1341.83	241.95	0.336	0.737	0.06	

TABLE 2 Means, median, standard deviations, minimum, maximum, and comparison of error scores on double jump reaching task (DJRT) and anti-jump reaching task (AJRT) across the groups: typically-developing children (TDC) versus developmental coordination disorder (DCD), and moderate DCD (m-DCD) versus severe-DCD (s-DCD).

	DJRT									
	Groups	Mean	Median	Std. deviation	Minimum	Maximum	U	z	р	d
	TDC	2.30	1	3.75	0	33	7102.5	1.710		0.20
AF	DCD	2.54	2	3.19	0	23	7192.5	-1.718	0.086	0.20
AE	m-DCD	2.11	1	2.16	0	10	650.5	1.166	0.244	0.25
	s-DCD	3.31	2	4.46	0	23	650.5	-1.166	0.244	0.25
	TDC	0.21	0	0.54	0	3	7456.0	-1.894	0.058	0.22
OTTE	DCD	0.39	0	0.84	0	4				0.22
CTE	m-DCD	0.38	0	0.90	0	4		-0.899	0.369	
	s-DCD	0.41	0	0.73	0	3	699.0			0.19
	TDC	0.23	0	0.62	0	4			<0.01	
	DCD	0.57	0	1.04	0	5	6814.0	-3.206		0.38
WTE	m-DCD	0.53	0	1.04	0	5				
	s-DCD	0.66	0	1.04	0	4	704.5	-0.747	0.455	0.16
	TDC	3.54	3	3.04	0	16				
	DCD	0.57	0	1.04	0	5	5925.5	-3.737	<0.001	0.45
TDE	m-DCD	0.53	0	1.04	0	5				
	s-DCD	0.66	0	1.04	0	4	651.5	-1.142	0.254	0.25

	AJRT									
	Groups	Mean	Median	Std. deviation	Minimum	Maximum	U	z	р	d
	TDC	2.31	2	2.41	0	15	7488.5		0.220	0.14
AE	DCD	2.99	2	3.40	0	17	/400.5	-1.227		0.14
AE	m-DCD	2.92	1	3.51	0	17	718.5	-0.496	0.620	0.10
	s-DCD	3.10	2	3.25	0	15	/18.5	-0.496	0.620	0.10
	TDC	0.06	0	0.27	0	2	0120.5	-0.492	0.623	0.05
CTT	DCD	0.04	0	0.18	0	1	8129.5			0.05
CTE	m-DCD	0.02	0	0.13	0	1		-1.148	0.251	0.25
	s-DCD	0.07	0	0.25	0	1	730.0			0.25
	TDC	0.45	0	0.76	0	4	7471.0		0.142	0.17
TAZED.	DCD	0.59	0	0.86	0	4	/4/1.0	-1.468		0.17
WTE	m-DCD	0.58	0	0.81	0	3	743.0	0.202	0.778	0.06
	s-DCD	0.59	0	0.94	0	4	743.0	-0.282	0.778	0.06
	TDC	2.63	2	2.31	0	13		2.515	0.001	0.45
TDE	DCD	4.07	3	3.42	0	21	5945.0	-3.715	<0.001	0.45
TDE	m-DCD	3.28	3	2.08	0	8	5.00 5	2041		0.46
	s-DCD	5.52	4	4.73	0	21	560.5	-2.041	0.041	0.46

 $AE, anticipatory\ error;\ CTE,\ central\ touch\ error;\ WTE,\ wrong-touch\ error;\ TDE,\ touch-down\ error.$

age over childhood (Ruddock et al., 2015), the combination of motor skill and executive function difficulties in children is likely to be a risk factor for persisting DCD (Wilson et al., 2020). Overall, the difficulty in inhibitory control and its integration with online motor control is a fundamental issue in children with DCD when performing speeded pointing movements.

Comparable performance of m-DCD and s-DCD sub-groups on the DJRT is somewhat at odds with sub-group differences observed for manual dexterity (e.g., pegboard placement, threading and drawing items of the MABC-2; McQuillan et al., 2021). Perturbation trials on both the DJRT and AJRT require rapid responses performed under open-loop control, while simple manual dexterity tasks are

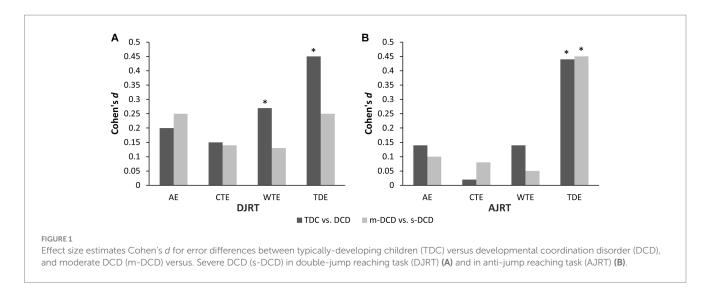


TABLE 3 Spearman correlations between movement time difference (MT^{diff}) and anticipatory error (AE), central touch error (CTE), wrong-touch error (WTE), touch-down error (TDE) on each task [double-jump reaching task (DJRT)] and anti-jump reaching task (AJRT)], presented as a function of group [typically developing children (TDC) versus developmental coordination disorder (DCD), and moderate-DCD (m-DCD) versus severe-DCD (s-DCD)].

DJRT								
			AE	СТЕ	WTE	TDE		
TDC	MT ^{diff}	rs	0.00	0.24	0.05	0.15		
(n = 201)	M1	p	0.953	< 0.001	0.443	0.030		
DCD	a complicat	rs	-0.17	0.26	-0.04	0.35		
(n = 82)	MT ^{diff}	р	0.115	0.018	0.693	< 0.001		
m-DCD	MT ^{diff}	rs	-0.08	0.40	-0.13	0.31		
(n = 54)	MII	р	0.565	0.003	0.351	0.023		
s-DCD	Martiff	rs	-0.34	0.01	0.11	0.40		
(n = 28)	MT ^{diff}	p	0.063	0.941	0.549	0.031		

AJRT									
			AE	CTE	WTE	TDE			
TDC	MT ^{diff}	rs	0.94	0.15	0.22	0.29			
(n = 201)	MI	Р	0.183	0.028	0.002	< 0.001			
DCD	MT ^{diff}	rs	0.243	0.144	0.266	0.517			
(n = 82)	WII	p	0.028	0.197	0.016	< 0.001			
m-DCD	MT ^{diff}	rs	0.17	0.08	0.30	0.46			
(n = 54)	M I ····	Р	0.202	0.561	0.024	< 0.001			
s-DCD	MT ^{diff}	rs	0.38	0.19	0.17	0.64			
(n = 28)	IVI I****	Р	0.041	0.310	0.378	< 0.001			

more closed-loop (or feedback dependent). Children with DCD "live on feedback" (Clark, personal communication, 2011) — a mode of control that is not optimal for tasks that demand rapid online corrections. Put another way, because the planning process is not complete before the start of a task (aka IMD), children with DCD rely more on feedback over the course of movement, and therefore have a slower, more iterative mode of motor control, adjusting their movements in successive steps. This raises the intriguing hypothesis

that the nature of the motor task (and the attendant demands it imposes on open-loop motor control) will determine whether performance difficulties are generalized across DCD sub-groups. Put another way, in the case of anti-jump reaching, demands on cognitive-motor coupling were complex enough to influence the performance of children with DCD, regardless of their motor severity.

More frequent touch errors in DCD (i.e., TDEs on both tasks and CTEs on the AJRT) suggest a generalized difficulty with endpoint and/or trajectory control, seen also in a range of other target-directed pointing and reaching tasks (Mandich et al., 2002; Wilmut et al., 2007; Hyde and Wilson, 2013). At the sub-group level, more TDEs in s-DCD relative to m-DCD on the AJRT suggests that endpoint control is more compromised in s-DCD under an inhibitory load. The absence of any group difference on AEs or WTEs suggests that children were well-oriented to task instructions and performed consistently in reference to task goals. Future research should examine performance accuracy on other measures of response inhibition and cancellation to determine the effect of different levels of response expectancy on performance.

Some distinctions in demographics and context between the Czech Republic and Australian study, may explain some of the discrepancies between our current and earlier studies. There has been accumulating evidence that residential context (or physical environment) is one of the main determinants of children's physical activity (Kimbro et al., 2011; Sharp et al., 2015), correlated also with physical fitness and motor coordination (Amador-Ruiz et al., 2018; Gallotta et al., 2022). In the earlier Australian studies, children were drawn from the large city of greater Melbourne, mainly its densely populated inner suburbs (Hyde and Wilson, 2013; Ruddock et al., 2015). Although we did not measure physical activity specifically, children in the Czech sample - recruited from the regional city of Olomouc and surrounding communities in Moravia - were more likely to engage in outdoor recreational activities than those in the Australian (urban) sample. One hypothesis worth testing is whether higher levels of physical activity in children meeting criteria for DCD may inoculate them against more severe functional impairments.

Our findings have some important implications for practitioners who work with DCD, most notably the importance of considering cognitive load when designing training tasks. Such tasks should be scaled in difficulty not only in motoric terms but also cognitive. In the case of dual-tasks, for example, dual-task interference tends to

be higher for more complex primary motor tasks compared with simple tasks, and the experience of fatigue is much higher in DCD (Krajenbrink et al., 2023). Cognitive-motor dual-tasks may be used as effective training tools, much like that demonstrated in the neurorehabilitation field (Li et al., 2020; Pereira Oliva et al., 2020; Johansson et al., 2023).

In sum, our study suggests that deficits in cognitive-motor coupling are prominent in children with DCD, regardless of the severity of motor skill impairment. Specifically, difficulties integrating cognitive control when performing a speeded task presents both a speed and accuracy cost. The likely downstream effect is performance difficulty on complex tasks that involve both visuomotor coordination and cognitive processing, e.g., dual-tasks like navigating on foot while solving a cognitive task, or sequential motor tasks that require cognitive problem solving. Future research should consider involving older children and adults, careful screening of comorbid conditions that may impact inhibition, prior levels of physical activity (i.e., motor experience), and consideration of IQ and its relationship to executive function and visio-spatial constructional ability (Vaivre-Douret et al., 2020).

Data availability statement

The datasets presented in this article are not readily available because of Human Subjects' protections. Requests to access the datasets should be directed to RA, reza.abdollahipour@upol.cz.

Ethics statement

The studies involving humans were approved by the Ethical Committee of the Faculty of Physical Culture, Palacký University Olomouc (FTK 46/2020), and participating schools. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin.

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Author contributions

RA, LV, KB, LB, TK, ZS, and PW: conceptualization, methodology, project administration, and visualization. RA, LV, and PW: formal analysis. ZS and PW: supervision. BS: interpretation of results. RA, LV, KB, LB, TK, ZS, BS, and PW: data curation, investigation, drafting, and review and editing. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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EDITED BY
Catherine Purcell,
Cardiff University, United Kingdom

REVIEWED BY Paola Chivers, University of Notre Dame Australia, Australia Reza Abdollahipour, Palacký University, Olomouc, Czechia

*CORRESPONDENCE
Richard Mills

☑ richard.mills@mmu.ac.uk

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Children with developmental coordination disorder are less able to fine-tune muscle activity in anticipation of postural perturbations than typically developing counterparts

Carla Harkness-Armstrong¹, Emma F. Hodson-Tole^{2,3}, Greg Wood^{3,4} and Richard Mills^{3,4}*

¹Centre for Physical Activity in Health and Disease, Division of Sport, Health, and Exercise Sciences, Brunel University London, Uxbridge, United Kingdom, ²Department of Life Sciences, Manchester Metropolitan University, Manchester, United Kingdom, ³Manchester Metropolitan University Institute of Sport, Manchester, United Kingdom, ⁴Department of Sport and Exercise Sciences, Manchester Metropolitan University, Manchester, United Kingdom

The majority of children with developmental coordination disorder (DCD) struggle with static and dynamic balance, yet there is limited understanding of the underlying neuromechanical mechanisms that underpin poor balance control in these children. Eighteen children with DCD and seven typically developing (TD) children aged 7–10 years stood with eyes open on a moveable platform progressively translated antero-posteriorly through three frequencies (0.1, 0.25 and 0.5 Hz). Myoelectric activity of eight leg muscles, whole-body 3D kinematics and centre of pressure were recorded. At each frequency, postural data were divided into transition-state and steady-state cycles. Data were analyzed using a linear mixed model with follow-up Tukey's pairwise comparisons. At the slowest frequency, children with DCD behaved like age-matched TD controls. At the fastest frequency, children with DCD took a greater number of steps, had a greater centre of mass variability, had a greater centre of pressure area, and tended to activate their muscles earlier and for longer than TD children. Children with DCD did not alter their postural response following prolonged exposure to platform movement, however they made more, non-structured postural adjustments in the medio-lateral direction as task difficulty increased. At the faster oscillation frequencies, children with DCD adopted a different muscle recruitment strategy to TD children. Activating their muscles earlier and for longer may suggest that children with DCD attempt to predict and react to postural disturbances, however the resulting anticipatory muscle excitation patterns do not seem as finely tuned to the perturbation as those demonstrated by TD children. Future work should examine the impact of balance training interventions on the muscle recruitment strategies of children with DCD, to ensure optimal interventions can be prescribed.

KEYWORDS

balance, postural control, dyspraxia, electromyography, motor control, entropy halflife

1. Introduction

Developmental coordination disorder (DCD) is a movement disorder characterized by reduced motor competence and poor motor coordination, in the absence of other identifiable neurological and/or medical disorders (American Psychiatric Association, 2013). Affecting 5–6% of school-aged children (Zwicker et al., 2012), children with DCD experience significant problems in their fine and/or gross motor skills (Geuze et al., 2001). Most children with DCD also experience significant difficulties with both static and dynamic balance, which can lead to secondary issues such as non-participation in physical activity (Fong et al., 2011) and an increased risk of tripping and falling (Scott-Roberts and Purcell, 2018). As balance is integral in the successful performance of most functional skills (Huxham et al., 2001), it is essential to study the underlying mechanisms that may underpin poor balance control in children with DCD, to ensure that optimal interventions can be prescribed.

It is well established that, even for highly repetitive or simple balance tasks, human movement patterns are varied (Hausdorff, 2007; Turnock and Layne, 2010). However, this variation is not random, with patterns that can be quantified evident in the changes that occur. This time-based organization of variation, or structure, in movement patterns is recognized as an important feature of a neuromuscular system that can adapt to perturbations and changes in the surrounding environment (Bolton, 2015). Variation in walking characteristics of typically developing (TD) children (age 3-14 years) is less structured (more random) than those of adults (Hausdorff et al., 1999). Therefore, studying structure within movement patterns can reveal variations in the growth and maturation of the motor control system. Structure also exists in the muscle activation and coordination that drives movements (Hodson-Tole and Wakeling, 2017; Wakeling and Hodson-Tole, 2018). These structures can change in response to postural control challenges (Ferrari et al., 2020), highlighting the importance of neuromuscular drive in determining motor behaviors.

Postural control can be distinguished into reactive (feedback) or anticipatory (feedforward) responses, whereby postural adjustments are either made subsequent, or prior, to a balance perturbation. Responses to postural disturbances also scale to the level of postural threat (Adkin et al., 2000) and depend of the size of the perturbation. For instance, during smaller perturbations, an ankle strategy is often effective, whereby torque generated about the ankle joint is sufficient to maintain balance (Massion, 1994). In larger perturbations, a more severe response may be required, such as a hip strategy, whereby large, rapid movements are generated about the hips to regain centre of mass (COM) equilibrium (Horak and Nashner, 1986). As we develop across the lifespan, we learn to adapt to different perturbations through mechanisms that are dynamic and flexible (Haddad et al., 2013). However, individuals with DCD often present with a poor organization of body movements in relation to the global environment (Green and Payne, 2018), therefore it is important to assess the postural responses of those with DCD during balance perturbations.

Reactive and anticipatory mechanisms of postural control have been described previously for single discrete perturbations in children with DCD. During unexpected perturbations, Cheng et al. (2018) found that children with DCD reacted later than TD children to a forward push, whereas Fong et al. (2015) reported no group differences when reacting to a backward moving platform. During planned movements, children with DCD presented with fewer anticipatory muscle activations when kicking a ball and climbing stairs (Kane and Barden, 2012), and had a shorter duration between muscle activity onset time and peak activation than TD children during a *Y*-balance test, which was suggested to be a potential mechanism to compensate for a less-effective feedforward control system (Yam and Fong, 2019). Whilst knowledge of postural control during single perturbations is important, it is also essential to assess movement strategies during continuous dynamic situations (such as a moving base of support), to fully understand the underlying mechanisms that may contribute to poor balance control (Horak et al., 2009). The oscillating platform paradigm causes both reactive and anticipatory postural control strategies to be generated to overcome the same perturbation (Mills and Sveistrup, 2018).

While these reactive and anticipatory postural control strategies have been studied in children with other motor impairments (e.g., cerebral palsy; Mills et al., 2018), to our knowledge, they have not been studied in children with DCD during continuous dynamic movement. Additionally, no previous work has studied the structure of postural sway characteristics in children with DCD, nor evaluated the association with muscle activation and coordination. Therefore, the primary aim of this study was to compare postural responses to continuous platform oscillations between children with DCD and TD children. The secondary aim of this study was to determine if children with DCD were able to modify postural responses after prolonged exposure to platform movement. We hypothesized that children with DCD would be less able to adapt their postural responses compared to TD children after prolonged exposure to platform movement.

2. Materials and methods

2.1. Participants

Eighteen children with DCD and seven TD children participated in this study. Children with DCD were recruited through parental support groups on social media (e.g., Facebook). TD children were recruited via social media and convenience sampling (e.g., sibling of child with DCD). Participant characteristics are shown in Table 1. Children in the DCD group satisfied the Diagnostic and Statistical Manual of Mental Disorders

TABLE 1 Mean \pm standard deviation participant characteristics.

	DCD	TD
N (male/female)	18 (13/5)	7 (2/5)
Age (years)	9 ± 1	9 ± 1
Height (m)	1.41 ± 0.07	1.31 ± 0.09
Body Mass (kg)	38.9 ± 9.6	29.7 ± 12.4
MABC-2 Percentile (Overall)	2±3	-
MABC-2 Percentile (Balance)	3±3	56±25
ADHD	90 ± 13	-

DCD, Children with developmental coordination disorder; TD, typically developing children; MABC-2, movement assessment battery for children.

(DSM-5) criteria (American Psychiatric Association, 2013), whereby they exhibit substandard motor ability, relative to their chronological age, since early development. Prior to data collection, parents/guardians completed the Developmental Coordination Disorder Questionnaire (Wilson et al., 2009) to confirm that their child had significant movement difficulties that interfered with balance, did not suffer from any general medical condition known to affect sensorimotor function, and had no diagnosed learning difficulties (DSM-5 criteria B, C, D). If any known medical conditions or learning difficulties were identified, these children were excluded from the study. Children with DCD were required to score ≤ 5th percentile (overall), reflecting definite motor impairment (DSM-5 criteria A), and ≤15th percentile (balance subscale), reflecting 'risk' of motor impairment, on the Movement Assessment Battery for Children, Second Edition (MABC-2; Henderson et al., 1992). TD children were required to score > 15th percentile (balance subscale), reflecting no motor impairment. Parents/guardians also completed the Attentional Deficit Hyperactivity Disorder (ADHD) Rating Scale - VI (DuPaul et al., 1998). The institutional research ethics committee granted ethical approval. Written informed consent was obtained from parents/guardians and written assent given by children, in accordance with the Declaration of Helsinki.

2.2. Experimental protocol

The experimental protocol for this study was adapted from others described previously (Bugnariu and Sveistrup, 2006; Mills and Sveistrup, 2018). Participants stood upright with eyes open and feet shoulder width apart in the centre of a moveable platform. The platform was driven by electromagnetic propulsion, controlled via custom written software (Labview v19 SP1, National Instruments, Austin, Texas) through a DAQ card (USB-6210, National Instruments). Participants were instructed to maintain balance and avoid taking steps unless necessary. If steps were taken, participants were instructed to return to their initial position as quickly as possible. The platform translated 10 cm peak-to-peak in the antero-posterior direction. Two trials of ten sinusoidal oscillations at a frequency of 0.1 Hz, twenty oscillations at 0.25 Hz, and forty oscillations at 0.5 Hz (Figure 1A) were presented, with frequency changes presented sequentially and automatically. Participants were aware that platform frequency would increase, however they were not informed as to when this would occur.

Full body kinematics were collected at 100 Hz using a 10-camera motion analysis system (Qualisys v2021.1, Gothenburg, Sweden). Passive retro-reflective markers (n=47) were positioned on all body segments (modified Plug-in Gait model). Two additional markers were positioned on the oscillating platform to record its position. For outcome measures described below, head and trunk angle, and whole-body COM were calculated in Visual 3D (v2021.06.2, C-Motion, Rockville, MD). Bilateral surface electromyography (EMG; Delsys Inc., Natick, United States) from rectus femoris (RF), biceps femoris (BF), tibialis anterior (TA), and medial gastrocnemius (MG) muscles were collected at 1000 Hz in Qualisys. Centre of pressure data were collected using a Kistler force plate (Type 9281B, Kistler Instrument Corp., Winterthur, Switzerland) at 1000 Hz. Force data were recorded in BioWare software (v5.4.3.0), synchronized to motion data by the Qualisys trigger.

2.3. Outcome measures

At each platform frequency, the number of cycles containing a step were manually counted at the time of data collection and verified using motion capture data. Centre of pressure (COP) area was calculated using a 90% confidence ellipse. COM displacement variability in the antero-posterior and medio-lateral directions were assessed in terms of each signals standard deviation (SD) and the timescale over which short-term fluctuations in the signal persisted, calculated as the Entropy Halflife (EnHL). In the antero-posterior direction, both absolute and adjusted data are presented, whereby platform displacement was subtracted from COM data. To calculate EnHL, the COM in the antero-posterior and medio-lateral directions were split into equal length epochs containing all cycles within a single platform oscillation frequency. Each signal was high-pass filtered (2nd order Butterworth, 10 Hz cut-off) to attenuate temporal oscillations imposed by the platform movement (Figure 2A). The filtered signal was standardized (mean = 0, SD = 1) and a reshape timescale approach (Zandiyeh and Von Tscharner, 2013) used to generate restructured time series with increasing time intervals (1 ms - 6s) between consecutive data points (Figure 2B). The sample entropy (SampEn) of each reshaped signal was calculated using a freely available software (Goldberger et al., 2000), with m=1 and r=0.2. SampEn provides the conditional probability that a time series of m data points remains affiliated, with a tolerance of r, if a data point is added to it (Richman and Moorman, 2000). Resulting SampEn values increase (indicating less regularity) as the reshape scale increases, reflecting the breakdown of short-term signal fluctuations (Figure 2B). The series of SampEn values produced were normalized to the maximum SampEn calculated for the original time series (when m = 0 and r = 0.2). This normalization means the reshape timescale at which SampEn = 0.5 represents the timescale at which the signal transitions from containing regular, structured fluctuations to being random (Zandiyeh and Von Tscharner, 2013) called the EnHL. These analyses were completed using custom written code in Wolfram Mathematica (version 11.1.1).

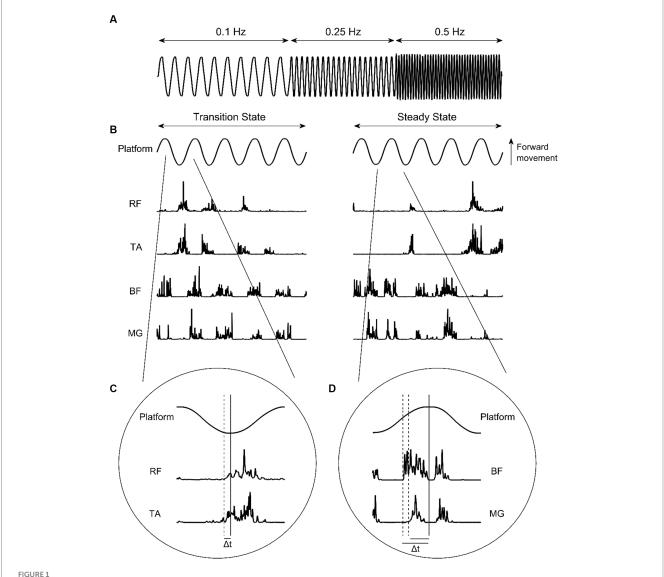
Head anchoring index (AI) was calculated using Eq. (1) (Mills and Sveistrup, 2018) to determine the stabilization strategy of the head in relation to both the global environment and the trunk segment:

$$AI = \left[\sigma_r^2 - \sigma_a^2\right] \div \left[\sigma_a^2 + \sigma_r^2\right] \tag{1}$$

where σ_a is the SD of the absolute head angle relative to the global coordinate system, and σ_r is the SD of the head relative to the trunk segment. A positive AI indicates a head-stabilized-in-space strategy. A negative AI indicates a head-stabilized-to-trunk strategy.

To calculate muscle activity onset latencies, EMG signals were decomposed into time-frequency space using an EMG specific wavelet analysis approach (Von Tscharner, 2000). Specifically, a filter bank of k=11 non-linearly scaled wavelets with central frequencies spanning 6.90–395.44 Hz was used to resolve the EMG signal intensities into time/frequency space. Total intensity was calculated as the sum of the signal power contained within wavelets $1 \ge k \le 10$, providing a representation of the signal power at each time point whilst removing effects of low frequency signal components (i.e., contained within the first wavelet, k=0).

The occurrence of muscle activity in respect to the relevant platform change of direction were identified manually using the *ginput*



(A) Platform oscillation frequencies. (B) Platform oscillations at 0.5 Hz and corresponding EMG intensities from the rectus femoris (RF), tibialis anterior (TA), bicep femoris (BF), and medial gastrocnemius (MG) during transition-state and steady-state cycles. (C) Identification of anterior muscle activity onset. (D) Identification of posterior muscle activity onset. Solid vertical lines indicate platform change of direction. Dashed vertical lines indicate muscle activity onset. At indicates muscle onset latency.

function in MATLAB (R2022a, MathWorks Inc., Natwick, MS, USA). To be considered for inclusion as muscle activity, EMG intensity had to meet or exceed two SDs above baseline (defined as the quiet period prior to trial start) and last for more than 50 ms (Mills and Sveistrup, 2018). For RF and TA, this was when the platform transitioned from backward to forward direction. For BF and MG, this was when the platform transitioned from forward to backward direction (Figures 1B–D). To remove subjectivity of this method, a custom MATLAB script was subsequently used. Firstly, the EMG intensities at the manually identified muscle activity onset times were determined, and averaged for each muscle to calculate an onset threshold. Activity onset times were then automatically adjusted using the script, so that all activity onsets for a specified participant occurred when EMG intensity surpassed their defined muscle threshold. Lastly, the total activity time of each muscle 'burst' was calculated as the time

between activity onset, and the first subsequent instance that the EMG intensity envelope dropped below the onset threshold. All muscle activity data were expressed as a percentage of half-cycle time, to allow for comparisons between different platform frequencies. Muscle activity bursts were coded as anticipatory where they occurred before change of direction, and as reactive where they occurred after change of direction.

For AI and EMG data, platform frequencies were sub-divided into 'transition-state' and 'steady-state'. Transition-state was defined as the first 3 cycles at 0.1 Hz, and the first 5 cycles at 0.25 and 0.5 Hz. Steady-state was defined as a period within the last half of each frequency that contained 5 cycles without stepping at 0.1 Hz, and a period of 8–10 cycles without stepping at 0.25 and 0.5 Hz, whereby the movement of the platform is predictable (Bugnariu and Sveistrup, 2006).

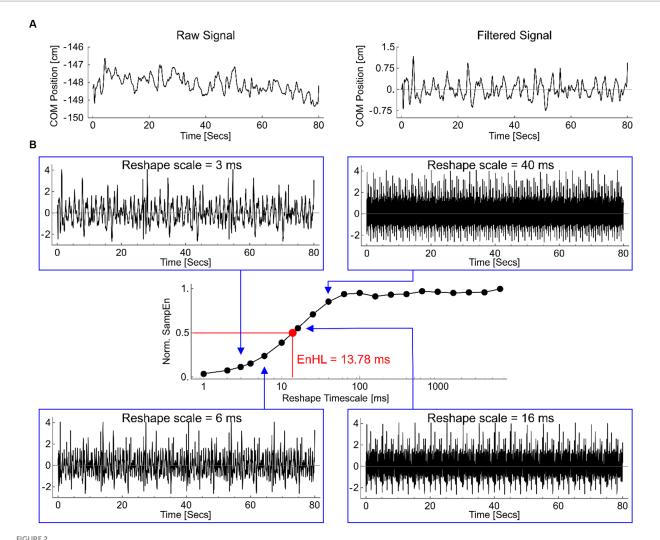


FIGURE 2 (A) An example medio-lateral COP displacement signal, from 0.25 Hz platform oscillation, as recorded (left) and after filtering (right). (B) Filtered signal reshaped at timescales of 3 ms (top left), 6 ms (lower left), 16 ms (lower right) and 40 ms (top right). Note the original repeating pattern of fluctuations is reduced as the reshape timescale increases. The normalized sample entropy values (SampEn) for each of these signals, and for all other reshape timescales, are shown in the central graph (log scale on x-axis). The timescale at which the normalized SampEn = 0.5 is highlighted (red), defining the EnHL for this signal as 13.78 ms.

2.4. Statistical analysis

All statistical analyses were completed using RStudio (RStudio 1.3.959). Descriptive statistics (Table 1) are reported as mean ± standard deviation (SD). A linear-mixed model (LMM; lme4 package; Bates et al., 2015) was developed to quantify differences for each outcome measure (number of steps, COM SD, COM EnHL, COP area, head anchoring index, muscle onset latency and total excitation time) between groups (DCD vs. TD), platform frequencies (0.1 Hz vs. 0.25 Hz vs. 0.5 Hz) and platform state (transition vs. steady-state) (fixed effects). Participant ID was included as a random effect. Assumptions of linearity and normality distributions of the model were checked visually, and homogeneity of variance assessed using Levene's Test (p > 0.05; Levene, 1960). Estimated means for each variable were derived from the model using the emmeans package, and are reported as mean ± standard error (SE). To identify between-group and between-state differences, Tukey's pairwise comparisons were conducted. Statistical significance was set at p < 0.05. Effect sizes (ES) were also calculated using the *effsize* package, and considered trivial (< 0.2), small (≥ 0.2 to <0.6), moderate (≥ 0.6 to <1.2), large (≥ 1.2 to <2.0), or very large (≥ 2.0) (Batterham and Hopkins, 2006), and are presented as ES \pm 90% confidence intervals. ES were considered unclear if the 90% confidence intervals included substantial positive and negative values ($\geq \pm 0.2$; Hopkins et al., 2009).

3. Results

3.1. Stepping responses

One child with DCD took steps during 1 cycle at $0.1\,\mathrm{Hz}$ and $0.25\,\mathrm{Hz}$. Three children with DCD took steps during 1 cycle at $0.25\,\mathrm{Hz}$. No TD children took any steps at either $0.1\,\mathrm{Hz}$ or $0.25\,\mathrm{Hz}$. At $0.5\,\mathrm{Hz}$, 16 out of 18 children with DCD, and six out of seven TD children took steps throughout the trial. LMM estimated means showed that

children with DCD took steps during more cycles to maintain balance than TD children at 0.5 Hz (8 \pm 1 vs. 3 \pm 2, large ES, 1.17 \pm 0.47, p = 0.129), and at the other two frequencies (vs. 0.1 Hz, 8 \pm 1 vs. 0 \pm 1, large ES: 1.75 \pm 0.35; p < 0.001; vs. 0.25 Hz, 8 \pm 1 vs. 0 \pm 1, large ES: 1.72 \pm 0.35; p < 0.001).

3.2. COM variability

3.2.1. COM standard deviation

Children with DCD had a greater LMM estimated COM SD, than TD children in the medio-lateral direction at 0.1 Hz (1.20 \pm 0.19 vs. 0.67 \pm 0.29 cm, moderate ES, 0.75 \pm 0.83, p = 0.661) and 0.5 Hz (1.84 \pm 0.19 vs. 0.97 \pm 0.29 cm, large ES, 1.26 \pm 0.83, p = 0.133), and antero-posterior direction at 0.25 Hz (4.10 \pm 0.13 vs. 3.67 \pm 0.20 cm, large ES, 1.25 \pm 1.16, p = 0.465) and 0.5 Hz (4.58 \pm 0.13 vs. 3.77 \pm 0.20 cm, very large ES, 2.30 \pm 1.16, p = 0.019) (Figure 3). In children with DCD, COM SD increased with task difficulty in both medio-lateral (moderate ESs: 0.93–0.96; p > 0.05) and antero-posterior (large ESs: 1.36–1.70; p < 0.01) directions (Figure 3), whereas there was no change in TD children (unclear ESs; p > 0.05). When platform displacement was accounted for in the antero-posterior direction, all observed differences between groups and platform frequencies were still present (Figures 3C,F).

3.2.2. COM entropy halflife

At 0.1 Hz, children with DCD had a longer LMM estimated COM EnHL in the medio-lateral direction $(20.49 \pm 0.99 \text{ vs. } 17.04 \pm 1.57 \text{ ms},$ moderate ES, 0.88 ± 0.79 , p = 0.441), and a shorter COM EnHL in the antero-posterior direction (34.14 $\pm\,0.86$ vs. 36.10 $\pm\,1.33$ ms, moderate ES, 0.64 ± 0.81 , p = 0.777) than TD children (Figure 4). In children with DCD, COM EnHL decreased with increased task difficulty in both medio-lateral (moderate [0.1 vs. 0.25 Hz, p = 0.438] to large [0.1 & 0.25 vs. 0.5 Hz, p < 0.05] ESs: 0.68–1.95) and antero-posterior directions (large to very large ESs: 1.81–6.74; *p* < 0.001). COM EnHL differences in the antero-posterior direction were still present when accounting for platform displacement (moderate to large ESs: 0.86-2.30; p<0.05) (Figure 4C). In TD children, COM EnHL was similar regardless of platform frequency in the medio-lateral direction (Figure 4A), however COM EnHL decreased with increased task difficulty in the antero-posterior direction (very large ESs: 2.72-7.46; p < 0.001). When accounting for platform displacement, differences in 0.1 vs. 0.5 Hz (very large ES: 2.02 ± 0.93 ; p = 0.008) and 0.1 vs. $0.25 \,\mathrm{Hz}$ (very large ES: 2.36 ± 0.93 ; p = 0.001) were still present in both groups (Figure 4C).

3.3. COP area

No difference in LMM estimated COP area was detected between groups at 0.1 Hz (46 ± 30 vs. $35\pm39\,\mathrm{cm}^2$, unclear ES, 0.12 ± 0.95 , p=0.999) and 0. 25 Hz ($51\pm30\,\mathrm{vs.}$ $46\pm39\,\mathrm{cm}^2$, unclear ES, 0.05 ± 0.95 , p=0.999), however children with DCD had a greater COP area than TD children at 0.5 Hz ($250\pm30\,\mathrm{vs.}$ $113\pm39\,\mathrm{cm}^2$, large ES, 1.60 ± 0.95 , p=0.069). COP area increased with task difficulty in both children with DCD (very large ESs: 2.33-2.39; p<0.001) and TD children (moderate ESs: 0.78-0.91; p>0.05).

3.4. Anchoring index

Despite some individual participants adopting a head-stabilized-in-space strategy or head-stabilized-to-trunk strategy, average data indicate no clear head stabilization strategy in either group (AI of <0.2 or >0.2; Figure 5). There were no group differences detected during transition or steady-state cycles at any platform frequency (unclear ESs; p>0.05), and no state differences detected in either group (unclear ESs; p>0.05).

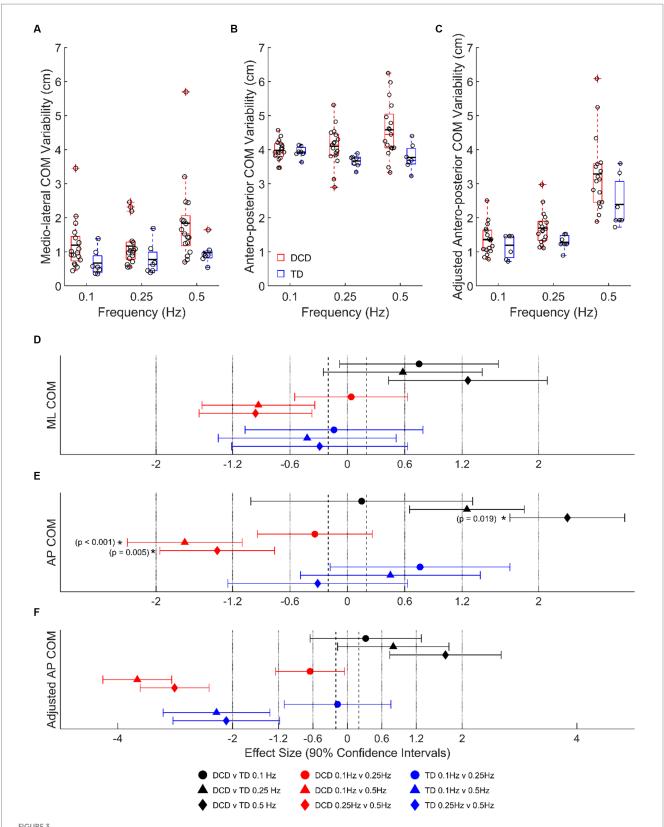
3.5. Muscle activity

LMM estimated muscle activity data for transition-state and steady-state cycles are shown in Table 2. In general, both groups tended to activate their muscles earlier and for longer as task difficulty increased. At 0.25 Hz, muscle excitation occurred earlier in children with DCD in the RF (moderate ES: 1.08 ± 1.07), TA (large ES: $1.62\pm1.32)$ and MG (large ES: 1.49 ± 0.85) during transition-state cycles, and in the MG (moderate ES: 1.07 ± 0.85) during steady-state cycles than TD children. Muscle excitation duration of the MG was longer in children with DCD (moderate ES: 0.93 ± 1.03) than TD children during steady-state cycles. At 0.5 Hz, muscle excitation of the MG occurred earlier in children with DCD during both transitionstate (moderate ES: 1.13 ± 0.79) and steady-state cycles (large ES: 1.90 ± 0.81), and for longer in the BF (moderate ES: 1.02 ± 1.04) and MG (large ES: 1.58 ± 0.92) during transition-state cycles, and in the BF (moderate ES: 1.05 ± 1.04) and MG (large ES: 1.31 ± 0.94) during steady-state cycles than TD children.

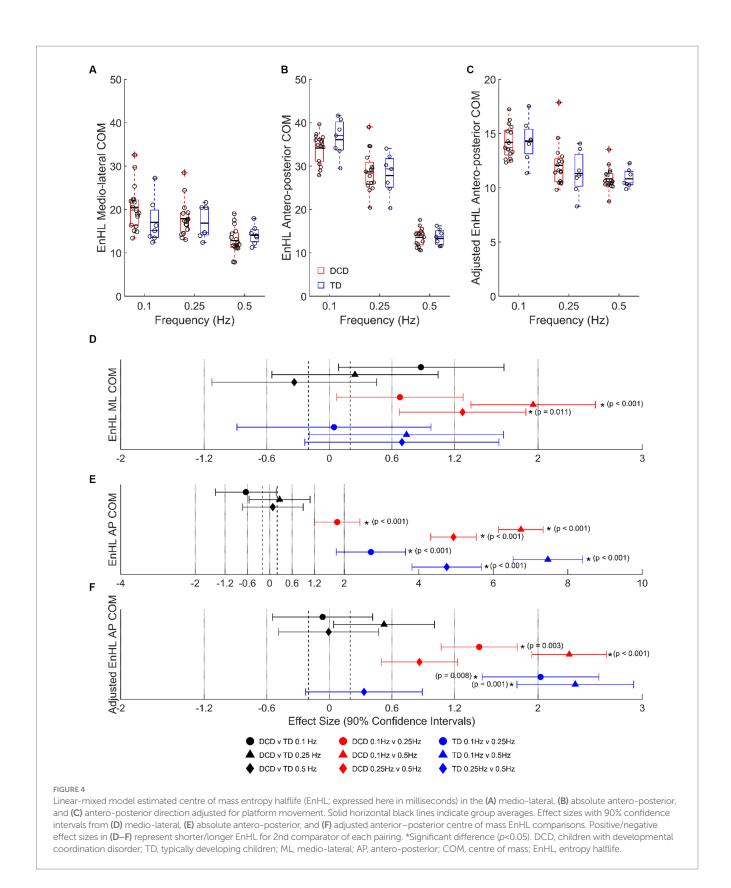
At 0.25 Hz, children with DCD generally activated their muscles at a similar time between platform states (except TA excitation occurred later in steady-state), however excitation duration was longer in steady-state cycles for the TA (small ES: 0.49 ± 0.61) and GM (small ES: 0.52 ± 0.58) than in transition-state cycles. During steady-state cycles, TD children tended to activate their muscles earlier and for shorter durations than in transition-state cycles, however all effect sizes were unclear. At $0.5\,\mathrm{Hz}$, no clear trends were observed in muscle excitation onset time or excitation duration between platform states in either group. Full ES comparisons can be found in Supplementary Figures S1, S2.

4. Discussion

This study is the first to assess the postural and neuromuscular responses of children with DCD using a continuous balance perturbation paradigm. As expected, children with DCD were generally more unstable than TD children, particularly at the highest platform frequency. An increase in the number of children who took steps at 0.5 Hz reflects the increased difficulty of the task for both groups (Streepey and Angulo-Kinzler, 2002). However, children with DCD took steps more often than TD children to maintain balance (large ES). Children with DCD also had a greater COM variability (SD) than TD children in both the antero-posterior (large to very large ESs) and medio-lateral (moderate to large ESs) directions (Figure 3), indicating greater postural sway. This was further supported by the greater COP area covered by children with DCD (large ES).

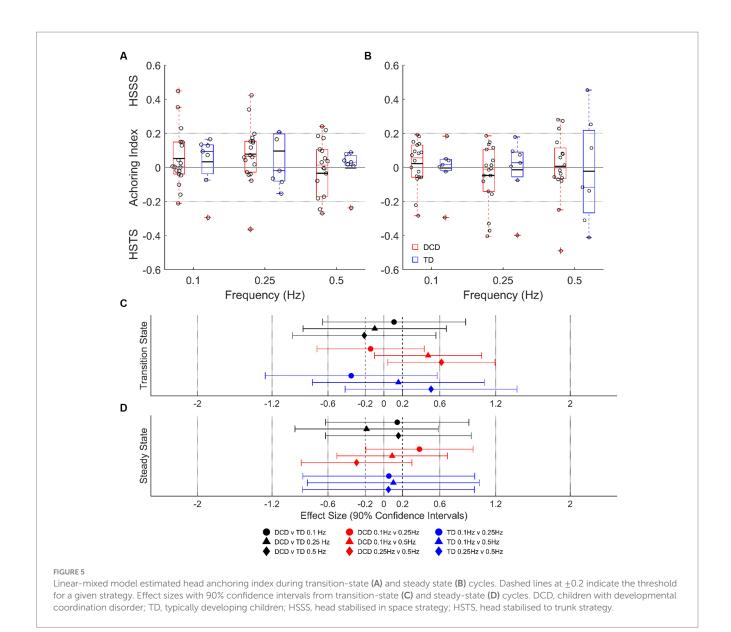


Linear-mixed model estimated centre of mass variability, based on signal standard deviation, in the (A) medio-lateral, (B) absolute antero-posterior, and (C) antero-posterior direction adjusted for platform movement. Solid horizontal black lines indicate group averages. Effect sizes with 90% confidence intervals from (D) medio-lateral, (E) absolute antero-posterior, and (F) adjusted antero-posterior centre of mass variability comparisons. Positive/ negative effect sizes in (D-F) represent smaller/greater variability for 2nd comparator of each pairing. *Significant difference (p < 0.05). DCD, children with developmental coordination disorder; TD, typically developing children; ML, medio-lateral; AP, antero-posterior; COM, centre of mass.



At the fastest platform frequencies, children with DCD tended to adopt a different muscle excitation strategy to TD children. Activating their muscles earlier and for longer may suggest that children with DCD attempt to predict and react to postural disturbances, however the resulting anticipatory muscle excitation patterns do not seem as

finely tuned to the perturbation as those demonstrated by TD children. Additionally, children with DCD made more, non-structured (random) postural adjustments in the medio-lateral direction as task difficulty increased. Therefore, data from the current study indicate an altered neuromuscular coordination in children with DCD, which



should be considered in future training interventions to improve balance control.

Despite the reduced stability of children with DCD, there was no detected difference in their global stabilization strategy compared to TD children. Children with DCD showed no preference for either a head-stabilized-to-trunk strategy, or a head-stabilized-in-space strategy (Figure 5), whereas other populations with known balance deficits, such as children with cerebral palsy (Mills et al., 2018) and adults with Parkinson's disease (Mesure et al., 1999), adopt a head-stabilized-to-trunk strategy. This may be explained by a poor organization of body movements in relation to the global environment, often associated with DCD (Green and Payne, 2018).

Children with DCD did however, adopt a different neuromuscular strategy to TD children at the faster platform frequencies. Generally, the organization of muscle excitation was distal to proximal in children with DCD, indicating an ankle strategy was implemented to maintain balance (Massion, 1994). Whilst this was also the case for the anterior muscles of TD children, there were some instances whereby average posterior muscle excitation was ordered proximal to distal

(Table 2). This may indicate that TD children were able to switch between an ankle and hip strategy to maintain balance (Horak and Nashner, 1986). Children with DCD tended to activate their muscles earlier and for longer than TD children, regardless of platform state (Table 2). Whilst this does suggest that children with DCD attempt to predict and react to postural disturbances (Cordo and Nashner, 1982), the resultant anticipatory muscle excitations are different to those demonstrated by TD children. Thus, a lack of appropriate muscular reactions to balance perturbations may explain poor dynamic balance control in children with DCD.

Previous work has shown that children with DCD do not make postural adaptations when exposed to repeated discrete perturbations (Cheng et al., 2022). During our continuous perturbations, neither group made postural adjustments with prior knowledge of platform movement at the fastest platform frequency, as both muscle excitation onset time and total excitation duration remained similar between transition-state and steady-state cycles. However, this likely reflects the increased difficulty of the task at 0.5 Hz, as TD children were able to make postural adjustments with prior knowledge of platform

TABLE 2 Linear-mixed model estimated mean ± standard error timing of muscle activity during transition-state and steady-state cycles.

			Transitio	n-state		Steady-state				
		Onset la	itency (%)	Total excitation time (%)		Onset latency (%)		Total excitation time (%)		
		DCD	TD	DCD	TD	DCD	TD	DCD	TD	
	RF	9.13 ± 3.72	9.79 ± 4.47	7.03 ± 1.93	2.39 ± 2.44***	3.82 ± 3.03	4.33 ± 4.09	$10.07 \pm 1.67^{\dagger\dagger}$	9.71 ± 2.31****	
0.111	TA	-1.40 ± 4.45	4.45 ± 5.75	7.02 ± 1.51	5.12 ± 1.95	7.08 ± 3.69 ^{††}	2.84 ± 6.25	5.96 ± 1.25	3.71 ± 2.12	
0.1 Hz	BF	5.51 ± 5.98	-10.81 ± 10.69	6.87 ± 2.79	13.53 ± 4.99	0.81 ± 5.98	-15.11±7.94**	6.29 ± 2.79	2.47 ± 3.71 ^{†††}	
	MG	-4.67 ± 3.21	-13.28 ± 5.43**	5.90 ± 1.75	7.59 ± 2.95	-2.80 ± 3.21	$-0.57 \pm 5.01^{\dagger\dagger}$	8.73 ± 1.75 [†]	7.02 ± 2.72	
	RF	-8.64 ± 5.58	8.13 ± 8.21**	21.06 ± 4.74	14.77 ± 6.92	-3.68 ± 5.19	8.66 ± 8.21	17.34 ± 4.38	26.38 ± 6.92	
0.2511	TA	-10.81 ± 4.00	4.44±6.17***	10.09 ± 1.78	9.89 ± 2.71	$-6.62 \pm 3.85^{\dagger}$	-1.16±6.17	12.66 ± 1.69 [†]	9.02 ± 2.71	
0.25 Hz	BF	-16.98 ± 3.88	-10.62 ± 5.89	15.19 ± 2.59	12.92 ± 3.91	-13.82 ± 3.74	-13.01 ± 6.49	16.51 ± 2.49	12.84 ± 4.33	
	MG	-20.64 ± 2.16	-8.58 ± 3.46***	12.41 ± 1.58	11.17 ± 2.53	-22.27 ± 2.16	-13.63 ± 3.46**	$14.98 \pm 1.58^{\dagger}$	10.43 ± 2.54**	
	RF	-17.08 ± 5.46	-3.95 ± 9.23	33.64±7.90	30.84 ± 13.36	-15.69 ± 5.84	-18.43 ± 8.48	42.70 ± 8.46	25.69 ± 12.27	
0.511	TA	-19.84 ± 4.24	-17.07 ± 6.80	25.89 ± 3.29	19.97 ± 5.28	-19.18 ± 4.65	-20.74 ± 6.80	$18.85 \pm 3.44^{\dagger\dagger}$	23.34±5.28	
0.5 Hz	BF	-21.19 ± 3.30	-13.50 ± 4.91	39.64±7.08	18.90 ± 10.43**	-20.32 ± 3.22	-16.11 ± 4.91	44.40 ± 7.08	23.10 ± 10.43**	
	MG	-25.98 ± 2.06	-16.69 ± 3.31**	32.16 ± 3.25	14.47 ± 5.21***	$-29.67 \pm 2.20^{\dagger}$	-14.01 ± 3.31***	29.89 ± 3.45	15.25 ± 5.21***	

Negative onset latencies indicate muscle excitation occurred before platform change of direction.

RF, rectus femoris; TA, tibialis anterior; BF, bicep femoris; MG, medial gastrocnemius; DCD, children with developmental coordination disorder; TD, typically developing children.

movement at 0.25 Hz (Table 2). At 0.25 Hz, TD children activated their muscles earlier and for a shorter duration during steady-state cycles, which may suggest that they were able to better anticipate platform movement compared to transition-state cycles. In contrast, there were no changes in muscle excitation onset times between platform states in children with DCD, and muscle excitation duration was indeed longer in steady-state cycles. Overall, data from the current study indicate an altered neuromuscular coordination in children with DCD, which should be considered in future training interventions to improve balance control.

While children with DCD exhibited greater postural sway than TD children (Figure 3), the structural organization of the anteroposterior COM variability (EnHL) did not differ between groups (Figure 4). This suggests that to maintain balance, the control strategies adopted by children with DCD resulted in a similar temporal organization of the antero-posterior COM movement as TD children, possibly explaining the similarity in the global kinematic outcome measures described above. However, surprisingly, the EnHL of the medio-lateral displacement of children with DCD became shorter as platform difficulty increased, whereas there was no change in TD children. This suggests that children with DCD made more, non-structured (random), postural adjustments in a plane orthogonal to platform movement as task difficulty increased. Previous work has shown those with DCD to explore more action space during a defined task by increasing available degrees of freedom (Golenia et al., 2018). Therefore, this increased, less structured variability in the medio-lateral plane, may be a compensatory mechanism as a result of the way children with DCD manage the degrees of freedom problem (Latash et al., 2007). It may also be explained by a lack of stiffening and/or appropriately organized recruitment of hip ab/adductor muscles, which are important for medio-lateral stability (Winter et al., 1996). However, as we did not measure muscle activity in these muscles, further work is required to confirm or deny this notion.

Some limitations should be acknowledged. Firstly, our sample size is small and does not include an even distribution of male/female participants. While sex differences in postural control have been shown previously in TD children (Smith et al., 2012), exploring sex differences between and within children with DCD and TD children was outside the scope of the current manuscript. Furthermore, it was not possible to accurately explore sex differences due to the insufficient number of data per sub-level (e.g., TD male participants, n = 2). Future work with larger sample sizes is needed. EMG data were only collected for eight lower limb muscles, yet conclusions are generalized to wholebody postural control. Further, our assumption that postural movement in the antero-posterior direction would be solely controlled by flexor/extensor muscles meant that all eight muscles considered for analysis were flexor/extensor muscles. Future work should therefore consider collecting EMG data from more muscles, and consider the role that ab/adductor and rotational muscles may play in ensuring postural stability in the antero-posterior direction. Future work should also consider assessing the EnHL of EMG data, to identify whether there are any differences in the temporal organization of muscle activity.

To conclude, data from the current study indicate that while children with DCD were not able to perform the task as well as TD children (more unstable), they were able to complete the task, actively working toward making similar global postural adjustments as TD children. However, to achieve a similar global stabilization strategy, children with DCD generated this response with a different neuromuscular strategy, activating their muscles earlier and for longer than TD children. Children with DCD also made more, non-structured, movements in a plane orthogonal to platform displacement as task difficulty increased, suggesting they utilize more degrees of freedom to overcome balance perturbations than TD children. Future work should examine the impact of balance training interventions on the muscle excitation patterns and coordination strategies of children with DCD, to ensure that appropriate

^{*}Small, **moderate or ***large effect size difference between DCD and TD.

[†]Small, ††moderate, †††large, or ††††very large effect size difference between transition-state and steady-state cycles.

interventions to improve balance can be prescribed. Future work should also consider the role of attentional deficits of children with DCD on postural control during continuous balance perturbations.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by the Faculty of Science and Engineering Research and Ethics Governance. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin.

Author contributions

CH-A: Data curation, Formal analysis, Writing – original draft. EH-T: Conceptualization, Funding acquisition, Methodology, Supervision, Writing – review & editing. GW: Conceptualization, Funding acquisition, Methodology, Supervision, Writing – review & editing. RM: Conceptualization, Funding acquisition, Methodology, Project administration, Supervision, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fnhum.2023.1267424/full#supplementary-material

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EDITED BY
Catherine Purcell,
Cardiff University, United Kingdom

REVIEWED BY Jennifer Davies, Cardiff University, United Kingdom Jennifer Keating, Cardiff University, United Kingdom

*CORRESPONDENCE Emma Hodson-Tole ☑ e.tole@mmu.ac.uk

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Children with developmental coordination disorder have less variable motor unit firing rate characteristics across contractions compared to typically developing children

Maaike Esselaar^{1,2}, Johnny V. V. Parr^{1,2}, Greg Wood^{1,2} and Emma Hodson-Tole^{2,3}*

¹Department of Sport and Exercise Sciences, Manchester Metropolitan University, Manchester, United Kingdom, ²Institute of Sport, Manchester Metropolitan University, Manchester, United Kingdom, ³Department of Life Sciences, Manchester Metropolitan University, Manchester, United Kingdom,

Introduction: Understanding the nuances of neuromuscular control is crucial in unravelling the complexities of developmental coordination disorder (DCD), which has been associated with differences in skeletal muscle activity, implying that children with DCD employ distinct strategies for muscle control. However, force generation and control are dependent on both recruitment of motor units and their firing rates and these fine details of motor function have yet to be studied in DCD. The purpose of this study was therefore to compare motor unit characteristics in a small muscle of the hand during low level, handgrip contractions in typically developing (TD) children and children with DCD.

Methods: Eighteen children (9 TD vs. 9 DCD) completed a series of manual handgrip contractions at $10\pm5\%$ of their maximum voluntary contraction. High density surface electromyography was used to record excitation of the first dorsal interosseus muscle. Recorded signals were subsequently decomposed into individual motor unit action potential trains. Motor unit characteristics (firing rate, inter-pulse interval, and action potential amplitude) were analysed for contractions that had a coefficient variation of <10%.

Results and Discussion: This study found few differences in average motor unit characteristics (number of motor units: TD 20.24 \pm 9.73, DCD 27.32 \pm 14.00; firing rate: TD 7.74 \pm 2.16 p.p.s., DCD 7.86 \pm 2.39 p.p.s.; inter-pulse interval: TD 199.72 \pm 84.24 ms, DCD 207.12 \pm 103 ms) when force steadiness was controlled for, despite the DCD group being significantly older (10.89 \pm 0.78 years) than the TD group (9.44 \pm 1.67 years). However, differences were found in the variability of motor unit firing statistics, with the children with DCD surprisingly showing less variability across contractions (standard deviation of coefficient of variation of inter-pulse interval: TD 0.38 \pm 0.12, DCD 0.28 \pm 0.11). This may suggest a

more fixed strategy to stabilise force between contractions used by children with DCD. However, as variability of motor unit firing has not been considered in previous studies of children further work is required to better understand the role of variability in motor unit firing during manual grasping tasks, in all children.

KEYWORDS

Dyspraxia, muscle activity, electromyography (EMG), isometric contractions, HD-EMG, neuromotor control, developmental disorders

1 Introduction

Developmental coordination disorder (DCD) affects 5–6% of children (American Psychiatric Association, 2013) and is associated with poor learning and performance of motor skills (Parr et al., 2020a) and appropriate force control. While the cause of DCD remains largely unknown, there is growing evidence that individuals with DCD display fundamental differences in their brain structure (Gill et al., 2022), brain activation patterns (Scott et al., 2021), and possibly how their brain communicates with the contracting muscles controlling force production (as observed in a single participant case study; Parr et al., 2022).

Understanding the nuances of neuromuscular control is crucial in unravelling the complexities of DCD. This disorder has been associated with fundamental differences in skeletal muscle activity, implying that children with DCD employ distinct strategies for muscle control (Fong et al., 2018). To investigate these strategies, researchers have used surface electromyogram (sEMG) recordings, which capture the interference pattern of detected motor unit action potentials. Analysing sEMG signals from children with DCD has revealed they tend to activate their muscles later, for longer durations and with more co-contraction across agonist/antagonist pairs in perturbation based postural balance control tasks (Williams and Castro, 1997; Raynor, 2001; Johnston et al., 2002; Williams, 2002; Geuze, 2003; Fong et al., 2013) and play-based activities like throwing and catching a ball (Fong et al., 2015) and uni- and bi-lateral aiming tasks (Huh et al., 1998).

Although these studies provide insight into the general patterns of muscle activity that may impact task performance, the measures used from the sEMG do not reveal details of the neuromuscular control used to produce the resulting joint force profiles (Martinez-Valdes et al., 2018). This will depend on the behaviours of the individual functional units within the active muscles. In skeletal muscle, these functional units are termed motor units and comprise the α -motoneuron, its axon and the group of innervated muscle fibres (Sherrington, 1925). An increase in force output from a muscle can be achieved by either recruiting more motor units or by increasing the firing rate of the already recruited motor units. sEMG amplitude measures do not reveal the number of recruited motor units nor their firing rates because the signal that is measured is the interference pattern of all the detected motor unit action potential shapes and firing rates (Martinez-Valdes et al., 2018). Traditionally, studying individual motor unit behaviours was only possible using invasive, intramuscular needle or fine-wire electrode techniques (Merletti and Farina, 2009). This has made it unfeasible to study motor unit behaviours in some populations, including children.

Advances in the availability of algorithms that can decompose sEMG signals into the individual motor unit action potential trains, do now, however, make it possible to extract such information from this signal, which can be recorded less invasively (De Luca et al., 2006; Holobar and Zazula, 2006; Pope et al., 2016). The currently available algorithms rely on availability of multiple (i.e., more than two) signals, recorded simultaneously from surface electrodes placed relatively close to each other on the same muscle (typically known as high density EMG, HD-EMG). The proximity of the recording sites provides multiple views of the same motor unit action potentials and, in various ways, the algorithms use the similarity and differences in signal information content to estimate the action potential shapes and firing instances of detected units that sum to produce the muscle behaviour. Decomposing HD-EMG signals, therefore provides a means of revealing some of the individual motor unit behaviours that contribute to a given motor task using a signal that can be easily recorded in children (and other previously inaccessible patient populations).

Decomposing EMG signals into constituent motor unit action potential trains has enabled differences in motor unit behaviours to be identified in adult patient groups where motor control is affected by pathology. For example, in stroke survivors, paretic muscles exhibit lower motor unit firing rates than non-paretic (Rosenfalck and Andreassen, 1980; Young and Mayer, 1982; Mottram et al., 2014) even for the same level of force production (Gemperline et al., 1995). This suggests that paretic muscles need to recruit more motor units to produce a given force, likely influencing the metabolic cost and muscle fatiguability. In people living with Parkinson's disease motor unit firing rate has been found to be the same as healthy controls, however significant differences in the variability of firing rate are found (Wilson et al., 2020). This suggests that in mild-moderate Parkinson's disease motor dysfunction is linked to variability in motor output. These two examples highlight how different conditions associated with movement impairment, are underpinned by differences in the motor system responses to a given task. To date however, there have been few studies of motor unit behaviours in children (Herda et al., 2019; Miller et al., 2019), and we could not find any that had compared motor unit behaviours in typically developing (TD) children and those with DCD. Given the fundamental connection between motor unit behaviour and the ability to meet the time varying force requirements of any movement task, this seems a significant gap in the current literature.

The purpose of this study was therefore to investigate motor unit characteristics in a small muscle of the hand during low level (10% maximum voluntary contraction) isometric handgrip contractions in TD children and children with DCD. Handgrip was selected as the study task due to its direct relevance to daily functional tasks (e.g., squeezing a toothpaste tube, opening food jars). The first dorsal interosseus (FDI) was selected as the muscle to be studied as it is easily palpated and offers an accessible location for required HD-EMG sensors to be secured, even in smaller children. In addition, there is a strong correlation between grip strength and finger strength ($r \ge 0.93$), with the fingers on the radial side of the hand contributing ~60% to overall grip strength (MacDermid et al., 2004). The index finger accounts for 25% of total grip strength (MacDermid et al., 2004) and hence FDI can be considered to contribute to the task studied. Assessing the firing rate and relative range of action potential sizes from recorded HD-EMG signals will provide insight into the neuromuscular control strategy associated with handgrip and might highlight factors contributing to control deficits in children with DCD.

2 Materials and methods

2.1 Participants

Thirty-eight participants, aged 7–12 years, were recruited for the study which had been approved by the local ethics committee in the Faculty of Science and Engineering, Manchester Metropolitan University (ethics number 41284). Children in the DCD group were recruited via social media, local support groups and via the Dyspraxia Foundation. The TD group was recruited via a local scout group, siblings of the children with DCD and from the family of student and staff members of the Manchester Metropolitan University.

Children in the DCD group were classified based on the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) criteria (American Psychiatric Association, 2013), whereby they exhibit substandard motor ability, relative to their chronological age. Prior to data collection, parents completed the Developmental Coordination Disorder Questionnaire (DCDQ, Wilson et al., 2009) to confirm that their child had significant movement difficulties that interfered with their child's daily lives. Co-occurrence with ADHD was also recorded using the Vanderbilt ADHD Diagnostic Parent Rating Scale (Wolraich et al., 2003). Finally, parents confirmed that their child did not suffer from any general medical condition known to affect sensorimotor function and had no diagnosed learning difficulties.

For the DCD group, children who scored 57 or below on the DCDQ (classified as suspected DCD) and below 18 on the ADHD rating scale (indication no ADHD) were invited to take part in the study. For the control group a score of 58 or higher on the DCDQ (indication no DCD) and below 18 on the ADHD (indication no ADHD) were invite to the lab. The DCDQ score was therefore used as an initial indication of DCD, which was subsequently confirmed with the Movement Assessment Battery for Children–2 (MABC-2) (see section "2.2.1 Assessment of motor impairment").

On visiting the laboratory, participants and guardians were shown a pictorial overview of the study and were given the chance to ask questions and discuss what would be required of them. After, participants provided written assent and the guardian completed a written consent form.

2.2 Data collection procedures

2.2.1 Assessment of motor impairment

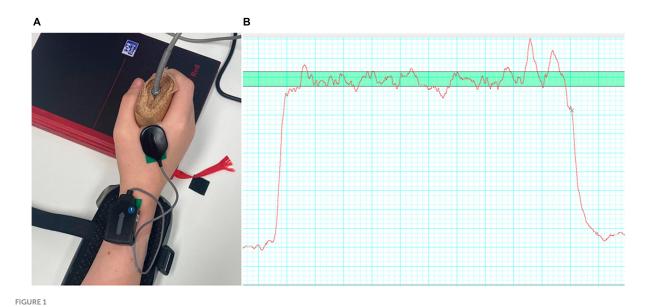
The MABC-2 is a test of motor impairment. The test assesses three domains: Manual Dexterity, Balance and Aiming and Catching with eight tasks in total. A total MABC-2 test score of up to 56 reflects a percentile score of 5% or less and denotes a significant movement difficulty, a total test score between 57 and 67 with a percentile score between the 5th and 15th suggest that the child is "at risk" of having a movement difficulty, any score above 67 or above the 15th percentile denotes no movement difficulty. In this study, children who scored at or below the 5th percentile for the total MABC-2 score were included in the DCD group while those who scored at or above the 20th percentile were allocated to the TD group.

2.2.2 Assessment of force production and motor unit activity

Participants sat at a table with 268.1 mm \times 476.6 mm size screen (Iiyama Co., Ltd, Iiyama, Japan) located 60 cm in front of them. The dominant hand was assumed to be the hand the child used to sign the assent form, and this was the hand with which all testing was completed. To collect surface EMG data, a four-pin surface array sensor (Delsys, Inc., Natick, MA, USA) was attached to the mid-belly region of the FDI muscle of the dominant hand. The diameter of each pin is 0.5 mm and they are placed at the corners of a 5 mm \times 5 mm square. Before the sensor was placed, the surface of the skin was prepared by shaving, applying, and removing tape to remove dead skin and dampening the skin with a paper towel. Data was recorded at 20 kHz using the EMGworks Acquisition (v. 4.8.0, Delsys, Inc., Natick, MA, USA).

The experimental protocol was based on a handgrip task, whereby participants were asked to repeatedly squeeze a hand-dynamometer (Parr et al., 2022, 2023). The dynamometer was attached to a PowerLab 4/25 T (AD Instruments, Bella Vista, NSW, Australia) that recorded the hand contraction force (in kilogrammes) via Labchart 8 software (ADinstruments, Sydney, NSW, Australia) at a sampling rate of 1000 Hz. Participants first completed three maximum voluntary contractions (MVC) with 1-min break in between each attempt. The force profile was presented to them on the screen using the Labchart interface. The peak force value achieved across the three recorded MVCs was used to set the target zone, 10% MVC \pm 5%, which was displayed as a 15 mm thick green band that extended the entire width of the screen (Figure 1).

During each trial participants were asked to squeeze the dynamometer, so that the force trace remained, as steady as possible and within the target zone. A single trial comprised 6×10 -slong contractions each separated by 10 s of rest. Feedback about the steadiness and accuracy of the force was provided during the practice trials and between trials. An audible tone, controlled through a custom PsychoPy (Peirce et al., 2019) script, signalled the start/end of each contraction. Eleven trials were completed in total (including one practice trial at the start) with 1 min rest between the trials for a total of 66 contractions per participant.



(A) Showing the EMG electrode placement on the hand of a participant; (B) and an example of a force trace (red line) with the target zone (green, horizontal bar) also shown.

2.3 Data analysis

2.3.1 Force data

Force data were extracted from the Labchart software, and each contraction was analysed for accuracy and steadiness of force production. Force accuracy was defined as the percentage of time participants were able to maintain their force output within the target zone. Force steadiness was defined as the coefficient of variance (CoV), calculated as the percentage of the force standard deviation to the mean force value for that contraction. Both force accuracy and steadiness were assessed from 1 to 10 s following the auditory "go" stimulus, as the first second was likely to contain dynamic fluctuations in force as participants ramped-up their force output. For each participant, we also calculated the standard deviation of force accuracy across all contractions (Accuracy SD), to express their contraction-to-contraction variability in task accuracy. These data were analysed using a bespoke Matlab (version R2021a) script.

Across recorded trials not every contraction attempt produced steady force outputs, with some showing consistently large fluctuations in magnitude. Therefore, to identify whether motor unit behaviours differed between the two groups it was important to ensure analysed data represented comparable force production behaviour. Thus, only contractions where the force CoV was 10% or below were included in the analysis. This threshold was defined prior to data analysis and based on previous work reported in Smits-Engelsman et al. (2003). The data of children who had at least 20 trials with CoV < 10% were included in the study.

2.3.2 Motor unit characterisation

Action potentials were extracted from recorded sEMG signals for each contraction from 1 to 10 s following the auditory "go" stimulus (as per force data) to provide individual motor unit action potential firing trains. This was achieved using the

precision decomposition (PD) III algorithm described by De Luca et al. (2006) and commercially available as NeuroMap software (v. 1.2.2, Delsys, Inc., Natick, MA, USA). The analysis processes involved pre-processing which includes filtering data at 20 Hz and a baseline correction, feature extracting, template matching, decomposing the EMG signal by matching identified MU templates onto the data and extracting them. The Neuromap Explorer software was then used to select, and export for further analysis, the motor unit information pertaining to units decomposed with \geq 85% accuracy within valid contractions (where force CoV < 10%). The accuracy measure provided by the software is based on the decompose-synthesise-decompose-compare approach described by De Luca and Contessa (2012).

From exported motor unit data, the following outcome measures were recorded for each participant: average number of motor units identified per contraction; their average firing rate (FR) (expressed as pulses per second; p.p.s.) and average inter-pulse-interval (IPI) (in milliseconds). As both average FR and average IPI are derived from the series of instantaneous measures generated by decomposition it is not possible to directly convert between the two, because such conversion is only possible between single data points (Johnson, 1994). The variability of IPI within each individual contraction was reported as the CoV of IPI (IPI-CoV, reported as the ratio of standard deviation: mean). By contrast, the standard deviation (SD) was calculated to determine the contraction-to-contraction variability of IPI (IPI-SD), IPI CoV (IPI-CoV-SD), and firing rate (FR-SD) across all contractions.

In addition, to investigate the diversity of characteristics in the recruited motor unit pool within participants, the average motor unit action potential amplitude (mV), average FR and average IPI across contractions were quantified for units within the 10th and 90th percentile for motor unit action potential amplitude. To account for differences in motor unit action potential amplitude stemming from other, extraneous, factors (e.g., different subcutaneous adipose thickness) the ratio of the average smallest

and largest action potential amplitudes (10th vs. 90th percentile) was calculated in each participant, essentially normalising data to facilitate comparison across groups.

2.3.3 Statistical analysis

For the within participant measures described above, a Shapiro-Wilk test confirmed the distribution of all dependent variables were normal (W=0.898-0.980, p=0.053-0.953). Independent t-tests were chosen to assess between group differences in age, MVC, force control (Force CoV in%, Accuracy, Accuracy SD), and measures describing motor unit characteristics (FR, IPI, IPI-CoV as ratio, FR-SD, IPI-SD, IPI-CoV-SD). Paired t-test was used to test for differences between firing of the biggest and smallest motor units within the two groups. Unless otherwise mentioned all tests were two tailed.

For all statistical tests, significant differences were considered to occur when p < 0.05. No adjustments for multiple comparisons were made, on the basis that this is an initial exploratory investigation of this topic and increasing the chance of a type II error could risk missing potentially useful findings (Rothman, 1990). The lack of previous motor unit data from TD children and those with DCD means it was not possible to complete *a priori* power calculations, and sample size was selected based on previous motor unit assessments in TD children (Miller et al., 2018, 2019). Therefore, here the effect size associated with each comparison (described above) was also calculated as Cohen's d, with \leq 0.1 considered small, 0.2–0.3 medium, 0.4–0.5 large effect size (Cohen, 1988). Average and standard deviation values for TD and DCD groups are reported, alongside the confidence interval (CI), in the following results.

3 Results

3.1 Participant characteristics

Of the 38 participants recruited 18 had a minimum 20 trials with CoV < 10% of these nine satisfied the criteria for DCD (MABC-2 score at or below the 5th percentile) (**Table 1**). The average age of the DCD group was 10.9 years old (range 10 to 12 years) and 9.4 years (range 7–12 years) for the TD group (**Table 1**). The TD group was significantly younger than the DCD group (t = -2.35, p = 0.032, 95% CI = -2.75 to -0.144, d = 1.27).

3.2 Force control characteristics

The TD group had an average MVC of 13.06 ± 4.46 kg and the DCD group had an average MVC of 15.35 ± 4.38 kg (Figure 2A). These values were not significantly different between the two groups (t = -1.213, p = 0.243, CI = -6.95 to 1.89, d = 0.6).

In total 453 contractions from the TD group and 500 contractions from the DCD group (both out of a total of 9 participants \times 6 contractions \times 11 trials = 594 contractions) met the criteria of a CoV in force of less than 10% and were included in the analysis. The TD group had an average of 50.33 \pm 11.92 valid contractions per person, while in the DCD group an average of 49.22 \pm 13.30 valid contractions per person. The number of

TABLE 1 Mean movement ABC-2, DCDQ and ADHD rating scale scores and age within participant group.

	Group			
Measure	DCD (N = 9)	TD (N = 9)		
MABC-2 Overall percentile score (percentile)	1.78 (1.39)	98.11 (1.27)		
MABC-2 aiming and catching (percentile)	6.56 (5.98)	91.56 (4.90)		
MABC-2 balance (percentile)	4.72 (7.79)	90.89 (6.48)		
MABC-2 manual dexterity (percentile)	7.72 (6.45)	93.11 (3.33)		
DCDQ score	24.8 (11.9)	65 (7.2)		
ADHD diagnostic parent rating scale score	17.4 (1.4)	15.3 (1.8)		
Age (Years)	10.89 (0.78)	9.44 (1.67)		
Sex split	7 male	6 male		

Standard deviation values are shown in parentheses. NM indicates not measured.

contractions included did not differ significantly between the two groups (t=0.19, p=0.85, 95% CI = -11.51 to 13.73, d=0.1). In addition, the CoV for force was $5.06\pm1.11\%$ for the TD group and $5.04\pm0.88\%$ for the DCD group. These values were not significantly different between the groups (t=0.048, p=0.962, 95% CI = -0.98 to 1.02, d=0.9) (Figure 2B).

When examining force accuracy, the TD group were accurate for $54 \pm 21\%$ of the contraction time while the DCD group were accurate for $50 \pm 8\%$ of the contraction time (**Figure 2C**). There was no statistically significant difference found between the two groups (t = 0.43, p = 0.67, CI = -0.13 to 0.20). However, it is noteworthy that the standard deviation of the force accuracy was significantly greater in one-sided testing (t = -1.84, p = 0.042, 95% CI = -0.05 to 0.003, d = 0.9), with the DCD group having a mean standard deviation of $15.3 \pm 1\%$ which was greater than that of the TD group, $12 \pm 3\%$ trial time (**Figure 2D**).

3.3 Motor unit characteristics and behaviours

The average number of motor units identified per contraction with accuracy \geq 85% was 20.24 \pm 9.73 in the TD group and 27.32 \pm 14.00 in the DCD group (**Figure 3A**). The average number of motor units per contraction did not differ between the groups (t = -1.25, p = 0.116, CI = -19.13 to 4.97, d = 0.6). The average ratio between the largest-smallest average motor unit action potential amplitude of the TD group was 7.40 ± 1.27 and 5.29 ± 0.98 for the DCD group. This difference was not significant (t = 0.19, p = 0.85, 95% CI = -11.51 to 13.73, d = 0.6) (**Figure 3B**).

The average FR exhibited notable similarity between the groups, with 7.74 \pm 2.16 p.p.s. for the TD group and 7.86 \pm 2.39 p.p.s. for the DCD group (**Figure 4A**). Again, there was no statistically significant difference between these values (t=-0.11, p=0.91, 95% CI = -2.40 to 2.16, d=0.1). The FR of the 10% largest and smallest motor units are shown in **Figure 4B**. In the TD group the mean FR was 2.22 \pm 1.75 p.p.s. and 10.82 \pm 3.77 p.p.s. for largest and smallest motor units, respectively. In the DCD group, the 10% largest units fired at 1.69 \pm 0.68 p.p.s., while the smallest units fired at 10.25 \pm 2.34 p.p.s. There was a significant

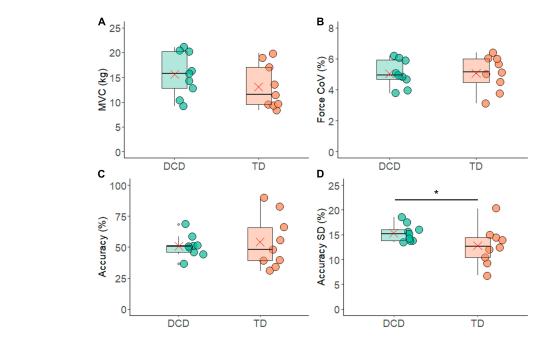


FIGURE 2
Box and whisker plots illustrating force measurements from each group. (A) Maximum voluntary contraction. (B) Co-efficient of variation (CoV) of force. (C) Force accuracy. (D) Standard deviation of force accuracy. In each figure mean and median group values are denoted by the \times symbol and horizontal line, respectively. The edges of the boxes represent the 1st and 3rd quartile values. Grey circle symbols indicate outlier values (identified in gg-plot as any values over 1.5 times the interquartile range over the 75th percentile or any values under 1.5 times the interquartile range under the 25th percentile), with the whisker edges representing the minimum and maximum values (excluding outliers). Significant differences ($\rho < 0.05$) between groups are denoted by the horizontal bar and asterisk (*).

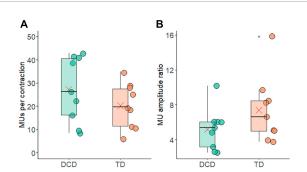


FIGURE 3

Box and whisker plots illustrating average motor unit characteristics from each group. (A) Number of motor units per contraction. (B) Ratio of the motor unit action potential amplitude from the largest and smallest motor units. In each figure mean and median group values are denoted by the x symbol and horizontal line, respectively. The edges of the boxes represent the 1st and 3rd quartile values. Grey circle symbols indicate outlier values (identified in gg-plot as any values over 1.5 times the interquartile range over the 75th percentile or any values under 1.5 times the interquartile range under the 25th percentile), with the whisker edges representing the minimum and maximum values (excluding outliers).

difference in the firing rate of the 10% smallest and largest motor units within the TD group (t = 8.07, p < 0.001, CI = 6.14 to 11.06, d = 2.7) and within the DCD group (t = 10.45, p < 0.001, CI = 6.55 to 10.56, d = 3.9). The between group difference for the FR of the 10% smallest motor units was not significant (t = 0.35, t = 0.731,

CI = -2.93 to 4.07, d = 0.2) nor was the FR of the 10% largest motor units (t = 0.74, p = 0.469, CI = -0.99 to 2.03, d = 0.4).

The average FR-SD of the smallest 10% was 1.86 ± 1.02 p.p.s. and 1.25 ± 0.47 p.p.s. for the TD and DCD group, respectively. For the largest 10% the average FR-SD was 1.05 ± 1.03 p.p.s. for the TD group and 1.01 ± 0.54 p.p.s. for the DCD group (**Figure 4C**). There was a significant difference in the FR-SD between the smallest and largest motor units in the TD group (t=2.61, p=0.031, CI=0.09 to 1.51, d=0.9), this difference was not significant in the DCD group (t=1.05, p=0.33, CI=-0.33 to 0.82, d=0.4).

The FR results were also reflected in the average IPI measures, where no meaningful differences emerged between the TD group (199.72 \pm 84.24 ms) and the DCD group (207.12 \pm 103 ms) (t = -0.167, p = 0.870, 95% CI = -101.79 to 86.9, d = 0.1)(Figure 5A). The average IPI-SD was slightly smaller in the DCD group (179.40 \pm 129.40 ms) compared to the TD group (182.31 \pm 79.56 ms), a difference that was not statistically significant (t = 0.59, p = 0.57, 95% CI = -58.21 to 102.66, d = 0.3) (Figure 5B). The IPI-CoV was also slightly smaller in the DCD group (0.79 \pm 0.16) compared to the TD group (0.94 \pm 0.25) (Figure 5C), and while there was a trend of significance during one-sided testing (t = 1.443, p = 0.080, 95% CI = -0.6 to 0.35, d = 0.7) it failed to reach significance. However, the IPI-CoV-SD was significantly lower in the DCD group (0.28 \pm 0.11) compared to the TD group (0.38 \pm 0.12) (**Figure 5D**) for one-sided testing (t = 1.93, p = 0.036, 95% CI = -0.01 to -0.22, d = 0.9).

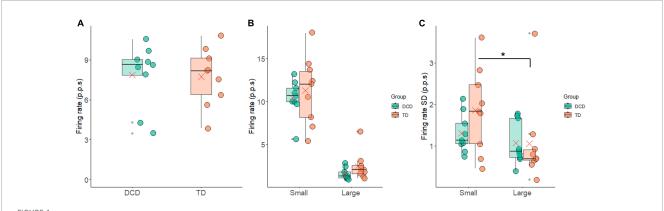
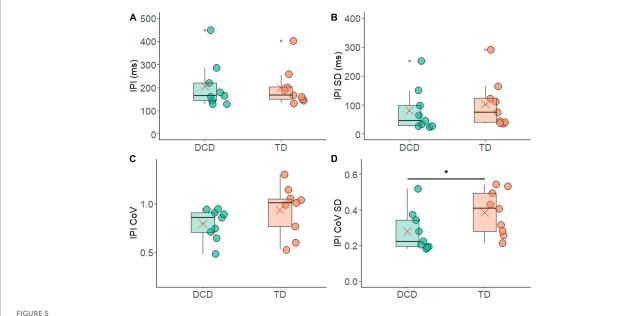


FIGURE 4
Box and whisker plots illustrating the firing rate measures from each group. (A) Average firing rate. (B) Average firing rate of the 10% largest and smallest motor units. (C) Standard deviation in firing rate of the 10% largest and smallest motor units. In each figure mean and median group values are denoted by the \times symbol and horizontal line, respectively. The edges of the boxes represent the 1st and 3rd quartile values. Grey circle symbols indicate outlier values (identified in gg-plot as any values over 1.5 times the interquartile range over the 75th percentile or any values under 1.5 times the interquartile range under the 25th percentile), with the whisker edges representing the minimum and maximum values (excluding outliers). Significant differences (p < 0.05) between groups are denoted by the horizontal bar and asterisk (*).



Box and whisker plots illustrating the inter pulse interval measures from each group. (A) Average inter-pulse interval (IPI). (B) Standard deviation of the average inter-pulse interval. (C) Co-efficient of variance of the inter-pulse interval. (D) Standard deviation of co-efficient of variance of the inter-pulse interval. In each figure mean and median group values are denoted by the \times symbol and horizontal line, respectively. The edges of the boxes represent the 1st and 3rd quartile values. Grey circle symbols indicate outlier values (identified in gg-plot as any values over 1.5 times the interquartile range over the 75th percentile or any values under 1.5 times the interquartile range under the 25th percentile), with the whisker edges representing the minimum and maximum values (excluding outliers). Significant differences (p < 0.05) between groups are denoted by the horizontal bar and asterisk (*).

4 Discussion

This study was the first to examine the number of detected motor units and their characteristics (ratio of action potential sizes, FR and IPI) in the FDI during low level (10% MVC) handgrip isometric contractions in TD children and children with DCD. This study found few differences in most of the motor unit characteristics when force steadiness was controlled for.

There were no differences in MVC magnitude between the DCD and TD groups. The selection of contractions in which the

force produced was maintained within the target window with the CoV < 10% also meant there were no significant differences in the force steadiness profile between groups. This indicates that all children whose data were included in the analysis were able to complete the task, and to do so with similar proficiency, given that the average number of contractions that met the analysis inclusion criteria was $\sim\!50/66$ in both groups. However, the percentage of time participants were able to maintain the target force output (force accuracy) was quite low in both groups (**Figure 2C**), indicating the task did provide a degree of challenge to both groups.

Esselaar et al. 10.3389/fnhum.2023.1294931

Observation of the participants leads us to believe this reflects the challenge of moving the force into the target window at the onset of the contraction, rather than maintaining the force once the target was achieved. It should however be noted that the TD group was significantly younger than the DCD group, which needs to be considered when interpreting findings.

Previous research has shown that as children mature, their skill at controlling force increases. For example, Smits-Engelsman et al. (2003) showed that children, aged 5–12 years, were able to perform an isometric finger press task with minimal accuracy deviation (2.4%) and no association between the accuracy deviation and age. However, they found that the force variability (measured as CoV) decreased significantly with age. Here we found no differences in the force variability (measured as CoV) nor in the accuracy between the DCD group and the TD group (Figure 2) when controlling for force stability as defined by CoV < 10%, despite the DCD group being significantly older than the TD group (\sim 1.5 years older). The matching of force steadiness and accuracy performance between the DCD and TD groups could suggest a delay in the development of neuromotor strategies to stabilise force production in children with DCD. This is in agreement with earlier work by Smits-Engelsman et al. (2008), who also found that the standard deviation and the CoV in index finger press isometric force contractions were similar between younger TD children (7-9 year-olds) and older children (11 year-olds) with DCD.

Greater variability in force output has functional implications, for example linking to serious handwriting problems (Smits-Engelsman et al., 2001). This variability has been attributed to a high level of noise in the neuromotor system, making it more difficult to complete perception-action calculations and hence successfully complete fine motor tasks (Smits-Engelsman et al., 2008). Contamination of the neuromotor signal can occur from sources both internal and external to the nervous system (van Galen et al., 1993; van Galen and de Jong, 1995). Within the peripheral nervous system, neuromotor noise can be introduced through recruitment and/or rate coding of motor units. Here, we show that (for the FDI muscle during a handgrip task) the number of motor units detected, and their size, firing rate and inter-pulse interval did not differ between the DCD and TD group. As such, recruitment of units and the average firing characteristics (i.e., rate coding) of the recruited motor unit pool do not seem to be the source of any differences in neuromotor noise that might exist between TD children and those with DCD.

However, differences were found in the variability of motor unit firing statistics, i.e., variability in the rate coding. In contrast to what may be predicted based on the neuromotor noise theory, less variability was found in the motor unit firing statistics between contractions. Specifically, the variability in IPI-CoV between contractions, represented by IPI-CoV-SD, was significantly smaller in the DCD group compared to the TD group (Figure 5). In addition, the TD group exhibited a significant difference in the standard deviation of firing rate between the largest and smallest motor units (Figure 4). This difference was not however found in the DCD group, again indicating smaller contractioncontraction variability in this group. The children with DCD therefore repeatedly produced the same force output patterns with less variance in the firing rate statistics in the recruited motor unit pool across contractions. The implication of this smaller variance is unclear, however it may reflect patterns of motor unit behaviour were more fixed (e.g., fewer motor unit combinations detected) across contractions. If so, this could increase fatiguability if the task were to be repeated over very extended periods of time.

It is important to note that the magnitude of differences in variance found here are quite small (although the effect size was large in some cases) and come from a relatively small sample size. However, when taken together with the lack of consideration of variability in previous studies of motor unit firing in children (Miller et al., 2018, 2019; Herda et al., 2019), it is suggested that further studies are required to better understand the role of variability in motor unit firing during manual grasping tasks, in both TD children and those living with DCD.

Indeed, wider study of the role of motor system variability within the context of the greater movement variability that is a hallmark of DCD (Geuze and Kalverboer, 1987; Williams et al., 1992; Volman and Geuze, 1998; Bo et al., 2008; Mackenzie et al., 2008; Smits-Engelsman et al., 2008; Roche et al., 2016; Golenia et al., 2018; Parr et al., 2020a,b) seems important. The influence of motor unit recruitment on the relationship between within and between movement variability should also be considered. This is considering our finding that while the children with DCD had the same accuracy level, there was greater variation in their contraction-to-contraction accuracy (standard deviation of the accuracy, Figure 2). This suggests, that while their motor recruitment strategies were on average as successful as those of the TD children, they may constrain the adaptability that enables fine, contraction-to-contraction adjustments. Such exploration would benefit from consideration of the temporal characteristics of variability which have been applied in the study of gait dynamics (Hausdorff, 2007) and dynamics of muscle activity (Wakeling and Hodson-Tole, 2018; Ferrari et al., 2020), to provide further insight into the moment-to-moment adjustments in motor output that facilitate smooth, coordinated movement patterns.

This study is not without its limitations. This is the first time that motor unit behaviour has been assessed in children with DCD via high density surface EMG. Therefore, it was impossible to calculate an a priori power and sample size calculation. The relatively small sample size could therefore have contributed to some of the effect sizes being small, while others are strong to very strong. Because there were no differences in the number of trials that were included and the number of detected motor units between the two groups, it is suggested that the HD-EMG sensor worked equally well in both TD and DCD children. This provides confidence that the similarities and differences found reflect the underlying neurophysiological functioning of studied children, and not factors related to our experimental set up. However, the motor unit characteristics reported here are only from one, small hand muscle. The handgrip forces recorded reflect the sum of several muscles in the hand and forearm. As we record only from the FDI caution should be taken when interpreting these results.

In conclusion, this study found that when controlling for CoV in an isometric handgrip task, children with DCD performed as well as their TD counterparts. The underlying muscular control only differed in the contraction-contraction variance of the motor unit firing statistics. Therefore, the underlying motor unit recruitment patterns of TD children and children with DCD do not seem to differ. In contrast, features of motor unit rate coding across contractions did. This difference may indicate that the children with DCD proficiently achieved the task by employing

a different strategy in relation to the neural drive received by the recruited motor unit pool. However further work is required to confirm this finding, and to identify whether it is a general feature of neuromotor behaviour across other muscles of the hand and arm.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author.

Ethics statement

The studies involving humans were approved by the local Ethics Committee in the Faculty of Science and Engineering, Manchester Metropolitan University (Ethics number 41284). The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin.

Author contributions

JP: Conceptualization, Funding acquisition, Methodology, Software, Visualization, Writing – review and editing. GW: Conceptualization, Funding acquisition, Project administration, Writing – review and editing. EH-T: Conceptualization, Funding acquisition, Methodology, Writing – original draft, Writing – review and editing. ME: Conceptualization and development, Design of methodology, Review, Revision and approval of manuscript, Data curation, Formal analysis, Writing original draft, Software and Visualisation.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

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EDITED BY

Mariagiovanna Cantone,

Gaspare Rodolico Hospital, Italy

REVIEWED BY Reza Abdollahipour, Palacký University Olomouc, Czechia Yao-Chuen Li, China Medical University, Taiwan

*CORRESPONDENCE
Catherine Purcell

☑ PurcellC2@Cardiff.ac.uk

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Understanding factors that influence physical activity behavior in people with developmental coordination disorder (DCD): a mixed-methods convergent integrated systematic review

Catherine Purcell^{1*}, Nadja Schott², Victoria Rapos^{3,4}, Jill G. Zwicker^{4,5} and Kate Wilmut⁶

¹School of Healthcare Sciences, Cardiff University, Cardiff, United Kingdom, ²Institute of Sport and Movement Science, University of Stuttgart, Stuttgart, Germany, ³Rehabilitation Sciences, The University of British Columbia, Vancouver, BC, Canada, ⁴Brain, Behavior, and Development, British Columbia Children's Hospital Research Institute, Vancouver, BC, Canada, ⁵Department of Occupational Science and Occupational Therapy, The University of British Columbia, Vancouver, BC, Canada, ⁶Centre for Psychological Research, Oxford Brookes University, Oxford, United Kingdom

This systematic review synthesizes the literature on physical activity amongst people with DCD using the COM-B framework. The review questions were: (1) what is the Capability (C), Opportunity (O) and Motivation (M) for physical activity and (2) what does physical activity behavior (B) look like? A mixedmethods systematic review was conducted by searching eight databases (PubMed, APA PsycINFO, EMBASE, Scopus, Child Development and Adolescent Studies, Cochrane Library, Web of Science, CINAHL) up to July 2023. Data were extracted, thematically analyzed, and mapped to the COM-B model. The quality of studies was assessed with the Joanna Briggs Institute (JBI) critical appraisal tool. The protocol was registered with PROSPERO (CRD42022319127). Forty-three papers, 42 of which related to children, were included. Fifteen aligned with physical activity behavior, nine with physical capability, thirteen with psychological capability, one with social opportunity, one with physical opportunity, one with reflective motivation and three with automatic motivation. Pre-school-aged children with DCD engage in comparable levels of physical activity behavior, but differences emerge from 6 years of age. Characteristics of DCD result in reduced physical capability and less varied participation in physical activity. This impacts psychological capability, whereby lower self-perceptions

result in a negative feedback loop and reduce the motivation to participate. Barriers relating to social opportunities may result in poor reflective and automatic motivation, although there is evidence that interventions can enhance enjoyment in the short term.

KEYWORDS

developmental coordination disorder, motor skills disorder, physical activity, COM-B, behavior change

1 Introduction

Physical inactivity is one of the leading risk factors for premature mortality worldwide (World Health Organization [WHO], 2022), accounting for 5.3 million deaths annually (Lee et al., 2012). In the UK alone, physical inactivity contributes to one in six deaths and is estimated to cost £7.4 billion annually (Public Health England, 2016). Physical inactivity increases the societal and economic burden of mental and physical ill-health; however, despite the clear evidence of the health benefits of being physically active, over a quarter of the world's adult population is insufficiently physically active, and 81% of 11–17 year-olds were insufficiently physically active in 2022 (World Health Organization [WHO], 2022). Promoting physical activity is, therefore, a global public health priority (World Health Organization [WHO], 2022).

However, some families experience significant inequalities in opportunities for physical activity, such as families of children living with disabilities, who often experience greater environmental barriers (World Health Organization [WHO], 2020). One such group is children and adults with developmental coordination disorder (DCD). DCD is a neurodevelopmental disorder affecting 5–15% of school-aged children (Hamilton, 2002) that significantly impacts a child's ability to learn motor skills and perform everyday activities, including getting dressed, tying shoelaces, using cutlery, handwriting, playing games or sports, or riding a bicycle (Zwicker et al., 2012). Ultimately, these motor deficits harm academic performance, vocational choices and leisure pursuits (Zwicker et al., 2012). Secondary consequences of DCD include low self-esteem, depression, anxiety, loneliness, problems with peers and withdrawal from participating in physical and social activities (Zwicker et al., 2018; Izadi-Najafabadi et al., 2019; Harris et al., 2021). Furthermore, the motor and psychosocial difficulties associated with DCD profoundly impact quality of life (Zwicker et al., 2018) and persist in adulthood (Harris et al., 2021).

Previous research has explored physical inactivity in children and adults with DCD and identified that people with DCD are less physically fit (Schott et al., 2007), less physically active (Cairney et al., 2012), have lower perceived athletic competence and tend to avoid participation in physical activity (Rivilis et al., 2011). These findings are supported by a recent systematic review that explored differences in physical activity levels and the impact of these differences (Mercê et al., 2023). The authors concluded that the 16 included studies identified lower levels of moderate and vigorous physical activity amongst children with DCD, with implications across physical and psychological domains reported (Mercê et al., 2023). A recent scoping review also examined psychosocial factors

related to physical activity among children with DCD based on social cognitive theory, self-determination theory and the theory of planned behavior (Kwan et al., 2022). The authors concluded from the 14 papers that physical literacy-based interventions targeting perceived motor competence and motivation might effectively promote physical activity in children with DCD (Kwan et al., 2022).

However, despite these findings and increased intervention efforts to increase physical activity amongst people with DCD, most lack evidence-based behavior change theories. Behavior science approaches are based on the idea that successful behavior change depends first and foremost on a clear definition of the problem: who needs to change what behavior, in what way and what is required to do so? Once the behavior is clearly defined, in the present case that people with DCD avoid physical activity or engage in physical activity only to a limited extent, behavior change interventions can be developed.

Many behavior science models assume that successful behavior change interventions must consider three essential aspects: motivation, competence and situation. The COM-B model (Michie et al., 2011) was developed following a comprehensive review and consultation of 19 existing frameworks of behavior change interventions, none of which incorporated a full range of intervention functions or policies, therefore the COM-B provides a comprehensive and coherent link to a model of behavior. The COM-B model (Michie et al., 2011) posits that Behavior (B) occurs as a result of a bi-directional interaction between three components: Capability (C), Opportunity (O) and Motivation (M) and, as such, can contribute insights into physical activity behavior. This model explains that to perform a particular behavior; one must feel they are physically and psychologically able to do so (C), have the physical and social opportunity (O) and want or need to carry out the behavior more than other competing behaviors (M).

While some of the previous literature and systematic reviews have explored individual components of the COM-B, this is the first systematic review that brings the COM-B components together to better understand the physical activity behavior of people with DCD. This is necessary to develop future behavior change interventions, using the Behavior Change Wheel, that aim to increase physical activity. Without a critical overview of the literature relating to capability, opportunity, and motivation for physical activity amongst people with DCD, there is a risk that interventions focus on components that do not result in behavior change. Additionally, a comprehensive overview of the literature enables future research to focus on any gaps identified, strengthening the evidence for future interventions. Therefore, this systematic review addressed the following questions: what is

this group's capability, opportunity and motivation for physical activity? and (Lee et al., 2012) what does physical activity behavior look like amongst people with DCD?

2 Methods

This systematic review was informed by the Joanna Briggs Institute (JBI) methodology for conducting mixed-method systematic reviews (Stern et al., 2020). The review protocol was registered on PROSPERO in March 2022 (reference number: CRD42022319127).

2.1 Eligibility criteria

We developed comprehensive inclusion and exclusion criteria to judge the eligibility of potential publications involving people with DCD and physical activity outcomes for inclusion in this systematic review. The criteria were developed *a priori* based on the results of a preliminary scoping search in CINAHL and were piloted on two papers identified through the initial search. CP and one other reviewer, KW, independently applied the eligibility criteria. The reviewers discussed potential changes; however, the eligibility criteria did not need to be updated prior to application. The full eligibility criteria are detailed below.

2.1.1 Study design

Qualitative, quantitative, and mixed-method studies written in any language and peer-reviewed were included. This review did not include systematic reviews, meta-analyses, study and review protocols, commentaries, editorials, gray literature, conference posters or abstracts, although reference lists were used to enhance search results. Only English language articles were identified, so translation was unnecessary.

2.1.2 Participants

We included studies concerning children (under 18 years) or adults (18+ years) who met the Diagnostic Statistical Manual of Mental Disorders (DSM) or International Classification of Diseases (ICD) criteria for DCD (or at least 2 out of 4 criteria). Studies that reported co-occurring diagnoses/characteristics were included if the article's primary focus was DCD. Articles were excluded if they did not include a standardized motor assessment, or where another condition or visual impairment could explain the motor difficulties, or where motor difficulties were a consequence of a lack of opportunity.

2.1.3 Intervention

The focus of this systematic review was not on interventions; however, any studies that included interventions, even when the primary outcomes were not relevant to this review, were included if relevant baseline data were reported.

2.1.4 Comparators

For interventions including randomized controlled trials or pre-post intervention studies of any duration, articles were included if a comparator group did not receive a physical activity intervention or if the comparator group was a typically developing (TD) group.

2.1.5 Outcomes

Articles that reported outcomes in line with physical activity Behavior (B), Capability (C), Opportunity (O), and Motivation (M) were included. In addition, we considered other outcomes if they were measured alongside a COM-B component. A non-exhaustive list of examples of eligible outcomes are presented in **Table 1**.

2.2 Search strategy

We conducted a preliminary search of CINAHL on the 16th of March 2022 to scope the literature relevant to the review questions. The scoping exercise helped ensure that there were no current or ongoing reviews on the topic of interest, refine the aims and eligibility criteria for this systematic review, estimate the amount of published work available, and, therefore, the resources needed to complete this systematic review. Relevant articles identified from the scoping search of CINAHL were also used to develop a full search strategy; keywords in the titles and abstracts and the index terms used to describe the articles were organized into search strings. The search period was not limited. We used the following keywords and MeSH (medical subject heading) terms: developmental coordination disorder; motor skills disorders; DCD; probable DCD; significant motor difficulties; motor development; dyspraxia; motor competence; physical activity; sedentary behavior; exercise; physical performance; sport; aerobic exercise; fitness; motor activities; anaerobic exercise and participation.

2.3 Data sources

We searched the following electronic databases for peer-reviewed articles between the 6th May 2022 and the 27th May 2022: PubMed; APA PsycINFO; EMBASE; Scopus; Child Development and Adolescent Studies; Cochrane Library; Web of Science; CINAHL via EBSCO and ERIC. A final search was conducted on the 10th July 2023 to ensure any articles published after the 27th May were captured; no additional articles were identified for inclusion. No date restrictions were applied to the searches; all publications up to the date of the searches were considered.

2.4 Article screening

References were imported into Rayyan (Ouzzani et al., 2016), and duplicates were removed. Initially, the titles and abstracts of articles were screened independently by CP and KW against the eligibility criteria, and any conflicts that arose were resolved through discussion and, where necessary, by the third reviewer, NS. The screening process was reported in the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA) 2020 flow diagram (Page et al., 2021) (see Figure 1).

2.5 Critical appraisal

We assessed the methodological quality of included articles using the established JBI critical appraisal tools for the following study designs: Randomized Controlled Trials (RCTs) and quasi-experimental studies (Tufanaru et al., 2017); analytical cross-sectional studies, case reports and cohort studies (Moola et al., 2015) and qualitative research (Lockwood et al., 2015).

We adopted the method and classification outlined by Edwards et al. (2016) to judge quality, and included articles were assessed against the pre-determined criteria. Quantitative and qualitative components of mixed-method studies were appraised separately using the appropriate critical appraisal instruments. Each paper received an overall score based on the number of criteria met (13 for RCTs, 10 for qualitative and cohort studies, 9 for quasi-experimental studies and 8 for analytical cross-sectional studies and case reports). A point was deducted from the total available score if a criterion was considered not applicable to a particular article. A percentage score allowed the normalization of scores; 0–40% were considered low quality, 40–70% moderate quality and 70–100% high quality.

Each article was assessed independently by one of the authors, and then all scores were checked by a paired reviewer (e.g., CP reviewed KW).

2.6 Data extraction

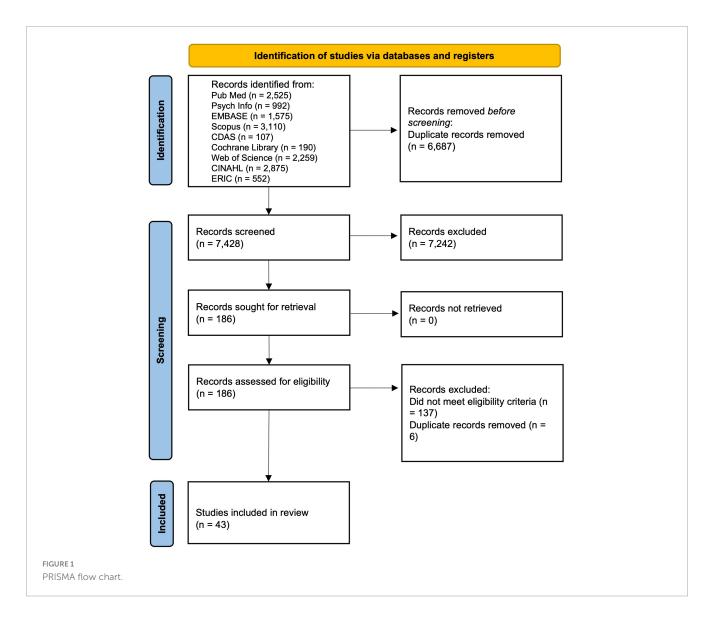
A piloted template (Supplementary material, online supporting information) was used to extract the study design, sample characteristics, diagnostic criteria, methodology and summary outcomes. Data were independently extracted by a single author and checked by a paired reviewer. The review team conducted consensus checks and resolved discrepancies through discussion. Studies were grouped into one of seven categories: physical capability; psychological capability (C); physical opportunity; social opportunity (O); reflective motivation; automatic motivation (M) and behavior (B).

2.7 Data transformation

One reviewer (CP) employed a convergent integrated approach to synthesize the data: this involved narrative interpretation of the quantitative results from experimental studies (including the quantitative component of mixed-method studies) in a way that answered the review questions by a repeated detailed examination. Specifically, quantitative and qualitative findings were initially synthesized separately; quantitative data was converted into "qualitized data" through transformation into narrative interpretation, followed by integration of both sets of findings. This

TABLE 1 Examples of facilitators to physical activity behavior and common measures framed within the COM-B.

COM-B components	Possible facilitators of physical activity behavior	Possible measures
Capability: individual's physical an	d psychological capacity to engage in the l	behavior
Psychological: the capacity to engage in necessary thought processes	Need knowledge of suitable local activity opportunities Need to know about easy and manageable activities that are safe	Children's self-perception and adequacy in predilection for physical activity
Physical: the capacity to engage in the necessary physical processes	Motor coordination problems Reduced fitness	Adolescent Motor Competence Questionnaire, Developmental Coordination Disorder Questionnaire, Movement Assessment Battery for Children/Movement Assessment Battery for Children – 2nd ed, Test of Gross Motor Development, Bruininks Oseretsky Test of Motor Proficiency/Bruininks Oseretsky Test – 2nd ed, Canadian Agility and Movement Skill Assessment; Physical Activity Questionnaire
Opportunity: all factors lying outs	ide the individual that makes the performar	nce of the behavior possible or prompt it
Social: cultural milieu that dictates the way we think about things	Family and peers provide encouragement to be active. Want to be active with other people with DCD who understand their current situations. Opportunity to be part of a group, to create accountability and provide encouragement.	Social support for exercise behavior scale
Physical: physical opportunity provided by the environment	Need accessible and pleasant walking routes or groups —good pavements or footpaths, safe and greenspace. Need appropriate and accessible recreational spaces and differentiated programs	Neighborhood environment scale Presence of recreational facilities index Participation and environment measure for children and youth
Motivation: all brain processes tha	t energize and direct behavior	
Automatic: emotions and impulses arising from associative learning and/or innate dispositions	Social interaction with other people is a motivation to be active. Want to take part in physical activity that is fun and enjoyable	Physical literacy in children questionnaire Physical exercise self-efficacy scale Exercise self-identity scale Perceived behavioral control. Positive and negative affect schedule short form Children's assessment of participation and enjoyment/preferences for activities of children
Reflective: evaluations and plans	Understand the physical and mental health benefits of physical activity	Canadian assessment of physical literacy Physical literacy in children questionnaire



approach provides greater insights and preserves the integrity of both sets of findings (Stern et al., 2020).

2.8 Data synthesis and integration

We narratively integrated quantitative and qualitative data at the interpretation level using the COM-B components to identify the relationship between and within quantitative and qualitative findings. The narrative summary was scrutinized by the review team for accuracy.

3 Results

3.1 Characteristics of included studies

We screened 7,428 titles and abstracts and assessed 186 full texts for eligibility. Forty-three papers met the eligibility criteria and were included in the review (see **Figure 1**).

The characteristics of studies included in this systematic review are presented in **Table 2**. All included studies considered children or adolescents, apart from one (Tan et al., 2022), which followed up with participants at the age of 25 years.

3.1.1 Design

Studies included six randomized controlled trials (Howie et al., 2016, 2017; Yu et al., 2016a; Bonney et al., 2017a; Noordstar et al., 2017; Sit et al., 2019), four quasi-experimental studies (Meek and Sugden, 1997; Poulsen et al., 2007; Cairney et al., 2010; Green et al., 2011), 25 cross-sectional studies (Cairney et al., 2005a,b, 2007, 2019; Poulsen et al., 2008, 2011a; Baerg et al., 2011; Fong et al., 2011, 2018; Silman et al., 2011; Engel-Yeger et al., 2012, 2015; Beutum et al., 2013; Kwan et al., 2013; Batey et al., 2014; Noordstar et al., 2014; Cermak et al., 2015; Yu et al., 2016b, 2021; King-Dowling et al., 2018, 2019; Wright et al., 2019; Brown et al., 2021; James et al., 2021; Li et al., 2021), four longitudinal studies (Cairney et al., 2006, 2017; Joshi et al., 2015; Tan et al., 2022), two qualitative studies (Barnett et al., 2013; Zimmer et al., 2020), one case series study (Kane and Bell, 2009) and one mixed-method study (Adams et al., 2018).

TABLE 2 Characteristics of the included studies, including author [reference], study design, percentage quality score, sample, diagnostic criteria and summary of outcome.

Author	Study design	% Quality score	Sample	DMS IV ar	nd DSM 5*	DSI	M IV	DSN	1 5	Summary outcome
				Criterion A	Criterion B	Criterion C	Criterion D	Criterion C	Criterion D	
				Poor acquisition and execution of motor skills	The deficits interfere with activities of daily living	Not due to general medical condition and does not meet criteria for PDD	Motor difficulties are in excess of any intellectual deficits	Onset of symptoms is in the early developmental period	Not better explained by intellectual, visual or neurological condition	
Behavior										
Cairney et al., 2006	Long	100%	44 DCD 537 TD 9–14 years	BOTMP-SF		Children with known learning disabilities and physical health problems were excluded				Children with DCD participated less in structured and free play activities, but this did not change with age.
Poulsen et al., 2008**	CS	75%	60 boys with DCD 113 boys without DCD 10–13 years	MABC-2 ≤15th	Parent report	Excluded if had a diagnosed neurological/ psychiatric condition	Excluded if IQ < 70 on SIT-R3			LPA and MVPA both sig lower in DCD vs peers. Boys with DCD spent less time in structured and unstructured PA compared to peers.
Cairney et al., 2010 Same sample as PHAST study	QE	75%	111 pDCD 1972 TD Start age for sample: 9 years 11 months	BOTMP and MABC <6th			Kaufman brief IQ test			Divergence in free-play activity occurs for females with probable DCD, but not for males.
Green et al., 2011	QE	75%	193 pDCD 4138 TD 7 years and 12 years	ALSAPC coordination test (one item per scale from the MABC), <15th	23 item ADL scale completed by parent	Excluded those with known visual/neurological condition	Excluded those scoring below 70 on short form of WISC-III			Boys with p-DCD were less physically active than boys without DCD (no group difference in girls).
Baerg et al., 2011 Same sample as PHAST study	CS	88%	32 DCD (12.8 years) 30 DCD/ADHD (12.9 years) 48 TD (12.7 years)	MABC-2, <15th			Used the KBIT-2 to determine typical IQ			Girls: DCD/ADHD > control for daily step count. No other differences in step count. Boys: no differences in daily step count between groups.
Poulsen et al., 2011a	CS	75%	60 DCD boys 113 boys without DCD 10–12 years 11 months	MABC, <15th	Parent identified difficulties with daily living skills	Excluded if had a previously diagnosed neurological or medical disorder	Excluded if IQ < 70 on SIT-R3			Boys with DCD reported fewer (MVPA) activities, and majority of PA in DCD group were completed individually or in the home environment.
Beutum et al., 2013	CS	75%	9 pDCD (8 years, 10 months) 9 TD (8 years, 11 months)	MABC-2, <15th	Parents reported on activities of daily living		Teachers reported no cognitive difficulties			Children with DCD: participated in less MVPA, had higher BMIs, decreased strength and cardiovascular fitness.

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Author	Study design	% Quality score	Sample	DMS IV ar	nd DSM 5*	DSI	M IV	DSM	15	Summary outcome
				Criterion A	Criterion B	Criterion C	Criterion D	Criterion C	Criterion D	
				Poor acquisition and execution of motor skills	The deficits interfere with activities of daily living	Not due to general medical condition and does not meet criteria for PDD	Motor difficulties are in excess of any intellectual deficits	Onset of symptoms is in the early developmental period	Not better explained by intellectual, visual or neurological condition	
Cermak et al., 2015**	CS	75%	US: 31 DCD (9.3 years) and 44 TD (9.5 years) Israel: 22 DCD (8.7 years) and 21 TD (girls [9.0 years])	MABC-2, ≤15th	Parent's report challenges in ADL				Parental report regarding medical history	In both Israel and the US, children with DCD demonstrated reduced physical activity, increased sedentary behavior, poorer fitness and increased overweight vs typical children.
Howie et al., 2016	RCT	62%	21 DCD or rDCD 10–12 years	MABC-2	DCD-Q				Parents reported no known behavioral or neurological disorders	No significant differences in time spent in sedentary, light, moderate or vigorous physical activity between the intervention and control periods.
Howie et al., 2017	RCT	62%	Sample as Howie et al. (2016)					Sample as Howie et al. (2016)		Participants described being more confident, stronger, having improved fitness and an increased willingness to participate in sports and physical activity.
King- Dowling et al., 2019 Same sample as CATCH study	CS	75%	111 pDCD (4.9 years) 177 rDCD (4.9 years) 301 TD (5 years)	MABC-2, <5th for pDCD and 6th–16th for rDCD	Determined via interview with parents			Young children, so in early developmental period	Excluded if had a diagnosis of a medical condition affecting motor control	No group differences for amounts of daily activity. Children with p-DCD accumulate MVPA in shorter bouts.
Cairney et al., 2019 Same sample as CATCH study	CS	100%	287 rDCD 301 TD 4–5 years of age	MABC-2, <16th	Determined via parent semi-structured interview				Excluded those with any known neurological or physical condition which might affect motor control	No group differences for BMI percentile or physical activity. Children in the rDCD group had significantly lower aerobic and musculoskeletal fitness, and larger waist circumference.
James et al., 2021 Same sample as CATACH study	CS	75%	288 at rDCD (4.9 years) 301 TD (5.0 years)	MABC-2, <16th				Young children, so in early developmental period	Parents reported medical history	When adjusting symptoms of ADHD, children at risk of DCD are less active than their TD peers.

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Summary outcome

Study

Sample

Author

DSM IV

DSM 5

DMS IV and DSM 5*

TABLE 2 (Continued)

Author	Study design	% Quality score	Sample	DMS IV ar	nd DSM 5*	DSI	M IV	DSM	15	Summary outcome
				Criterion A	Criterion B	Criterion C	Criterion D	Criterion C	Criterion D	
				Poor acquisition and execution of motor skills	The deficits interfere with activities of daily living	Not due to general medical condition and does not meet criteria for PDD	Motor difficulties are in excess of any intellectual deficits	Onset of symptoms is in the early developmental period	Not better explained by intellectual, visual or neurological condition	
Yu et al., 2016b	CS	100%	43 DCD 87 TD 7–10 years	MABC-2	Teacher confirmed motor difficulty					Children with DCD reported poorer physical self-concept on health, coordination, and sporting ability. Girls with DCD had a lower level of PA compared to boys with DCD or TD children.
Bonney et al., 2017a	RCT	85%	43 DCD divided into two groups Females aged 13–16 years.	MABC-2, <16th BOTMP 2nd Edition	Self-report questionnaire on perceived motor competence ADL questionnaire completed in week 1			No diagnosis of a significant medical condition known to affect motor performance was noticed nor reported.	Recruited from mainstream high school assumed no intellectual or cognitive impairment.	Both the Task-oriented Functional Training (TFT) and Wii training groups showed significant improvement in muscular strength, motor proficiency, running and agility, predilection for physical activity and generalized self-efficacy.
Cairney et al., 2017 Same sample as PHAST study	Long	100%	97 pDCD 1857 TD Starting age 9–10 years	Bruininks–Oseretsky Test of Motor Proficiency 1st Edition (BOT-SF).					Excluded children with known physical or learning difficulties	Cardiorespiratory fitness was lower in DCD at each time point. CRF decline for both groups over time and this was steeper in DCD. Physical activity explained a small part of the difference in CRF.
King- Dowling et al., 2018** Same sample as CATCH study	CS	88%	111 DCD 177 rDCD 301 TD Children 4–5 years	MABC-2, DCD < 5th, rDCD between 6th and 16th					Parents confirmed motor difficulties not due to another condition	There was a large main effect of DCD group on both musculoskeletal and aerobic fitness performance, children with DCD had the greatest fitness deficits. No significant group differences regarding MVPA.
Fong et al., 2018	CS	75%	52 DCD (7.5 years) 61 TD (7.2 years)	BOMPT < or equal to 42 OR MABC-2, <5th	DCD-Q				Excluded those with diagnosed disorders which would better explain the difficulties	After accounting for effects of age, gender, height, lean mass, and fat mass, the total activity diversity score remained independently associated with leg BMC in children with DCD, explaining 5.1% of the variance. PA diversity score was not associated with leg BMC.

TABLE 2 (Continued)

Author	Study design	% Quality score	Sample	DMS IV ar	nd DSM 5*	DSI	M IV	DSM	1 5	Summary outcome
				Criterion A Poor acquisition and execution of motor skills	Criterion B The deficits interfere with activities of daily living	Criterion C Not due to general medical condition and does not meet criteria for PDD	Criterion D Motor difficulties are in excess of any intellectual deficits	Criterion C Onset of symptoms is in the early developmental period	Criterion D Not better explained by intellectual, visual or neurological condition	
Yu et al., 2021	CS	63%	73 DCD 99 TD 6–10 years	MABC-2, <u>≤</u> 5th	MABC-2 Checklist or the Caregiver Assessment of Movement Participation completed by teachers and/or parents				No known neurological or intellectual impairments or other medical conditions	33% of children with DCD met MVPA guidance. DCD had poorer FMS proficiency in jumping and catching and running, jumping, catching, and kicking.
Psycholog	ical Capab	ility								
Cairney et al., 2005b	CS	75%	44 DCD 556 TD 9–14 years	BOTMP-SF, ≤10th			Excluded those with known learning disabilities/physical health problems			Generalized Self-Efficacy: Children with DCD reported lower self-efficacy to PA.
Cairney et al., 2005a	CS	88%	44 DCD 556 TD 9–14 years	BOTMP-SE, ≤10th			Excluded those with known learning disabilities/physical health problems			Regardless of gender, children with DCD had lower self-efficacy toward physical activity and participated in fewer organized and recreational play activities. Girls with DCD had the lowest mean scores of all children.
Poulsen et al., 2007	QE	100%	60 DCD 113 TD Mean age 11 years	MABC	Reported difficulties with tasks of daily living	No reported intellectual impairment	No reported diagnosed emotional, neurological or motor disorder			Group differences in loneliness and sports participation and social-physical participation. Relationship between MABC score and loneliness was mediated by sports participation.
Cairney et al., 2007	CS	100%	44 pDCD 546 TD 9–14 years	BOTMP-SF, ≤10th		Children with known learning disabilities and physical health problems were excluded				Children with probable DCD reported lower average enjoyment scores, lower perceived adequacy, higher percentage body fat and lower cardiorespiratory fitness. Negative correlation between probable DCD and enjoyment of PE.
Kane and Bell, 2009	Case series	70%	3 DCD children	BOTMP-SF	DCD-Q and leisure section of Canadian Occupational and Performance Model	Excluded if had any known neurological condition	Excluded is WISC < 70			Self-efficacy for PA is a key contributor to participation in PA.

Author	Study design	% Quality score	Sample	DMS IV ar	nd DSM 5*	DSI	M IV	DSM	15	Summary outcome
				Criterion A	Criterion B	Criterion C	Criterion D	Criterion C	Criterion D	
				Poor acquisition and execution of motor skills	The deficits interfere with activities of daily living	Not due to general medical condition and does not meet criteria for PDD	Motor difficulties are in excess of any intellectual deficits	Onset of symptoms is in the early developmental period	Not better explained by intellectual, visual or neurological condition	
Silman et al., 2011	CS	100%	61 pDCD 61 TD 12–13 years	MABC-2, ≤15th			KBIT-2 used to determine IQ			Lower perceived adequacy in DCD compared to peers. Perceived adequacy and physical activity were significant mediators. in the relationship between DCD and fitness.
Engel-Yeger et al., 2012	CS	100%	33 DCD (7.67 years) 33 TD (7.84 years)	MABC,<15th			Diagnosed by a pediatrician/ neurologist			Children with DCD showed lower preference to participate in activities compared to peers.
Kwan et al., 2013 Same sample as PHAST study	CS	100%	19 pDCD 42 TD 13–14 years	MABC-2, 15th	Had MABC-2 scores across two time points with both falling below 15th, taken as indication of ADL		KBIT-2 used to determine IQ			Poorer physical activity cognitions in DCD compared to peers. Attitudes and subjective norms for PA partially mediated the relationship between DCD and PA.
Batey et al., 2014 Same sample as PHAST study	CS	88%	29 pDCD (13.3 years) 76 TD (13.2 years)	MABC-2, <15th			Excluded if <70 on Kaufman IQ test			A direct effect of DCD on PA was observed for boys, but not for girls. Neither task efficacy nor barrier efficacy influenced the relationship between DCD and PA.
Noordstar et al., 2014	CS	88%	31 DCD 31 controls 7–12 years	MABC	Reported difficulties with activities of daily living				No underlying neurological disorders were present	No difference between groups for perceived athletic competence scores, but low perceptions of athletic competence were more common in the DCD group.
Engel-Yeger et al., 2015	CS	75%	37 DCD 24 TD 6.10-9 years	MABC,<15th	All had been referred for therapy	Excluded if they had positive neurological signs/visual impairments				Children with DCD showed lower adequacy of physical activity compared to peers. Children with lower adequacy of physical activity showed lower motor performance (predicted 78% of total MABC score).

TABLE 2 (Continued)

Author	Author Study % design Quality score		Sample	DMS IV ar	nd DSM 5*	DSI	DSM IV		15	Summary outcome
				Criterion A	Criterion B	Criterion C	Criterion D	Criterion C	Criterion D	
				Poor acquisition and execution of motor skills	The deficits interfere with activities of daily living	Not due to general medical condition and does not meet criteria for PDD	Motor difficulties are in excess of any intellectual deficits	Onset of symptoms is in the early developmental period	Not better explained by intellectual, visual or neurological condition	
Noordstar et al., 2017	RCT	69%	Intervention group: 20 DCD Care as usual group: 11 DCD All 8 years	MABC-2	DCD-Q			Stated this met as children were between 7 and 10 yrs	Had no known neurological disorders	No effect of the different interventions on leisure PA or total PA.
Wright et al., 2019	CS	100%	60 TD 19 At risk 38 DCD 6–12 years	MABC-2, ≤15th rDCD, <5th DCD	Parents reported that their child had difficulty performing recreational and daily activities			Stated was all in early developmental period (not clear how determined)	Children with an intellectual disability or medical condition were excluded	Children with DCD had lower PA predilection and adequacy regarding PA, higher body fat percentage, received less logistic support. TD children had increased muscle strength compared to the DCD and at risk groups.
Zimmer et al., 2020**	Qual	80%	6 DCD 10–12 years	MABC-2, <16th	DCD-Q				No reports of known conditions which would better explain motor difficulties	Three themes captured experiences of stress in physical education for children at risk for DCD: (1) "they hurt me" (2) "it's hard for me" (3) "I have to."
Physical or	pportunitie	es	'	'	'	'				
Adams et al., 2018	QE and Qual	80%	162 Physical Therapists (survey) + plus 10 with interview data 9 DCD (interviews) 9–12 years and parents	MABC-2, <16th	Being treated or had been treated for motor difficulties by physical therapists			Considered by physical therapists treating the children	Considered by the physical therapists treating the children	Barriers to participation included motor impairment, insufficient numbers to create a team and lack of inclusive practice.
Social opp	ortunities									
Barnett et al., 2013	Qual	70%	8 child and parent pairs All boys 13–15 years	MABC-2, ≤5th	MABC-checklist, ≤5th	BPVS score > 70	Parents reported no serious physical, sensory impairment			Majority were physically inactive but wanted to be more active. Cited poor motor skill, lack of motivation and reports of fatiguing easily, difficulty traveling to activities, negative comments from peers and teachers' lack of understanding of DCD as barriers to increasing PA.

TABLE 2 (Continued)

Author	Study design	% Quality score	Sample	DMS IV ar	nd DSM 5*	DSM IV		DSM 5		Summary outcome
				Criterion A	Criterion B	Criterion C	Criterion D	Criterion C	Criterion D	
				Poor acquisition and execution of motor skills	The deficits interfere with activities of daily living	Not due to general medical condition and does not meet criteria for PDD	Motor difficulties are in excess of any intellectual deficits	Onset of symptoms is in the early developmental period	Not better explained by intellectual, visual or neurological condition	
Reflective	Reflective motivation									
Meek and Sugden, 1997	QE	63%	197 7-year-olds 197 11-year-olds 59 14-year-olds	томі-н	A checklist of behaviors associated with DCD was completed	Recruited from mainstre schools so assumed no learning, emotional or physical difficulties)			No significant differences at either 7 or 11, but by 14 years of age the children with DCD had formed significantly lower attitudes than their class peers.
Automatic	motivation	n								
Sit et al., 2019	RCT	69%	Intervention group: 64 DCD and 64 TD Control group: 67 DCD and 67 TD 6–10 years	MABC-2, <5th DCD and 6th-16th rDCD	MABC-2 checklist				Those with visual, neurological or intellectual impairment were excluded	Fundamental movement skills training group spent more time in MVPA and reported greater enjoyment of PA after intervention which was not the case for the control group.
Li et al., 2021 Same sample as CATCH study	CS	89%	288 rDCD 301 TD 4–5 years	MABC-2, <16th					IQ > 70 (except for one child) no other medical condition which may lead to motor impairments.	Children with rDCD reported more internalizing problems which physical activity and BMI did not mediate.

^{*}Criteria summarized.

^{**}Does not state which version of the diagnostic criteria they were and so have been assigned on the basis of the publication date.

CT, randomized controlled trial; CS, cross sectional; Qual, qualitative; QE, quasi-experimental; Long, longitudinal.

3.1.2 Setting

Nineteen studies were conducted in Canada (Cairney et al., 2005a,b, 2006, 2007, 2010, 2017, 2019; Kane and Bell, 2009; Baerg et al., 2011; Silman et al., 2011; Kwan et al., 2013; Batey et al., 2014; Joshi et al., 2015; King-Dowling et al., 2018, 2019; Zimmer et al., 2020; Brown et al., 2021; James et al., 2021; Li et al., 2021), seven in Australia (Poulsen et al., 2007, 2008, 2011a; Beutum et al., 2013; Howie et al., 2016, 2017; Wright et al., 2019), six in Hong Kong (Fong et al., 2011, 2018; Yu et al., 2016a,b, 2021; Sit et al., 2019), three in the United Kingdom (Meek and Sugden, 1997; Green et al., 2011; Barnett et al., 2013), three in the Netherlands (Noordstar et al., 2014, 2017; Adams et al., 2018) two in Israel (Engel-Yeger et al., 2012, 2015), one in Israel and the US (Cermak et al., 2015), one in Finland (Tan et al., 2022) and one in South Africa (Bonney et al., 2017a).

3.1.3 Identification of DCD

Twenty-one studies used the DSM-IV (American Psychiatric Association [APA], 1994) criteria for DCD; in line with our inclusion criteria. All administered a motor assessment (criterion A), the test component of the Movement Assessment Battery for Children second edition (MABC-2) (Henderson and Sugden, 2007) was used in 24 studies, the Bruninks Test of Motor Proficiency (BOTMP) (Bruininks and Bruininks, 1978) was used in eight studies, the test component of the Movement Assessment Battery for Children first edition (MABC) (Henderson et al., 1992) was used in six studies, a combination of the BOTMP and MABC or MABC-2 was used in three studies, the Zurich Neuromotor assessment (Largo et al., 2001) and the Test of Motor Impairment-Henderson (TOMI-H) (Stott et al., 1986) were each used in one study. A total of 12 studies described how participants met criteria B and C, and 17 described criterion D. Twenty-two studies used the DSM-5 (American Psychiatric Association [APA], 2013), in line with our inclusion criteria, all administered a motor assessment, 16 described how participants met criterion B, seven described criterion C, and 20 described criterion D. Authors typically used probable DCD (pDCD) to describe participants aged under 5 years or when all diagnostic criteria had not been assessed and at risk of DCD (rDCD) when participants fell between the 6th and 16th percentile on a standardized motor assessment. None of the studies included children under the age of 4 years old.

3.1.4 Quality of the studies

Based on the study design, appropriate critical appraisal tools were used; percentage scores for methodological quality are presented in Table 2. Total percentage quality scores ranged from 62 to 100%; therefore, all included articles were of moderate to high quality. Randomized controlled trials ranged from 62 to 100%, quasi-experimental studies from 63 to 100%, cross-sectional studies from 63 to 100%, longitudinal studies from 71 to 100%, qualitative studies from 70 to 80%, the mixed-method study was 80% and case series study was 70%.

3.2 COM-B analysis

The results are presented within the framework of the COM-B model. A few of the included studies touched upon multiple

components of the COM-B model (Yu et al., 2016a,b); these studies were aligned to a single component based on the primary focus of the study.

3.2.1 Physical capability: physical strength, skill, or stamina (capacity to engage in the necessary physical processes)

Nine articles were best aligned with this component of the COM-B model (Fong et al., 2011, 2018; Joshi et al., 2015; Yu et al., 2016a,b; Bonney et al., 2017a; Cairney et al., 2017; King-Dowling et al., 2018; Li et al., 2021).

In terms of physical skill, Fong et al. (2011) used the Children's Assessment of Participation and Enjoyment (CAPE) questionnaire (Imms, 2008) to determine whether motor ability and weight status were associated with physical activity participation diversity in children aged 6-12 years with (N = 81) and without (N = 67)DCD. Children with DCD had significantly lower CAPE total participation intensity scores than TD children. Specifically, the authors highlighted that motor ability was positively correlated with CAPE total diversity scores in children with DCD, accounting for 7.6% of the variance in CAPE total diversity scores. In other words, children with DCD who presented a higher motor skill level participated in more formal, recreational and skill-based activities. Conversely, weight status was negatively correlated with total CAPE and recreational activity diversity scores, indicating that children with higher weight status participated in fewer activities. Therefore, physical skill, motor impairment and weight status contributed to a lack of participation in physical activity in children with DCD. This is supported by Fong et al. (2018), who compared bone mineralization and activity patterns of 52 children with DCD (mean age 7.5 years) and 61 TD children (mean age 7.2 years). After accounting for age, sex, height, lean mass and fat mass, bone mineralization and activity participation were lower in children with DCD compared to TD children. The authors recommended that children with DCD should be encouraged to participate in various activities, not just physical activity, to improve bone mineralization in prepubertal years.

Taking a different approach to physical skill, Yu et al. (2016a) conducted a quasi-randomized controlled repeated measures single-blind trial to measure FMS using the Test of Gross Motor Development-2nd edition (Ulrich, 2004). They found that FMS training effectively improved both locomotor skills (jumping) and object-control skills (catching and kicking) of children aged between 8 and 10 years with DCD (N = 38); improvements in object-control skills (catching and throwing) were sustained for at least 6 weeks. FMS training also effectively improved the self-perceived physical competency of children with DCD in terms of physical coordination, physical strength and physical fitness immediately after the training. In a follow-up study, Yu et al. (2016b) examined differences in FMS proficiency, physical self-concept and physical activity in children aged 7-10 years with DCD (N = 43) and age-matched TD children (N = 87). They found that physical activity was correlated with FMS proficiency. Children with DCD reported significantly poorer selfconcept on physical coordination and sporting ability, which was more pronounced for girls with significantly lower physical activity levels.

In a later study, Yu et al. (2021) explored differences in FMS in a large sample of children with DCD (N=73) and TD children (N=99) aged 8–9 years; they explored whether FMS was associated with moderate to vigorous physical activity (MVPA) and sedentary behavior. Using accelerometry to assess MVPA and five components of FMS (running, jumping, throwing, catching, kicking) from the Test of Gross Motor Development-2nd edition (Ulrich, 2004), they found that children with DCD had significantly poorer FMS proficiency in terms of specific movement patterns (jumping and catching) and outcomes (running, jumping, catching, and kicking). However, there were no significant differences in MVPA and sedentary behavior between children with DCD and TD. However, specific FMS movement patterns (running, jumping, catching) were closely related to MVPA and sedentary behavior in children, moderated by motor coordination status and sex.

In relation to physical strength and stamina, as part of the Physical Health Activity Study Team (PHAST), Joshi et al. (2015) found that children with probable DCD (pDCD; N = 103) had higher body mass index (BMI) and waist circumference than TD children; this difference between groups increased from baseline when the children were 9–10 years old over the 5-year study period. Boys with pDCD had a more rapid increase in BMI and waist circumference than girls with pDCD. Physical activity levels did not mediate or moderate the relationship between pDCD and measures of body composition. However, physical activity was negatively associated with measures of body composition. Likewise, using the same cohort, Cairney et al. (2017) evaluated whether physical activity levels could account for poor fitness among children with pDCD over a 5-year period. They reported that children with pDCD had poorer cardiorespiratory fitness compared to TD children; however, cardiorespiratory fitness in pDCD children at age 9 years was comparable, with a slight increase noted at age 14 years, which could not be explained by differences in selfreported physical activity at these ages.

Further evidence of differences in physical stamina and physical strength comes from King-Dowling et al. (2018), who examined differences in children at 4–5 years with DCD (N=111), at risk of DCD (N=177) and TD children (N=301) from the Coordination and Activity Tracking in Children (CATCH) sample to determine whether vigorous physical activity (VPA) levels mediated differences in health-related fitness. They found a significant main effect of the DCD group on musculoskeletal and aerobic fitness performance; however, daily VPA was similar across groups and did not explain health-related fitness differences.

Using a gamification approach to physical strength and stamina, Bonney et al. (2017b) randomly allocated females aged 13–16 years with DCD to one of two intervention groups. The first intervention involved a 45 min Nintendo Wii session, and the second involved task-oriented functional training. Both interventions were held once weekly for 14 weeks. Blinded assessors measured outcomes at baseline and at the end of the intervention period, which included impairment-based outcomes (e.g., isometric strength), activity-based outcomes (e.g., a stair climbing test) and participation-based outcomes [e.g., Children's Self-perceptions of Adequacy in and Predilection for Physical Activity (CSAPPA) questionnaire; Hay, 1992]. Both interventions improved muscle strength, motor proficiency, functional performance, self-efficacy and participation in activities of daily living (ADLs). In addition, although there was no

statistically significant difference in aerobic stamina (running task) between pre- and post-test, significant changes were found in a predilection for physical activity and overall self-efficacy score. Improvements in participation in ADLs were also observed.

3.2.2 Psychological capability: knowledge, psychological strength, skill, or stamina (capacity to engage in necessary thought processes)

Thirteen articles best aligned with the psychological capability component of the COM-B model (Cairney et al., 2005a,b; Poulsen et al., 2007; Kane and Bell, 2009; Silman et al., 2011; Engel-Yeger et al., 2012, 2015; Kwan et al., 2013; Batey et al., 2014; Noordstar et al., 2014, 2017; Wright et al., 2019; Zimmer et al., 2020).

Regarding psychological skill, Cairney et al. (2005b) quantitatively explored whether 9-14-year-old children with pDCD (N = 44) report lower levels of self-efficacy toward physical activity and engage in less free play and organized activities than their TD peers (N = 546) taking sex into account. Although girls with DCD had the lowest mean scores, all children with pDCD reported lower self-efficacy scores to participate in physical activity and lower levels of participation in free and organized play compared to children without DCD. In a follow-up study, Cairney et al. (2005a) investigated the effect of sex on the relationship between pDCD and self-reported participation in organized and recreational free-play activities. Data from 44 pDCD children and 556 TD children aged between 9 and 14 years showed that regardless of sex, children with pDCD had lower self-efficacy toward physical activity and participated in less organized and free-play activities than TD children. Again, girls with pDCD had the lowest mean scores of all children.

These findings are supported by a more recent qualitative study by Zimmer et al. (2020), who explored physical education experiences among six children at risk of DCD (10–12 years) through two semi-structured interviews with each child. To describe the stressors that children at risk of DCD experience in relation to physical education, three themes were identified using interpretative phenomenological analysis within the framework of relatedness, competence and autonomy: (a) they hurt me, referring to psychological and physical harm sustained from peers; (b) it's hard for me, referring to difficulties in taking part in activities and (c) I have to, referring to perceived teacher's demands. The authors highlight that while the stressors these children experienced interfered with fulfilling their basic psychological needs for relatedness, competence and autonomy, they primarily used coping strategies to minimize their experiences of stress.

Likewise, in a case-series study of three children aged 9–11 years with DCD, Kane and Bell (2009) evaluated a 6-week group exercise program and measured self-perceived adequacy, synonymous with psychological skill, for physical activity as one of the outcomes. Only one of the three children saw a considerable improvement in self-efficacy (pre-test: 55; post-test: 73) as measured by the Children's Self-Perceptions of Adequacy in and Predilection for Physical Activity (Hay, 1992). While this child did not see any changes in Bruininks-Oseretsky Test of Motor Proficiency (BOTMP) (Bruininks and Bruininks, 1978) scores, the self-rated performance of their motor goals improved. One of the other children also rated their performance on their motor goals higher after intervention and had improved greatly on the BOTMP

but saw little change in their self-perceived adequacy for physical activity. The third child saw little to no change in motor skills and self-efficacy. Thus, it appears that the relationship between motor performance and self-perceived psychological skills for physical activity is not the same for everyone. Kane and Bell (2009) contend that both factors likely affect participation; therefore, both should be considered important outcomes when evaluating interventions designed to increase physical activity.

In a more extensive study, Engel-Yeger et al. (2012) examined preference differences between children aged 7 years with (N = 33)and without (N = 33) DCD to participate in leisure activities, their physical activity levels as reported by their sports teacher and whether reports from their sports teacher could predict participation preferences. Significant differences were found in participation preference between groups based on the Preference for Activities of Children (PAC) (King et al., 2007) and Teacher Estimation of Activity Form (TEAF), a measure of sports performance and adequacy of physical activity (Hay, 1992). They found that TEAF scores successfully predicted children's preference to participate in leisure activities, suggesting psychological skill is related to participation in physical activity. A more recent crosssectional study examined the relationship between self-efficacy and motor performance in 37 children with DCD and 24 TD children (6-9 years) (Engel-Yeger et al., 2015). Children with DCD scored significantly lower on all self-efficacy scores on the Perceived Efficacy and Goal Setting (PEGS) (Missiuna et al., 2004) compared to their TD peers and sports teachers rated children with DCD significantly lower on the TEAF (Hay, 1992). Lower TEAF scores were associated with poorer motor scores on the MABC (Henderson and Sugden, 2007) and lower self-efficacy on the PEGS (Missiuna et al., 2004). The authors suggest that with failed attempts to learn motor skills due to poor motor ability, children with DCD develop lower self-perceptions and lower self-efficacy, which then creates a negative feedback loop that reduces their motivation to practice motor skills.

In a further study by Noordstar et al. (2014), they looked at the differences and relationships between perceived athletic competence and physical activity in children aged 7-12 years with (N = 31) and without (N = 31) DCD. The DCD group participated in less total physical activity than the TD children, primarily driven by less participation in unorganized physical activity in children with DCD. In relation to perceived psychological skill, no significant group differences were seen in perceived athletic competence levels. However, when the authors split both the group with DCD and the group without DCD into sub-groups with "high" or "low" perceived competence, no difference in terms of physical activity was seen between the DCD group and the TD group when their level of perceived athletic competence was low. Conversely, when perceived athletic competence was high, TD children showed greater physical activity levels than children with DCD. These findings suggest that a perception of high physical athletic competence drives physical activity in children without DCD but not in children with DCD.

Further support for poor physical ability self-concept comes from Poulsen et al. (2007), who used the Self-Description Questionnaire-I (Marsh, 1990) to examine the relationship of self-concept with patterns of physical activity in 10-13-year-old boys with (N=60) and without (N=113) DCD. Not surprisingly, the boys with DCD reported significantly lower physical ability

self-concept than their coordinated peers. Significantly lower general and peer relations self-concept were also noted in children with DCD. Despite the small effect size, self-perceptions of peer relationships mediated low energy expenditure patterns, suggesting that the social context may have more influence on increasing physical activity than physical ability self-concept. Another recent study explored barriers and task self-efficacy toward physical activity (Batey et al., 2014). A subset of participants from the PHAST study, aged 13-14 years, were asked to complete the self-efficacy scale (Foley et al., 2008) to assess their perceived psychological stamina to complete different intensities and duration of physical activity (task efficacy) and their confidence in completing physical activity when faced with everyday barriers (barrier efficacy). An accelerometer was used to record activity for 1 week. The authors found that children with pDCD (N = 29) spent significantly less time in MVPA and had significantly lower task and barrier self-efficacy toward physical activity than their TD peers (N = 76).

Another study from the PHAST cohort was conducted by Silman et al. (2011), who examined whether perceived adequacy and physical activity mediated cardiorespiratory fitness, as measured by peak aerobic power, in children with (N = 61) and without (N = 61) pDCD at age 12–13 years. Overall, they found that children with pDCD had lower perceived adequacy toward physical activity; perceived adequacy and physical activity were significant mediators in the relationship between pDCD and peak aerobic power. In another study that utilized the PHAST cohort, Kwan et al. (2013) specifically explored the influence of physical activity cognition amongst boys aged 13-14 years with (N = 19)and without (N = 42) pDCD within the framework of the Theory of Planned Behavior (Ajzen, 1991). The authors found that boys with pDCD had poorer physical activity cognitions than TD boys. These differences were most evident in their attitude and perceived behavioral control related to being physically active, showing that the relationship between pDCD and MVPA is partially mediated by physical activity cognitions in boys with pDCD.

Similarly, Wright et al. (2019) anticipated that perceived competence, enjoyment and predilection for physical activity would be lower amongst children aged 6–12 years with DCD (N=38) or at risk of DCD (N=19) relative to TD children (N=60). They also hypothesized that there would be a significant difference in physiological characteristics between TD children, children at risk of DCD and children with DCD and children either at risk of DCD or with DCD would have lower cardiorespiratory fitness and physical activity levels. They found that children with or at risk of DCD reported lower scores on psychological constructs that are predictive of physical activity involvement relative to TD children. Children with or at risk of DCD also had multiple physiological deficits (e.g., muscle strength) and received less parental logistic support for physical activity involvement (e.g., transportation).

Cairney et al. (2007) also explored perceived enjoyment of physical education and examined correlations between enjoyment and body fat, cardiorespiratory fitness and perceived adequacy in children with pDCD (N=44) at 9–14 years. They found that children with greater perceived adequacy, lower body fat and higher cardiorespiratory fitness were more likely to enjoy physical education. They also noted that children with pDCD were more likely to be above the normal, healthy weight for their age, have

poorer physical fitness and perceive themselves as less adequate (the most significant contributing factor to the enjoyment of physical education) about their physical abilities than children without pDCD.

In an intervention study, Noordstar et al. (2017) compared a motor intervention alone (N=11) with a motor intervention coupled with a program to boost psychological skill (N=20) in 8-year-old children with DCD. Motor control perceived self-competence and general self-esteem all improved over time; however, there were no effects of the intervention group on any of these measures. Despite these positive changes in both groups, no differences were found in physical activity levels. Therefore, it would seem, in line with the findings from Noordstar et al. (2014), that a change in perceived athletic competence does not result in a behavior change amongst 8-year-old children with DCD in this context.

3.2.3 Physical opportunity: opportunities provided by the environment, such as time, location, or resource (physical opportunity provided by the environment)

Only one study considered the physical opportunity component of the COM-B model. In the only identified mixed-method study, Adams et al. (2018) explored the role of pediatric physiotherapists in promoting sports participation in children with DCD. A total of 162 physiotherapists completed a survey and 10 physiotherapists and 9 children with DCD (9–12 years) took part in interviews. Although nearly half of the physiotherapists surveyed signposted children with DCD to sports clubs, the interview data suggest that matching sports to children's motor ability wishes and preferences facilitated participation. Identified barriers included a lack of understanding of DCD and the motor difficulties experienced by children with DCD.

3.2.4 Social opportunity: opportunities as a result of social factors, such as social norms and social cues (cultural milieu that dictates the way we think about things)

There was limited literature exploring the social opportunity component of the COM-B model in the context of physical activity. One study, however, used semi-structured interviews with eight 12- to 15-year-old boys with DCD and their parents to examine barriers and facilitators to participation in physical activity (Barnett et al., 2013). Half of the children with DCD and all but one parent reported that teenagers with DCD did little physical activity. Dislike of competitive team games, lack of nearby resources, negative comments from peers and teachers, lack of motor skills and confidence, poor motivation, lack of time, fatigue and pain and lack of understanding of DCD were all constraints to participating in physical activity. In contrast, parental support and intervention activities (such as gym sessions) led to engagement and enjoyment in physical activity. The authors concluded that although teenagers with DCD disliked competitive team games, they reported many physical activities they enjoyed when social opportunities were facilitated and when they were motivated to be more physically active.

3.2.5 Reflective motivation: reflective processes, such as making plans and evaluating things that have already happened (evaluations and plans)

Only one article aligned with the reflective motivation component of the COM-B model. Meek and Sugden (1997) aimed to establish whether children (aged 7–8 years; $N=197,\,10-11$ years; $N=197,\,13-14$ years; N=59) with and without DCD form expectance-value combinations of attitudes prior to the completion of a novel physical activity that significantly differs from their class peers who had either previously played volleyball or not played volleyball. There were no significant between-group differences at age 7 or 11 years, but by 14 years of age, the children with DCD had formed significantly lower attitudes than their class peers. Furthermore, as age increased, attitudes decreased, suggesting that even prior to undertaking physical activity, negative attitudes existed amongst older children with DCD. Such personal barriers may interact with environmental constraints and lead to an overall lack of engagement in physical activity in teenagers with DCD.

3.2.6 Automatic motivation: automatic processes, such as our desires, impulses and inhibitions (emotions and impulses arising from associative learning and/or innate dispositions)

Two articles best aligned with the automatic motivation component of the COM-B model (Sit et al., 2019; Li et al., 2021).

Li et al. (2021) examined the connections between physical activity and weight status to internalizing problems using a modified version of the environmental stress hypothesis as part of the CATCH study. They found that preschool children (4–5 years) at risk of DCD (N=233) experienced more internalizing problems than their TD peers (N=274), including emotion control, withdrawal from social interactions and complaints of somatic responses. Neither physical activity nor BMI mediated the relationship between children at risk of DCD and internalizing problems. It could be argued that preschool children at risk of DCD may be as physically active as their typically developing peers at this age [demonstrated in other studies described in section "3.2.1 Physical capability: physical strength, skill, or stamina (capacity to engage in the necessary physical processes)"].

In a later study, Sit et al. (2019) hypothesized that children with DCD aged 6–10 years who received FMS training would improve their motor skills proficiency and have higher physical activity levels. They perceived competence and enjoyment compared to those receiving conventional physical education. The authors concluded that children in the FMS training group improved locomotor and object control skills and engaged more in MVPA. However, there were no differences to the control group, although children with DCD did report increased enjoyment in physical activity during their leisure time, which was sustained for up to 12 months.

3.2.7 Behavior: the product of perceived capability, opportunity and motivation

Fifteen articles best aligned with the behavior component of the COM-B model (Cairney et al., 2006, 2010, 2019; Poulsen et al., 2008, 2011a; Baerg et al., 2011; Green et al., 2011; Beutum et al., 2013; Batey et al., 2014; Cermak et al., 2015; Howie et al., 2016, 2017;

King-Dowling et al., 2019; Brown et al., 2021; James et al., 2021; Tan et al., 2022).

King-Dowling et al. (2019), using the CATCH sample, aimed to determine if there were differences in patterns of activity levels amongst preschool children (4-5 years) with pDCD (N = 111), children at risk of DCD (N = 177) and TD children (N = 301). They found that preschool children with pDCD and children at risk of DCD had comparable physical activity levels to their TD peers. However, preschool children with pDCD tended to accumulate their MVPA in shorter episodes of physical activity (King-Dowling et al., 2019). This pattern is consistent with evidence from Brown et al. (2021), who also used the CATCH sample to measure the BMI of children aged 4–5 years at risk of DCD (N = 288) and TD agematched children (N = 301). They also measured physical activity and sedentary behavior using an accelerometer whilst parents completed the Child Behavior Checklist (Achenbach, 1991). Both groups were found to engage in similar activity levels (5 h) during a 12-h awake period and movement behavior did not influence children's mental health based on parental reports. Taken together with the findings of King-Dowling et al. (2019), these results suggest that differences in sedentary time and physical activity may develop later in childhood.

An older sample supports the conclusion that differences in physical activity behavior may appear later in childhood. Beutum et al. (2013) recruited 9 children with pDCD and 9 TD children (aged 8 years) and found that children with pDCD participated in significantly less MVPA, had higher BMI and decreased strength and cardiovascular fitness. In addition, strength, activity type and family factors correlated significantly with MVPA for children with pDCD. In a larger cross-cultural study between the United States and Israel, 53 children with DCD and 65 TD children (aged 6-11 years) were recruited to measure relationships between children's motor coordination and their physical activity, sedentary behavior, fitness and weight status (Cermak et al., 2015). In Israel and the United States, children with DCD demonstrated significantly reduced physical activity, increased sedentary behavior, poorer fitness, and increased weight compared with TD children; no significant differences were found between the two countries. Differences in health-related fitness are also supported by the baseline data from a younger sample from the CATCH study (Cairney et al., 2019). Although no differences were observed between groups for BMI percentile or physical activity, children in the "at risk" DCD group (N = 287) had significantly lower aerobic and musculoskeletal fitness and larger waist circumferences compared to TD children (N = 301) at age 4-5 years (Cairney et al., 2019).

However, James et al. (2021) examined the effect of DCD risk amongst preschool children aged 4–5 years using the CATCH sample on MVPA levels when adjusting for ADHD symptomology. They reported that when adjusting for ADHD (particularly inattention), preschool children at risk of DCD (N=288) were significantly less active than their TD peers (N=301), suggesting that ADHD and DCD combined may have a negative impact on levels of physical activity in preschool-aged children. In an older PHAST sample, Baerg et al. (2011) compared physical activity using a 7-day accelerometry analysis of 12–13-year-old children with DCD (N=32), children with DCD/ADHD (N=30), and TD children (N=48). The accelerometer was used to assess step

count and activity energy expenditure of sedentary, light, moderate and vigorous levels of physical activity. The authors reported a sex and group interaction effect for average daily step counts and activity energy expenditure. Specifically, girls with DCD/ADHD had significantly more average step counts per day than TD girls. However, there was no difference between the average step count per day in girls with DCD compared to the DCD/ADHD and TD groups. There was also no significant difference between the average step count per day in boys with DCD, DCD/ADHD, or TD. The authors concluded that hyperactivity, as expressed in children with DCD/ADHD, appears to override the hypoactive behavior typically found in children with DCD. However, this finding was only found in girls and did not translate to boys with DCD/ADHD.

Cairney et al. (2006) explicitly explored whether the activity deficit between children with and without DCD widens or diminishes over time. In a cross-sectional study that administered a participation questionnaire to 44 children with DCD and 537 TD children (aged 9-14 years), they found that children with DCD participated in less structured and free play activities, but this activity deficit did not increase with age (Cairney et al., 2006). In a follow-up longitudinal study, the PHAST sample was used by Cairney et al. (2010) to recruit 111 children with pDCD and 1972 TD children at 9 years of age and followed them at ages 10 and 11 years using a participation questionnaire. The results indicate that divergence in free play activity over time occurs for females with pDCD but not for males. In another longitudinal study, Tan et al. (2022) analyzed longitudinal data to consider associations between the "at risk" status of DCD in childhood and physical activity in adulthood. Those children classified as "at risk" or "probably at risk" of DCD at 56 months were found to have a significantly lower number of steps over a 10-day period at 25 years of age compared to those children who were not at risk for DCD. Furthermore, statistical modeling indicated that DCD "risk status" increased time spent in sedentary light activity and decreased time spent in MVPA.

Other sex differences have also been noted. Using a sample from the Avon Longitudinal Study of Parents and Children (ALSPAC) sample, Green et al. (2011) explored whether children with pDCD (N = 193) had an increased risk of reduced MVPA compared to TD children (N = 4,138) at two time points (t1: 7-8 years; t2: 12-13 years) using accelerometry for 7 days. Boys with pDCD were less physically active than boys without pDCD at ages 7 and 12 years. There were no differences in levels of MVPA in girls with and without pDCD, which the authors suggest may reflect a generally low level of MVPA across the entire sample. Additionally, Poulsen (2008) asked parents of 60 boys aged 10-13 years with DCD and 113 boys without DCD to complete a 7-day leisure time diary and record the intensity, duration, content and social/physical environment of leisure time activities. A total daily score for low-intensity activities (LPA) and MVPA and the total metabolic (MET) levels were computed for 1 week's activities. Boys with DCD spent significantly less time engaged in MVPA compared to boys without DCD but spent significantly more time in LPA. This pattern of leisure physical activity contributed to significantly lower energy expenditure in boys with DCD compared to their peers. Interestingly, the highest percentage of out-ofschool time for both groups was devoted to sedentary, unstructured pursuits (e.g., television, electronic media). Boys with DCD had

significantly lower participation in structured (e.g., team sports) and unstructured (e.g., street ball, running games) social physical activities; no significant differences were noted between groups for physical non-social activities (e.g., individual sports). In a followup study, Poulsen et al. (2011b) explored the differences in the number and context of leisure-time personal projects reported in boys with and without DCD. Group-matched 10-12-year-old boys with (N = 60) and without DCD (N = 113) completed the Personal Project Analysis for Children (Christiansen, 2000). Boys with DCD identified significantly fewer MVPA, team sports, popular sports and structured physical activity personal projects than boys without DCD. Boys with DCD accounted for 81.4% of participants involved in no team sports and 74.4% of participants who participated in two or fewer activities involving MVPA. Furthermore, the majority of physical activity reported by participants with DCD were completed individually or in the home environment.

In terms of exploring whether interventions can improve physical activity levels in children with DCD, Howie et al. (2016) recruited 21 children with DCD or at risk of DCD (aged 10-12 years) to take part in a crossover active video game (AVG) intervention. The intervention (AVG, no AVG) periods were 16 weeks for 20 min a day, 4-5 days per week. Accelerometers at baseline and following each intervention period measured minutes of sedentary, light, moderate and vigorous durations alongside self-reported activity types. The authors found that the AVG intervention did not improve physical activity or sedentary time. In a follow-up study to determine barriers to interventions, Howie et al. (2017) considered why the AVG intervention did not increase physical activity in the same sample. Although some participants (N = 5) significantly increased their physical activity following the intervention, this was not the case for all participants. In addition, there were no relationships between engagement with the AVG in terms of playing time and changes in physical activity, suggesting that levels of engagement did not explain these individual differences. Therefore, the exact barriers to AVG interventions remain unclear.

4 Discussion

The evidence relating to physical capability was of moderate to high quality (63-100%) and suggests that children with DCD have poorer motor skills, lower bone mineralization and participate in less varied formal, recreational and skill-based activities (Fong et al., 2011, 2018). Children with DCD also have higher BMI and waist circumference, which is especially the case for boys (Joshi et al., 2015). Physical activity levels do not seem to account for these differences (Joshi et al., 2015), nor does physical activity mediate the poorer cardiorespiratory fitness seen in children with DCD (Cairney et al., 2017) and VPA does not explain differences in health-related fitness (King-Dowling et al., 2018), although it is possible that lower levels of physical activity in DCD may be a consequence of these differences. FMS differences have also been found in children with DCD (Yu et al., 2021), with FMS proficiency correlated with physical activity (Yu et al., 2016b). In addition, there is evidence that FMS proficiency can be improved with FMS training (Yu et al., 2016a). Both Nintendo Wii and task-oriented functional interventions appear to improve muscle strength, motor proficiency, functional performance, self-efficacy and participation in ADLs (Bonney et al., 2017a), at least in the short term. In the context of physical capability, the evidence suggests that children with DCD have poorer physical skills and physical strength, resulting in poorer physical stamina.

Overall, the evidence relating to the psychological capability, which was of moderate to high quality (69-100%), suggests that children with DCD have lower levels of self-efficacy and perceived athletic competence toward physical activity, with the lowest selfefficacy reported amongst girls (Cairney et al., 2005a,b; Poulsen et al., 2007; Silman et al., 2011; Kwan et al., 2013; Batey et al., 2014; Wright et al., 2019), despite fitness differences being found in boys with DCD. Qualitative research also suggests that the stressors experienced by children with DCD around compulsory physical education are often managed using coping strategies (Zimmer et al., 2020), which are important self-management approaches given that physical activity interventions may not improve motor skills or self-efficacy (Kane and Bell, 2009). As a result, children with DCD may instead develop lower self-perceptions, lower self-efficacy and perceive themselves as less adequate in their physical abilities than children without DCD (Cairney et al., 2007), creating a negative feedback loop that reduces their motivation to practice motor skills and participate in physical activity (Engel-Yeger et al., 2012, 2015). Interestingly, perceptions of high physical athletic competence may drive physical activity in children without DCD but not in children with DCD (Noordstar et al., 2014, 2017) (see Dreiskämper et al., 2022 for an extensive discussion of this in TD children).

One article of high quality (80%) aligned with the physical opportunity component of the COM-B and identified that signposting children with DCD to sports clubs required consideration of children's motor skills, wishes and preferences with a lack of understanding identified as a barrier to participation (Adams et al., 2018). Social opportunity, lack of motor skills and confidence, poor motivation, lack of time and fatigue and pain are all reported barriers to participation in physical activity (Barnett et al., 2013) based on one high-quality study (70%). The evidence relating to reflective and automatic motivation, which was of moderate to high quality (63-100%), suggests that young people with DCD have negative attitudes toward physical activity (Meek and Sugden, 1997). Even preschool children at risk of DCD have greater internalizing problems than their typically developing peers (Li et al., 2021). However, one study found that following an FMS intervention, children with DCD reported increased enjoyment in physical activity during their leisure time, which was sustained for up to 12 months (Sit et al., 2019).

Taken together, the evidence aligning with the behavior component of the COM-B, which was of moderate to high quality (62–100%), suggests that levels of physical activity appear unaffected in pre-school children with DCD (Cairney et al., 2019; King-Dowling et al., 2019; Brown et al., 2021) unless there is co-occurring ADHD (James et al., 2021). Older children with DCD (>6 years) have lower step counts (Tan et al., 2022), lower levels of LPA and MVPA (Poulsen et al., 2008; Beutum et al., 2013; Cermak et al., 2015), higher BMI, decreased strength (Beutum et al., 2013), poorer fitness (Cermak et al., 2015) and participate in less structured and free play activities which do not change with age (Cairney et al., 2006), although divergence in free play activities was found for females with pDCD over time (Cairney et al., 2010). Other sex differences have also been noted whereby, compared to

girls, boys with pDCD are generally less physically fit (Baerg et al., 2011; Green et al., 2011) and engage in less physical activity (Batey et al., 2014). In terms of improving physical activity levels, there is currently insufficient evidence to support the implementation of home-based AVG interventions for children aged 10–12 years with DCD (Howie et al., 2016, 2017).

4.1 Strengths and limitations

This is the first systematic review that has considered physical activity amongst children with DCD in the context of a well-established behavior change model, the COM-B (Michie et al., 2011). The conduct of this review was supported by a multi-disciplinary team specializing in DCD research. The review followed the JBI methodology, which is well known for the conduct of rigorous evidence synthesis to promote and implement evidence-based decisions. Using JBI critical appraisal tools allowed for a detailed and nuanced assessment of different study designs.

However, the strict adherence to the eligibility criteria, specifically the need for authors to have explicitly stated how two or more of the DCD diagnostic criteria had been met, may have resulted in some relevant papers not being included. Furthermore, multiple studies drew on the same sample from the PHAST study between 2010 and 2017 (Cairney et al., 2010, 2017; Baerg et al., 2011; Kwan et al., 2013; Batey et al., 2014; Joshi et al., 2015) and the CATCH study between 2018 and 2021 (King-Dowling et al., 2018, 2019; Cairney et al., 2019; Brown et al., 2021; James et al., 2021; Li et al., 2021). These samples may not capture the demographic heterogeneity of the wider DCD population.

4.2 Future research

Based on the COM-B model of behavior change (Michie et al., 2011), future research could consider the reflective motivation and physical and social opportunities for children with DCD to engage in physical activity, an area generally neglected to date. Furthermore, based on the reviewed literature, there appear to be inconsistencies in implementing the diagnostic criteria for DCD in research. There were limited examples of all diagnostic criteria being considered. Therefore, future research should ensure the careful description of all criteria before grouping samples as DCD, rDCD, pDCD and TD. This will enable a more precise picture to emerge and opportunities for meta-analysis. In addition, only one identified study considered physical activity in adults with DCD (Tan et al., 2022). There is, therefore, a gap in understanding the capability, opportunity, motivation and behavior of adults with DCD in the context of physical activity.

4.3 Practical implications and recommendations

There is some evidence suggesting that FMS training (Yu et al., 2016a), Nintendo Wii interventions, and task-oriented functional interventions (Bonney et al., 2017a) may improve physical capability and that this, in turn, may improve participation

in physical activity. However, recommendations for future interventions can be derived from the data obtained in this systematic review using the Behavior Change Wheel (BCW). The COM-B model forms the hub of the BCW, a systematic behavioral science tool for developing and characterizing interventions for health behavior change (Michie et al., 2011). The BCW is a synthesis of 19 behavior change models described in the literature and was developed because other existing models do not account for the full range of possible interventions for systematic health promotion intervention planning (Atkins and Michie, 2015). The BCW sits around the COM-B model and provides nine intervention functions. These are categories through which behavior can be changed: (i) training (e.g., feedback on behavior; self-monitoring of behavior, instruction on how to perform a behavior); (ii) enablement (e.g., social support, goal setting, action planning, coping planning, selfmonitoring of behavior); (iii) coercion (e.g., feedback on behavior, social comparison); (iv) education (e.g., information about health consequences, feedback on behavior; prompts, cues; self-monitoring of behavior); (v) environmental change; (vi) role models; (vii) persuasion (e.g., information about health consequences, feedback on behavior); (viii) incentive (e.g., feedback on behavior; self-monitoring of behavior); and (ix) restrictions.

Based on this framework, for example, interventions to enhance the perceived psychological capability of children with DCD could include training people involved in providing physical activity opportunities to enable greater differentiation. Likewise, to enhance the social opportunity for physical activity, interventions could consider facilitating family or matched peer-based physical activities as part of daily routines. Interventions might include restructuring the environment to facilitate failure-free physical activity opportunities, preferably from a young age, to enhance reflective and automatic motivation.

5 Conclusion

Although preschool-aged children with DCD may engage in similar levels of physical activity behavior, differences emerge from 6 years of age; this age may align with greater expectations but also increased self-evaluation. Due to the nature of DCD, children's reduced physical capability results in less participation in varied formal, recreational and skill-based activities, which limits their opportunity to enhance their physical capability. This may impact psychological capability, whereby children with DCD develop lower self-perceptions and lower self-efficacy, which feeds into this negative feedback loop that reduces their motivation to participate in physical activity. Barriers relating to physical and social opportunities to participate in physical activity have been identified that may result in negative attitudes and poor reflective and automatic motivation toward physical activity; however, there is some evidence that interventions, for example, using a Nintendo Wii or active video games, can enhance enjoyment, at least in the short-term. In the context of physical education, there is some indication that some children with DCD adopt coping strategies to minimize the psychological impact of compulsory participation in physical activity; however, the sustainability of adopting top-down cognitive strategies needs further investigation.

Author contributions

CP: conceptualization, data curation, formal analysis, investigation, methodology, project administration, resources, validation, visualization, writing – original draft, Writing – review and editing. KW: conceptualization, data curation, formal analysis, investigation, methodology, validation, visualization, writing – review and editing. NS: conceptualization, formal analysis, methodology, validation, writing – review and editing. JZ: conceptualization, formal analysis, methodology, validation, writing – review and editing. VR: formal analysis, validation, writing – review and editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

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EDITED BY
Catherine Purcell,
Cardiff University, United Kingdom

REVIEWED BY
Paola Chivers,
The University of Notre Dame Australia,
Australia
Kate Wilmut,
Oxford Brookes University, United Kingdom

*CORRESPONDENCE
Jaebum Park

☑ parkpe95@snu.ac.kr
Jae Kun Shim

☑ jkshim@umd.edu

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Anthropometric, physical activity, and psychological characteristics of Korean adults with and without developmental coordination disorder (DCD)

Min Joo Kim¹, Soo Mi Nam², Byeol Kim³, Ilhyeok Park⁴, Jaebum Park^{5*} and Jae Kun Shim^{1,6*}

¹Department of Mechanical Engineering, Kyung Hee University, Yongin-si, Republic of Korea, ²Division of Sports Science, Hanyang University, Ansan-si, Republic of Korea, ³Institute of Sport Science, Seoul National University, Seoul, Republic of Korea, ⁴Department of Physical Education, Seoul National University, Seoul, Republic of Korea, ⁵Department of Physical Education, and Advanced Institute of Convergence Science, Institute of Sport Science, Seoul National University, Seoul, Republic of Korea, ⁶Department of Kinesiology, and Neuroscience and Cognitive Science Program, The University of Maryland, College Park, MD, United States

Developmental Coordination Disorder (DCD), also known as Dyspraxia, is characterized by movement difficulties in individuals without discernible neurological disorders or identifiable medical conditions. Previous studies from various countries have highlighted disparities in anthropometric, physical activity, and psychological characteristics between children diagnosed with DCD and their typically developing (TD) peers. These differences are influenced by sociocultural norms and geographical locations. However, little attention has been given to scrutinizing analogous differences in adult populations, particularly within Republic of Korea. This study aims to address this knowledge gap by employing a battery of questionnaires to assess anthropometric, physical activity, and psychological traits in a cohort of 377 Korean adults, encompassing those with DCD (n = 54) alongside TD counterparts (n = 323). It was hypothesized that Korean adults with DCD would exhibit higher body mass index and lower ratings in physical activity and psychological characteristics than TD, consistent with the previous studies performed in other countries on children. The results showed no statistically significant differences between the DCD and TD groups in anthropometric characteristics such as weight (kg), height (cm), and body mass index. The prevalence of walking and biking for daily commuting in daily routines within Korean society might have contributed to the mitigation of anthropometric among individuals with/without DCD. Statistically significant differences were found in physical activity levels at work and recreational settings, as shown in physical activity scores and duration. The DCD group also displayed lower scores across several psychological characteristics, including exercise adherence, intrinsic motivation, self-efficacy, physical self-concept, exercise expectations, and intrinsic regulation. These findings underscore the necessity of incorporating sociocultural dynamics when investigating anthropometric, physical activity, and psychological characteristics in adults with DCD. Their perceived difficulties in

fine motor skills were also significantly poor than TD. Future research studies are warranted to elucidate the underlying mechanisms driving the observed patterns in this study, thus contributing to a more nuanced comprehension of how DCD manifests within specific sociocultural contexts.

KEYWORDS

developmental coordination disorder, adults, dyspraxia, physical activity, GPAQ, psychological characteristics

1 Introduction

Developmental Coordination Disorder (DCD) is a commonly diagnosed neurodevelopmental disorder, and the diagnostic and statistical manual of mental disorders-5 (DSM-5) is utilized for the clinical identification of DCD (American Psychiatric Association [APA], 2013). Notably, DCD affects motor skills and coordination, specifically called dyspraxia. These symptoms typically emerge in childhood, while the symptoms are partly extended to adulthood (American Psychiatric Association [APA], 2013). The prevalence of DCD has primarily been documented within the pediatric population, with a prevalence of 5-6% (Blank et al., 2019). However, prevalence rates among school-aged children exhibit significant variability influenced by factors such as country and ethnicity, spanning from 1.4 to 32.8% (Wright and Sugden, 1996; Lingam et al., 2009; Tseng et al., 2010; Valentini et al., 2015; De Milander et al., 2016; Amador-Ruiz et al., 2018). The prevalence of DCD within the adult population remains relatively underexplored. Nonetheless, some studies suggest that approximately 75% of adults who were diagnosed with DCD during childhood may continue to experience the condition into their adulthood years (Visser et al., 1998; Kirby et al., 2008). However, this prevalence might underestimate actual instances because of possible under-recognition, stemming from medical professionals needing more familiarity with the condition (Wilson et al., 2013). These differences are due to the methodology, cut-off criteria, assessment tool, and participant characteristics, as pointed out in previous studies (Valentini et al., 2015). Cultural background was a possible constraint when studying the prevalence of DCD (Tsiotra et al., 2006). Diagnosing DCD is a multidisciplinary effort involving collaboration between child psychiatrists, developmental pediatricians, child neurologists, and specialized therapists skilled in occupational or physical therapy. This approach ensures accurate diagnosis while adhering to the DSM-5 published by the American Psychiatric Association (American Psychiatric Association [APA], 2013).

Individuals contending with DCD encounter challenges in acquiring fundamental motor skills essential for holistic development. Growing research suggests that children with DCD are associated with difficulties in sensory processing and integration, which can hinder effective interaction with their surroundings (Goyen et al., 2011; Allen and Casey, 2017). Furthermore, children with DCD often exhibit diminished function in visual perception and motor planning (Goyen et al., 2011). Children with DCD manifest heightened tactile (Loh et al., 2011) and movement sensitivities, as well as tendencies toward under responsiveness and sensation seeking, compared to typically

developing individuals (TD) (Zwicker et al., 2010). Consequently, their ability to perform everyday tasks, such as catching a ball or writing, is significantly compromised, impacting overall functional autonomy (Engel-Yeger and Hanna Kasis, 2010). Additionally, a prevailing pattern indicates that DCD frequently co-occurs with other disorders, including Attention-Deficit/Hyperactivity Disorder (ADHD), Autism Spectrum Disorder (ASD), and specific learning disabilities (Edwards et al., 2011; Blank et al., 2012; Zwicker et al., 2013b). Research on adults with DCD has received relatively little attention compared to children. A total of 75% of children with DCD continue to experience DCD into adulthood (Kirby et al., 2008).

Developmental Coordination Disorder exerts a profound and enduring impact, resonating across the lifespan and encompassing a broad spectrum of domains, including social interactions, physical and mental well-being, educational and professional achievements, and overall health-related quality of life (Karras et al., 2019). These far-reaching effects underscore the significance of understanding DCD, especially in the context of adulthood. Therefore, even as adults, individuals with DCD tend to avoid tasks that require motor skills due to slow and clumsy movements, resulting in lower levels of participation in routine physical activities and decreased quality of life (Cousins and Smyth, 2003; Zwicker et al., 2013a; Engel-Yeger, 2020). Furthermore, the challenges they face extend to basic motor skills essential for daily life, encompassing tasks like organizing, planning, time management, handwriting, using technological devices such as smartphones, and driving (Losse et al., 1991; de Oliveira and Wann, 2011; Kirby et al., 2011). This enduring pattern highlights the need for comprehensive understanding and effective intervention strategies. In alignment with wellestablished correlations between mental wellbeing and selfesteem in adolescents (Harrowell et al., 2017) and adults with DCD exhibit an elevated vulnerability to mood disorders (Sigurdsson et al., 2002; Harris et al., 2015; Verlinden et al., 2023). Moreover, individuals with DCD may display physical signs, including a higher prevalence of overweight or obesity in both children (Hendrix et al., 2014; Zhu et al., 2014; Yam et al., 2022) and adults with DCD (Wagner et al., 2011; Verlinden et al., 2023). These multifaceted challenges emphasize the complex nature of DCD and the need for holistic research and support.

In the absence of a conclusive medical intervention or remedy for DCD, about 75% of those affected persist in experiencing DCD throughout adulthood (Kirby et al., 2008; Reid, 2020). Yet, comprehension regarding the shifts in the physical, psychological, and behavioral traits among adults with

DCD is minimal (Meachon et al., 2022). Considering the inherent connection between child development and cultural elements (e.g., traditions, religion, and family contexts) that shape experiences (Hedegaard, 2011), it becomes conceivable that some individuals with DCD might have unknowingly engaged in activities that inadvertently aided in improving their condition. The interplay of relationships in daily life, influenced by culture and socio-historical circumstances, shapes developmental progress (Nelson and Iwama, 2010; Hedegaard, 2011).

The belief that proficiency in fundamental motor skills correlates with enhanced socioeconomic conditions is widespread (Armstrong et al., 2011; Pienaar et al., 2015). For instance, East Asian countries, where the norm is to use chopsticks for eating, foster distinct fine motor skills encompassing active muscle control, focused concentration, and adept visual and motor coordination (Ohtoshi et al., 2008; Lee et al., 2019). Consequently, even individuals diagnosed with DCD, raised in East Asian cultural contexts, might showcase comparable or superior fine motor skills and hand-eye coordination compared to their non-East Asian TD peers (Chow et al., 2001). Another example pertains to physical inactivity, where exercise is prescribed as a treatment for children with DCD (Smits-Engelsman et al., 2021). The report from the World Health Organization (WHO) suggests that socioeconomic culture can influence each country's physical activity levels and their trends in difference countries (World Health Organization [WHO], 2022). This reality emphasizes how the vast array of cultural contexts in various nations might pose a challenge to established observations concerning the disparities in physical, psychological, and behavioral aspects between individuals with DCD and TD, necessitating the adaptation of diagnosis criteria and mechanisms accordingly.

Nonetheless, there is a need for more investigation into how cultural factors influence physical, psychological, and behavioral traits in DCD. The distinctive cultural aspects of Republic of Korea may contribute to the development of fine motor skills in individuals with DCD. However, considering the findings from the Global Status Report on Physical Activity 2022, which reported relatively high physical activity rates of 70% for adult males and 59% for adult females in Republic of Korea, it is noteworthy that a vast majority of Korean adolescents aged between 11 and 17 years (91% male and 97% female) are classified as physically inactive according to the World Health Organization's 2023 report. This suggests the potential presence of a significant motor skills gap between Korean individuals with DCD and their typically developing counterparts. When comparing Korea's rates of physical inactivity with those of neighboring and other developed countries, a distinct trend emerges. Republic of Korea exhibits markedly higher levels of physical inactivity compared to other East Asian nations. Specifically, among adolescents, China and Mongolia report male-to-female inactivity rates ranging from 80 to 89% and 74 to 83%, respectively. For adults, China and Mongolia register male-to-female inactivity rates spanning from 12 to 16% and 18 to 19%, respectively. In contrast, Korea's adult population demonstrates levels of inactivity more closely aligned with those found in developed Western countries like the United States (32-48%) and the United Kingdom (32-40%). However, these Western counterparts report significantly lower male-to-female inactivity rates among adolescents, ranging from 64 to 81% in the United States and from 75 to 85% in the United Kingdom (World Health Organization [WHO], 2023, Country Profile). These rates underscore the complexity of assessing fine motor skills and physical activity related to DCD on a global or regional scale. To enhance the systematic diagnosis of the DCD population, it is imperative to conduct country-specific investigations that account for sociocultural factors.

Considering these distinct Korean socioeconomic conditions, cultural attributes, and physical activity patterns, our research seeks to assess and compare the anthropometric, physical activity, and psychological characteristics of adults with DCD and TD peers. It was hypothesized that Korean adults with DCD would exhibit higher body mass index and lower ratings in physical activity and psychological characteristics than TD, consistent with the previous studies performed in other countries on children.

2 Materials and methods

2.1 Participants

This research study was approved by the Institutional Review Board at Kyung Hee University (IRB No. KHGIRB-21-342). A total of 540 university students, aged between 18 and 24 years, were recruited using email outreach, flyer distribution, online postings on university student community platforms, and cross-institutional word-of-mouth referrals. All participants provided informed written consent. After undergoing a comprehensive three-stage screening process guided by the exclusion criteria outlined in Section "2.3 Procedures," the final cohort consisted of 377 participants.

2.2 Assessment tools

Three distinct sets of questionnaires were administered with the specific objectives of (1) categorizing participants into DCD and TD groups, (2) assessing physical activity patterns, and (3) evaluating perceived psychological characteristics. The administration of all questionnaires was facilitated through Google Forms. This section includes brief descriptions of each questionnaire set.

2.2.1 Participant group classification

The Adult Developmental Co-ordination Disorder/Dyspraxia Checklist (ADC) designed to screen adults for DCD (Kirby et al., 2010) was administered to classify participants into DCD and TD groups. ADC consisted of three subparts: part A for as a child, part B for current symptoms, and part C for current symptoms manifested by others. Participants indicated the frequency of these difficulties by marking on a Likert scale with options "never" [1], "sometimes" [2], "often" [3], or "always" [4]. Scores for each scale were then summarized, where lower scores indicate better performance. The cut-off was set at 80 points. The ADC study conducted by Kirby et al. (2010) reported strong internal reliability for each of the three subparts: part A ($\alpha = 0.91$), part B ($\alpha = 0.87$), and part C ($\alpha = 0.90$) (Kirby et al., 2010). Their study also established the tool's validity by demonstrating significant correlations between ADC's subparts and the Handwriting Proficiency Screening Questionnaire (HPSQ; Rosenblum, 2008) (subpart A: r = 0.68; subpart B: r = 0.75; subpart

C: r = 0.71; p < 0.001) (Kirby et al., 2010). In order to ensure the applicability of the ADC in the Korean context, a collaborative effort was undertaken with the original authors of the ADC study and bilingual professors who were not connected to this study. This collaborative effort focused on the translation and adaptation of the questionnaire, taking into consideration the specific characteristics of the Korean population. To ensure the applicability of ADC in the Korean context, a collaborative effort was undertaken with the original authors, orchestrating the translation and adaptation of the questionnaire while staying attuned to the specific nuances of the Korean population. The questionnaire was translated through a collaborative effort involving our research team and external experts, following a structured three-stage process. The initial translation underwent stringent scrutiny under the evaluation of two bilingual Korean professors specializing in physical education and kinesiology and are currently based in the United States. Building on their valuable insights, a panel of four experts conducted a second revision. A third iteration of revision and review concluded the translation process, ultimately leading to the development of the Korean version of the ADC (Kim et al., 2023). Reliability and validity of the Korean version of the ADC have not been conducted.

The DSM-5 outlines four essential criteria for defining DCD, which encompass the presence of a motor coordination skills deficit (criterion A), difficulties in motor skills exhibited in daily activities and school settings (criterion B), the onset of symptoms during the early developmental period (criterion C), and the exclusion of other medical conditions or diagnoses (criterion D). To assess participants' motor skill development, we employed the ADC questionnaire, which is recognized for its ability to provide insights into criteria A, B, and C of the DSM-5 (Meachon et al., 2022). Additionally, we addressed criterion D, which relates to excluding DCD that could be better explained by another medical cause, by excluding ADC questionnaire responses from participants with other medical diagnoses.

2.2.2 Body mass index (BMI)

Body mass index (kg/m²) was computed based on participants' self-reported weight (in kilograms) and height (in centimeters), taking into account the constraints posed by the pandemic.

2.2.3 Assessment of physical activities

The assessment of physical activity characteristics was conducted using the Global Physical Activity Questionnaire (GPAQ), a tool developed by the World Health Organization to comprehensively analyze distinct physical activity patterns and engagement levels (Armstrong and Bull, 2006). The GPAQ is a widely used assessment tool for measuring physical activity levels (Cleland et al., 2014). The GPAQ comprises 16 questions spanning the domains of (1) occupational activity, (2) recreational activity, and (3) travel to and from places. These domains are further broken down into six sub-domains, including (1) vigorous work [e.g., lifting or carrying heavy objects (approximately 20 kg or more)], (2) moderate work [e.g., repetitive lifting and moving of light objects (less than approximately 20 kg)], (3) vigorous recreation [e.g., running], (4) moderate recreation [e.g., fast walking], (5) transport [e.g., riding a bike], and (6) sitting [e.g., sitting at a desk]. We employed the Korean version of GPAQ (Lee et al., 2020). The validity of the Korean GPAQ demonstrated a significant correlation with accelerometer data (r = 0.34, p < 0.01), and its reliability exhibited moderate agreement for each domain, with Cohens' kappa values ranging from 0.38 to 0.70 (Lee et al., 2020). Participants were asked to specify their weekly engagement frequency in each category, providing details on the average duration in hours and minutes, along with the intensity (moderate or vigorous) of the activity. Vigorous activity entailed high-intensity physical activities that substantially elevated heart rate or caused heavy breathing. In contrast, moderate activity refers to moderately intense physical activities, leading to a mild increase in heart rate or slightly heavier breathing. The intensity and duration of physical activity within each domain were used to calculate the overall physical activity volume (metabolic equivalent of task-minute per week, [MET-min/week]) using the formula provided (World Health Organization [WHO], 2012). The tool demonstrates acceptable reliability and validity, incorporating adaptations to suit diverse populations across various countries and cultures (Bull et al., 2009; Trinh et al., 2009; Herrmann et al., 2013), inclusive of the specific context of Republic of Korea (Lee et al., 2020).

2.2.4 Assessment of psychological characteristics

A total of eight questionnaires were employed to assess participants' psychological characteristics across domains such as exercise adherence, intrinsic motivation, self-efficacy, physical self-concept, and intrinsic regulation, as summarized in **Table 1**. All questionnaires were completed using a Likert scale, and the scores were calculated as the average of the responses.

Exercise adherence pertains to consistent engagement in exercise activities and is often evaluated based on exercise frequency, intensity, and duration (Dishman, 1994). In this study, exercise adherence was gauged through the utilization of the Physical Activity Scale (PAS), Exercise Adherence Questionnaire (EAQ), and the Achievement Goal Questionnaire-Physical Education (AGQ-PE). The PAS was invented to assess their perceived exercise affecting physical activity adherence (Armitage and Sprigg, 2010), and the Korean version of the PAS comprising four questions with a 5-point Likert scale was modified and validated for the Korean university students (Park and Yoo, 2014). Its validity (r > 0.76) and reliability ($\alpha = 0.80$) of the Korean version were reported (Park and Yoo, 2014). In this study, we affirmed these findings with our calculated validity (KMO = 0.83) and reliability ($\alpha = 0.96$). The EAQ, introduced in 1991 to assess predisposing, enabling, and reinforcing factors affecting physical activity adherence, was validated for the Korean population (Oh et al., 2000). Its validity (KMO > 0.75) was high, and reliability ($\alpha > 0.63$) was significant (Oh et al., 2000). In this study, validity (KMO = 0.91) and reliability (α = 0.90) were calculated. Comprising 15 questions, the EAQ employs a 3-point Likert scale for responses. The total score is computed by summing all the assigned points (Corbin and Lindsey, 1994). The AGQ-PE encompasses 31 items and utilizes a 7-point Likert scale, ranging from 1 (not at all true for me) to 7 (very true for me). The questionnaire covers eight psychological traits: performance-approach goals, masteryapproach goals, performance-avoidance goals, mastery-avoidance goals, social responsibility goals, social relationship goals, and effort and persistence. Our choice of this questionnaire is underpinned by its established validity and reliability within the Korean context

TABLE 1 The list of questionnaires used to measure psychological characteristics.

Measurement category	Questionnaire	Number of questions	Reference scale
Exercise adherence	Physical activity scale (PAS)	4	5
	Exercise adherence questionnaire (EAQ)	15	3
	Achievement goal questionnaire-physical education (AGQ-PE)	4	7
Intrinsic motivation	Intrinsic motivation inventory (IMI)	5	7
Self-efficacy	Motivated strategies for learning questionnaire (MSLQ)	6	7
Physical self-concept	Physical self-description questionnaire (PSDQ)	40	6
Exercise expectations	Outcome expectations for exercise scale (OEE)	9	5
Intrinsic regulation	Behavioral regulation in exercise questionnaire-3 (BREQ-3)	4	5

(Park and Lee, 2010), allowing us to glean insights into individuals' exercise adherence attitudes amid the challenges posed by physical activities. As a result, our focus centered on the persistence domain extracted from the original questionnaire, and we have included the pertinent reference (Guan et al., 2006; Park and Lee, 2010). Its validity (KMO = 0.82) and reliability (α = 0.88) of the Korean version were reported (Park and Lee, 2010). Our assessment reported KMO = 0.81 for validity and α = 0.88 for reliability.

The Intrinsic Motivation Inventory (IMI) is a comprehensive measurement tool designed to evaluate an individual's experience with a specific activity. It has been utilized in numerous experiments exploring intrinsic motivation and self-regulation (Ryan, 1982; Plant and Ryan, 1985; McAuley et al., 1989). The IMI encompasses four distinct subscale categories: interest/enjoyment, perceived competence, perceived choice, and pressure/tension. Considering that the interest/enjoyment subscale is recognized as a self-report measure of intrinsic motivation (McAuley et al., 1989), our study specifically employed the interest/enjoyment subscale and adapted the questions to pertain to exercise. The IMI employs a 7-point Likert scale, where higher scores indicate stronger agreement with the posed questions. The validity (factor loading > 0.45) and reliability ($\alpha >$ 0.74) of the Korean version were reported (Um and Kim, 2003). In this study, validity (KMO = 0.94) and reliability ($\alpha = 0.94$) were calculated.

For the assessment of self-efficacy, which signifies an individual's confidence in their ability to perform a task, the Motivated Strategies for Learning Questionnaire (MSLQ) was employed. The MSLQ is a self-report tool crafted to evaluate college students' motivational orientations and their utilization of diverse learning strategies within university courses. Comprising two distinct sections, the MSLQ covers both motivation and learning strategies. The motivation section delves into students' objectives and the value they attribute to a particular course, their perceptions regarding their competence to excel in the course, as well as their level of test-related anxiety. Within this questionnaire, higher scores correlate with increased self-efficacy levels (Pintrich et al., 1991). Our study utilized the Korean version of MSLQ, which had been modified and validated for the Korean population (Jung and Park, 2013). The validity (GFI = 0.90) of the Korean version was reported (Park and Lee, 2012). In this study, validity (KMO = 0.91) and reliability (α = 0.96) were calculated.

The Physical Self-Description Questionnaire (PSDQ) is a multidimensional instrument crafted to assess physical self-concept across 11 distinct scales: strength, body fat, activity, endurance/fitness, sports competence, coordination, health, appearance, flexibility, global physical self-concept, and global esteem, comprising a total of 70 items (Marsh, 1996). Our study employed a Korean version of the PSDQ, which entails 40 items instead of the original 70 items (Kim, 2001). Its validity (factor loading >0.53) and reliability (r > 0.74) of the Korean version were reported (Kim, 2001). This study calculated validity (KMO = 0.92) and reliability ($\alpha = 0.95$).

The Outcome Expectations for Exercise Scales (OEE) draw from Bandura's self-efficacy theory, assessing an individual's convictions regarding the anticipated results of engaging in a particular behavior. This measurement is founded on the premise that outcome expectations significantly impact exercise behavior among adults (Resnick et al., 2000). Comprising a 9-item 5-point Likert scale, the OEE scale encompasses ratings from 1, representing low outcome expectations for exercise, to 5, signifying strong outcome expectations for exercise and physical activity. All items of the scale were employed in our study. The Korean version of OEE, which has been reported to have validity (factor loading >0.53) and reliability (r > 0.74) (Kim, 2001), was employed in this study. For our research, we conducted validity (KMO = 0.89) and reliability ($\alpha = 0.87$) calculations.

The Behavioral Regulation in Exercise Questionnaire-3 (BREQ-3) is a widely recognized assessment tool that encompasses a range of exercise motivation types, which include motivation, external regulation, introjected regulation, identified regulation, integrated regulation, and intrinsic motivation (Markland and Tobin, 2004; Wilson et al., 2007). In our investigation, our focus was on evaluating intrinsic regulation. Therefore, we selected a specific subset of questions that pertain to intrinsic motivation, which signifies engagement in an activity driven by its inherent enjoyment and satisfaction. This subset comprises four items. The Korean version of this particular subset has previously established its validity (factor loading >0.68) and reliability (r>0.76) (Lim and Hu, 2011). For our current study, we performed calculations for both validity (KMO = 0.86) and reliability (α = 0.96).

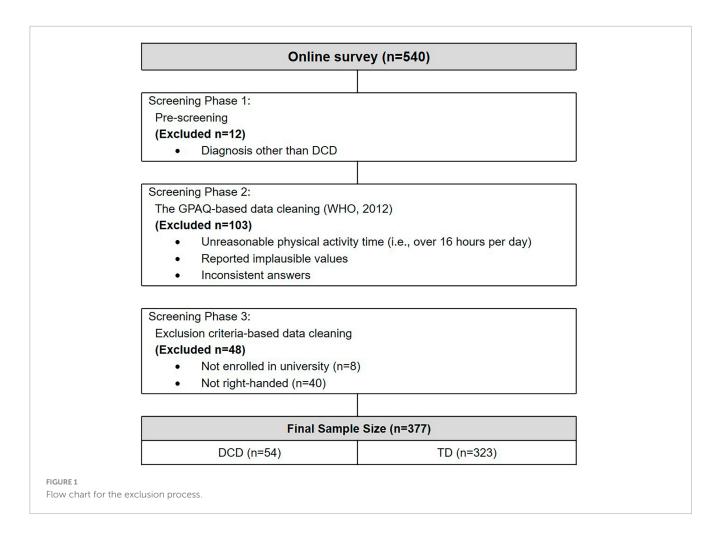
2.3 Procedures

After receiving informed consent from each participant, individuals in the DCD group scored 80 points or higher on the ADC. Participants also completed online surveys to provide their self-reported physical activity levels and psychological characteristics, elucidated in section "2.2 Assessment tools".

Our research team employed a 3-stage screening procedure. The study was conducted using an online survey methodology, initially collecting responses from 540 participants. The first phase of our data screening involved pre-screening to eliminate individuals with diagnoses other than DCD from ADC questionnaire. A total of 12 participants were excluded from the study (Figure 1). This exclusion comprised 10 participants with ADHD, 1 with dyspraxia, and 1 with dysthymic disorder. A recent international recommendation (Blank et al., 2019) states that DCD and dyspraxia are not the same, so we excluded individuals with other diagnoses. The subsequent screening phase encompassed the GPAQ data cleaning following its manual (World Health Organization [WHO], 2012). This process identified instances of unreasonable physical activity time (e.g., exceeding 16 h per day), implausible values, and conflicting responses, leading to the removal of 103 participants. These exclusions adhered to predefined criteria aimed at ensuring the accuracy and reliability of the physical activity data. In the last phase of the exclusion process, only those currently enrolled at a university and were right-handed were retained for the study. This criterion was informed by research on handedness, which revealed that left-handed individuals may face difficulties in their everyday experiences (Thomas et al., 2019). This methodological approach was undertaken to facilitate a nuanced comparative analysis amongst subjects with similar psychological maturation and socioeconomic standing levels. The participants who cleared all three stages of the screening procedure underwent an assessment using the ADC to classify them into either the DCD or TD group.

2.4 Statistics

We conducted descriptive statistics for the mean (M) and standard deviation (SD) of demographic and anthropometric features (i.e., age, height, weight, and BMI) and those variables related to ADC (i.e., total score, part A, B, and C, and subset score of fine motor skills). To test the assumptions for parametric statistics, we conducted the Shapiro-Wilk and Levene's tests for normality and equal variance on the demographic and anthropometric features, variables related to ADC (i.e., total score, part A, B, and C, and subset score of fine motor skills), physical activity (GPAQ) and psychology (i.e., PAS, EAQ, AGQ-PE, IMI, MSLQ, PSDQ, OEE, and BREQ-3). After discovering that these assumptions were not met, we conducted a Yuen's t-test between the DCD and TD groups. It's worth noting that Yuen's test does not rely on the assumptions of normality and equal variance. All statistical analysis was conducted in R (Version 4.3.1). Our statistical significance level was set at 0.05 for all analyses.



3 Results

3.1 Participants' demographic and anthropometric characteristics

Among the initial 540 participants recruited for the study, 377 participants cleared all three stages of the screening procedures. This final group comprised 170 males (45.1%) and 207 females (54.9%). The average age of the participants was 20.98 years. Participants were categorized into two groups based on their total ADC scores: 54 participants with DCD (ADC score equal to or over 80) and 323 participants classified as TD (ADC score below 80) (Table 2). These numbers revealed a prevalence rate of 14% for DCD within this study. No statistically significant differences were found between the DCD and TD groups regarding age, height, weight, and BMI (Figures 2A-D). We also compared ADC scores, including their perceived fine motor skills with scores from ADC related to fine motor skills (ADC items: A-1, A-2, A-5, A-6, A-8, B-1, B-2, B-4, B-5, B-6, B-7, and C-6) (Figure 2E). We found that the DCD group showed significantly higher scores in ADC total, ADC parts A, B, and C, and subset scores related to fine motor skills (Table 2).

3.2 Assessment of physical activities

We employed the GPAQ as an assessment tool for evaluating physical activities (**Figure 3**). The result of the GPAQ total indicated a significant difference between DCD and TD groups $[t_{(97.16)} = 3.98; p < 0.001]$. The DCD group exhibited statistically lower physical activity levels than the TD group. Within the DCD group, there were significantly lower levels of the work-moderate $[t_{(226.53)} = 2.39; p = 0.017]$, recreation-vigorous $[t_{(89.27)} = 3.53; p < 0.001]$, and recreation-moderate domains $[t_{(66.97)} = 3.08; p < 0.001]$, except transport domain $[t_{(65.98)} = 0.17; p = 0.869]$. These results suggest that adults with DCD are less physically active in most domains. Further analysis of the time (minute per day) dedicated to physical activity from the GPAQ demonstrated a significant difference between the DCD and TD groups. When

converted to minutes, the results also show how much less adults with DCD move compared to the TD group. Specifically, the DCD group spent significantly less time in the work-moderate $[t_{(212.27)}=2.03;\ p=0.043]$, recreation-vigorous $[t_{(116.16)}=4.29;\ p<0.001]$, and recreation-moderate domains $[t_{(51.78)}=2.32;\ p=0.024]$ compared to the TD group. However, no significant differences were found in the transport $[t_{(53.98)}=-0.14;\ p=0.886]$ and the sitting $[t_{(43.06)}=-1.87;\ p=0.067]$ domains.

3.3 Assessment of psychological characteristics

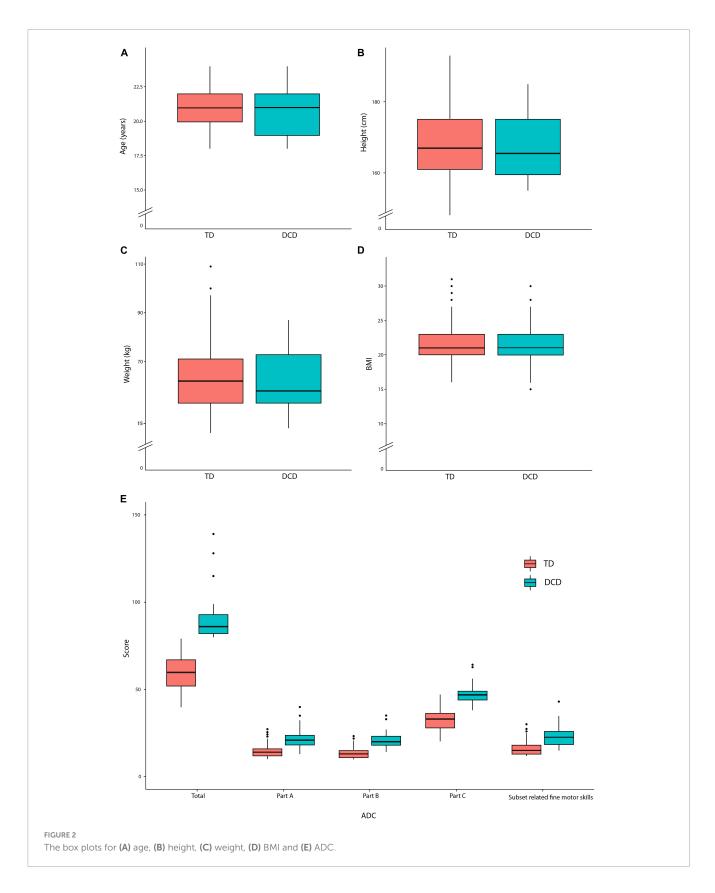
We employed several questionnaires to identify psychological characteristics (Table 1). We compared across all psychological characteristics-exercise adherence, intrinsic motivation, selfefficacy, physical self-concept, exercise expectation, and Intrinsic regulation. The DCD group exhibited statistically significantly lower scores across all psychological characteristics compared to the TD group. Exercise adherence of DCD group revealed significantly lower scores than TD in PAS $[t_{(53.52)} = 4.23;$ p < 0.001], EAQ [$t_{(41.87)} = 6.47$; p < 0.001], and AGQ-PE $[t_{(38.42)} = 2.46; p = 0.018]$, suggesting that the DCD group perceived that they have lack of willpower to continue exercising and did not think they have enough support of them surroundings or family and friends to continue their exercise (Figures 4A-C). Intrinsic motivation of the DCD group showed significantly lower scores than TD [$t_{(35.07)} = 5.00$; p < 0.001] (Figure 4D), which suggests that the DCD group was less motivated to participate in physical activities and less confident in performing motor skills. Self-efficacy scores of the DCD group were significantly smaller than TD [$t_{(43.66)} = 7.19$; p < 0.001] (**Figure 4E**), which suggests that the DCD group showed lower self-efficacy due to poor motor skills when participating in physical activities. Physical self-concept of the DCD group presented significantly lower scores than TD [$t_{(46.41)} = 7.29$; p < 0.001] (Figure 4F). As we mentioned in section "3.1 Participants' demographic and anthropometric characteristics," the DCD group was not different from the TD group regarding physical characteristics. However, the DCD group perceived themselves as less fit and less capable of performing

TABLE 2 The Yuen's t-test results in demographic and anthropometric characteristics of DCD and TD groups.

Items		Groups	[M (SD)]	t
		DCD	TD	
Age (years)		20.85 (1.65)	21.01 (1.67)	0.69
Height (cm)		166.92 (8.38)	167.85 (8.63)	0.80
Weight (kg)		61.87 (11.60)	62.44 (11.95)	0.01
BMI (kg/m ²)		22.09 (3.09)	21.83 (3.24)	-0.06
ADC	Total score	89.28 (11.20)	60.04 (9.69)	-20.76***
	Part A: as a child	21.60 (5.27)	14.20 (3.34)	-12.05***
	Part B: current perception of performance	20.50 (4.29)	13.40 (2.74)	-13.06***
	Part C: current feelings	47.15 (5.30)	32.44 (5.67)	-18.38***
	Score related fine motor skills	22.15 (3.62)	15.18 (5.89)	-8.40***

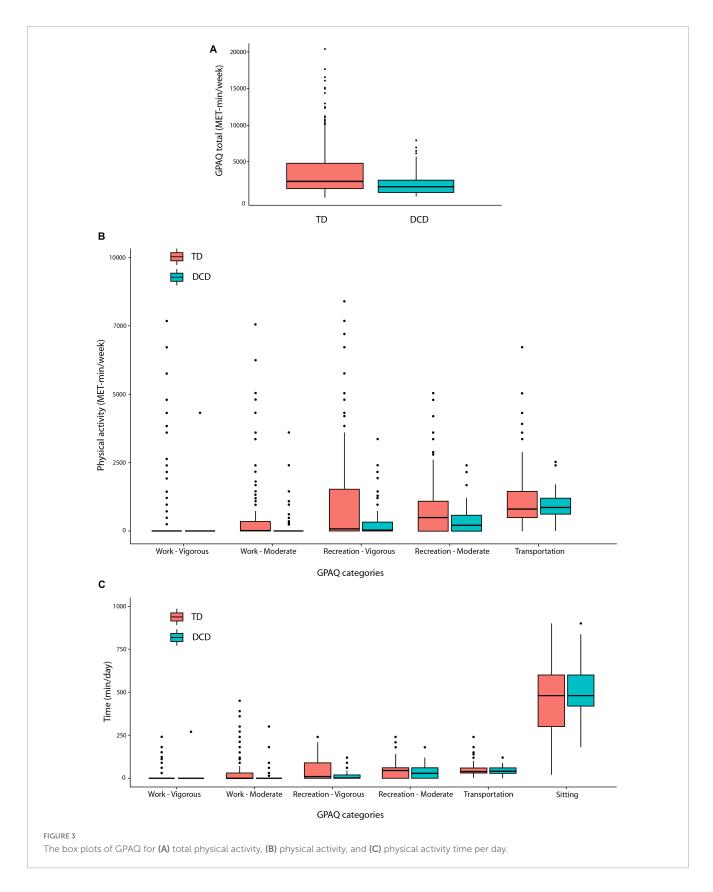
 $\label{eq:add} ADC: The adult developmental co-ordination \ disorder/dyspraxia\ checklist.$

^{***}p < 0.001.



sports. Exercise expectation of the DCD showed significantly lower scores than the TD group [$t_{(43.97)} = 3.62$; p < 0.001] (Figure 4G). Although not as much as the TD group, the DCD group generally expected exercise to promote their health and benefit from exercise.

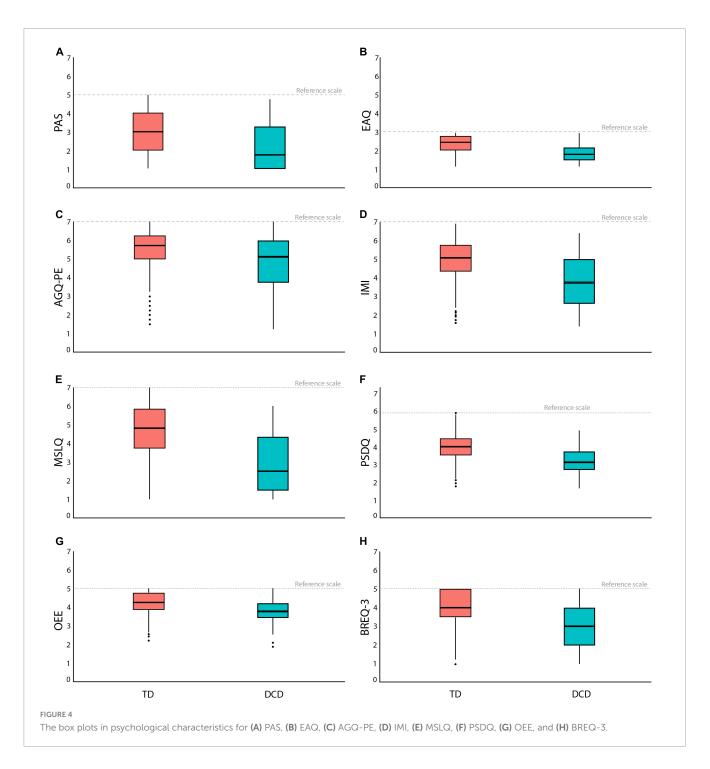
Intrinsic regulation of the DCD group revealed significantly lower scores than the TD [$t_{(39.34)} = 4.93$; p < 0.001] (**Figure 4H**), suggesting that the DCD group enjoyed less participating in or doing exercise.



4 Discussion

In this study, we found that despite exhibiting lower levels of physical activity and having lower levels of several

psychological factors related to exercise (e.g., exercise adherence, intrinsic motivation, self-efficacy, physical self-concept, exercise expectancy, and Intrinsic regulation), Korean adults with and without DCD did not differ significantly in terms of physical



characteristics like height, weight, or BMI. These findings align with previous research on adults with DCD, which showed reduced physical activity (Wilson et al., 2017) and low levels of psychological wellbeing (Tal-Saban et al., 2012; Purcell et al., 2015). However, the adults with DCD in our study who exhibited lower physical activity levels maintained BMIs within the normal range, similar to the typically developing population. This contrasts with earlier studies conducted in Canada (Cantell and Crawford, 2008) and Belgium (Verlinden et al., 2023), as well as with research on adolescents with DCD in Germany (Wagner et al., 2011), which reported higher BMIs among individuals with DCD.

The participants with DCD in this study were also found to have difficulty performing motor skills from childhood. Despite the noteworthy persistence of childhood DCD into adulthood, the existing body of research on DCD has predominantly focused on the pediatric population, with estimates ranging from 2% to 30% (Lingam et al., 2009; Blank et al., 2019; Kita et al., 2020). The limited adult DCD research available mirrors many traits commonly associated with childhood DCD, including heightened frustration, diminished competency, lower self-esteem, restricted engagement in daily activities (Jarus et al., 2011; Tal-Saban et al., 2014), and a compromised quality of life (Engel-Yeger, 2020), culminating in an overwhelming emotional burden (Cairney and Veldhuizen, 2013).

Kim et al. 10.3389/fnhum.2023.1280356

This congruence between childhood and adult DCD traits might have influenced the adoption of children's DCD diagnostic tools to adults. However, the broader context of human development underscores the significant influence of culture, leading to potential diversities in the observed physical, psychological, and behavioral characteristics within DCD adult populations across varying countries. Therefore, it is crucial to comprehensively understand the cultural associations underpinning DCD symptoms and then apply them to adult DCD diagnostic tools and interventions.

Culture stands as a potent yet often underestimated determinant of development and functioning (Matsumoto, 2001). Its influence extends beyond social dynamics, impacting cognitive processes and even biological responses (Schwartz et al., 2020) while also extending to motor skills (Cintas, 1995; Chow et al., 2001). The dynamic interplay between culture and context creates an environment where unique experiences shape psychological processes. For instance, a notable study discovered a substantial correlation between national culture and BMI across a cohort spanning 53 countries (Masood et al., 2019). The influence of culture extends to self-esteem as well. Research involving teenagers and young adults from 19 to 20 countries reveals that their self-esteem is not solely based on personal values but is shaped by the alignment with value priorities prevalent in their cultural surroundings (Becker et al., 2014). This cultural imprint even reverberates in the realm of physical activity levels, with cross-country disparities evident in a study encompassing 52 countries (Bann et al., 2019).

The assumption that elevated BMI in DCD has been differed by sample and other cultural factors. Like the present study, a longitudinal examination of the DCD population in Finland did not uncover statistically significant BMI differences compared to typically developing individuals (Tan et al., 2022). Conversely, previous literature shows that adults with DCD have a higher BMI (Cantell and Crawford, 2008; Verlinden et al., 2023), so it is important to consider the participant characteristics of these studies in interpreting the conflicting results. There is a possibility that other characteristics observed in individuals with DCD population, including enduring challenges in fine and gross motor skills (Cantell et al., 2003), writing (Barnett et al., 2011), time estimation (Tal Saban et al., 2014), learning to perform new tasks (de Oliveira and Wann, 2011), academic achievements/performance (Dewey et al., 2002; Alloway, 2007; Kirby et al., 2013; Harrowell et al., 2018), and mood disorder/anxiety (Cairney et al., 2010; Purcell et al., 2015; Omer et al., 2019), may not hold true across different countries. This observation holds substantial importance in terms of its potential impact on the accuracy of DCD diagnostic tests and the strategies for interventions. Considering that diverse cultural influences might lead to distinct patterns, the necessity of potentially modifying testing methods warrants careful consideration.

Although many studies have reported a higher prevalence of obesity among DCD population (Cantell and Crawford, 2008; Ferguson et al., 2015; Kumpulainen et al., 2016), our study did not reveal any significant statistical differences in height, weight, and BMI between adults with DCD and TD groups. The previous literature related to the population with DCD and their BMI and motor skills showed individuals with DCD with higher BMI and lower motor skills (Cantell and Crawford, 2008; Chivers et al., 2013). This divergence in findings could potentially be attributed to

an enduring cultural trend. A recent longitudinal study scrutinized the national obesity percentages across all genders and age groups in various regions of Republic of Korea between 2009 and 2018 (Nam et al., 2020). This investigation unveiled an escalating prevalence of obesity in both young men and women, particularly in the age range of 20-39 years (Nam et al., 2020). A plausible cultural explanation for this trend can be inferred from the statistics furnished by the Global Status Report on Physical Activity 2022: country profiles (World Health Organization [WHO], 2023), where physical inactivity in Republic of Korea was reported at 91% for male adolescents and 97% for females aged 11-17 years. Although these rates exhibit a slight reduction among adults aged 18 years and above, they remain notably high at 30% among males and 41% among females. Despite the characterization of Korean adults as physically inactive, BMI was in the normal range in all groups, and the results of this study may support the need to consider specificities such as race and culture. While our study did not identify discernible differences between the DCD and TD groups in terms of bicycle or walking use for commuting, notable contrasts emerged in occupational and leisure activity choices. The Korean adults with DCD appeared to gravitate toward pursuits that involve lower physical demands, as evidenced by fewer hours and reduced frequency of engagement. We hypothesize that when individuals find themselves in situations with limited options, the Korean DCD group tends to conform to prevailing norms. However, when presented with the choice to be less physically active, they readily opt for such alternatives. This result may be rooted in the context of our study participants, who were students living in an urban environment where walking and biking are customary modes of transportation, akin to many European countries. Consequently, they had to rely on relatively physically demanding modes of transportation. On the other hand, work is a realm where individuals can exercise independent decision-making. People can easily and independently identify jobs that entail less physical intensity and choose to avoid physically demanding roles. Given that all our participants were currently enrolled in school, it is plausible that their inclination toward occupations requiring greater physical exertion may have been limited, irrespective of their physical capabilities, motor competence, and confidence in engaging in physical activities. This aligns with the findings from our GPAQ analysis. Based on these observations, we recommend that adults with DCD diagnosis questionnaire consider emphasizing an individual's autonomy in actions. This would involve focusing on identifying actions driven by personal choice rather than actions influenced by cultural adaptation.

Psychological variables (Table 1) exhibited significantly lower values in the group of adults with DCD, consistent with findings from previous DCD studies conducted in various countries (Sigurdsson et al., 2002; Harris et al., 2015; Verlinden et al., 2023). Our study utilized a questionnaire to assess psychological states related to participation and persistence in exercise and physical activity. The results indicated a pronounced reluctance to engage in and sustain exercise, mirroring observations in previous research (Jarus et al., 2011; Engel-Yeger, 2020). This observed reluctance to participate in physical activities and maintain exercise regimens can be seen as an extension of a recurring characteristic among individuals with DCD, which begins in childhood. Children with DCD often shy away from various physical activities and physical

Kim et al. 10.3389/fnhum.2023.1280356

education classes due to their lower motor skill abilities. In our study, participants with DCD reported experiencing difficulties when performing daily activities and engaging in physical pursuits during their childhood years (see ADC Part A from Table 2). Additionally, their reduced motivation to participate in exercise, coupled with lower levels of self-efficacy and physical self-concept, may account for their decreased involvement in recreational physical activities compared to the TD population. Furthermore, our findings align with prior research, emphasizing that adults with DCD tend to exhibit lower overall psychological wellbeing compared to the general population.

The ADC questionnaire encompasses inquiries designed to assess difficulties in fine motor skills. These inquiries are rooted in extensive research highlighting challenges among adults with DCD regarding fine motor skills (Cantell et al., 2003), often translating to subpar handwriting performance (Barnett et al., 2011). Within the framework of the ADC questionnaire, we delved deeply into fine motor skills, subjecting questions related to challenges involving utensil uses for eating, handwriting, and grooming activities to meticulous statistical analysis. As mentioned in our Introduction, a question revolved around the influence of early exposure to chopstick usage in the Korean population on fine motor skill development. Yet, upon scrutinizing responses from both DCD and TD groups to fine motor skill questions within the ADC questionnaire, we discerned no statistically significant divergence in fine motor skills among Korean DCD and TD groups. This intriguing revelation suggests that cultures with a strong emphasis on honing fine motor skills may encounter difficulties in accurately identifying instances of DCD. This finding also hints at the possibility that our reported prevalence rate of 14% might be a conservative estimate, extending this trend to nations where chopstick use is prominent.

In conclusion, this study pioneers the investigation of anthropometric, physical activity, and psychological characteristics among Korean adults with DCD. While many of our findings align with global trends, deviations in specific aspects highlight the potential influence of culture on developmental trajectories, potentially leading to distinct patterns among adults with DCD. This underscores the value of comprehensive cross-cultural studies and subsequent adaptation of diagnostic mechanisms and intervention strategies. This study utilized perceived questionnaire responses to identify traits in adults with DCD and typically developing Korean adults. Our upcoming research phase will delve deeper into the social, physical, and neurological contributors and mechanisms underlying the differences between adults with and without DCD.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by the Institute of Review Board, Kyung Hee University. The studies were

conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

MK: Conceptualization, Data curation, Funding acquisition, Investigation, Methodology, Project administration, Resources, Visualization, Writing – original draft, Writing – review and editing, Formal analysis, Supervision. SN: Data curation, Formal analysis, Methodology, Writing – original draft, Writing – review and editing. BK: Writing – original draft, Writing – review and editing. IP: Data curation, Methodology, Writing – review and editing. JP: Conceptualization, Formal analysis, Funding acquisition, Investigation, Methodology, Resources, Software, Supervision, Writing – original draft, Writing – review and editing. JS: Conceptualization, Data curation, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Writing – original draft, Writing – review and editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

The authors declared that they were an editorial board member of Frontiers, at the time of submission. This had no impact on the peer review process and the final decision.

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EDITED BY Mariagiovanna Cantone, Gaspare Rodolico Hospital, Italy

REVIEWED BY
Judith Marguerite Gentle,
University of Surrey, United Kingdom
Cynthia Y. Hiraga,
São Paulo State University, Brazil

*CORRESPONDENCE
Rayan Falemban

☑ rafalemban@uqu.edu.sa;
☑ falembanra@cardiff.ac.uk

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Roadside experiences of parents of children with developmental coordination disorder and/or attention deficit hyperactivity disorder

Rayan Falemban^{1,2*}, Kate Wilmut³, Heather Hurst² and Catherine Purcell²

¹Department of Physical Therapy, Faculty of Applied Medical Sciences, Umm Al-Qura University,, Makkah, Saudi Arabia, ²School of Healthcare Sciences, College of Biomedical and Life Sciences, Cardiff University, Cardiff, United Kingdom, ³Centre for Psychological Research, Faculty of Health and Life Sciences, Oxford Brookes University, Oxford, United Kingdom

Introduction: Pedestrians are a vulnerable group at the roadside and previous research has identified that children with DCD and ADHD are at a heightened risk of pedestrian injuries. Despite this, limited research has explored parental perspectives of the pedestrian risks faced by children with DCD and/or ADHD. Understanding parents' perspectives provides a unique insight into the challenges children face every day and the concerns that parents perceive regarding their children's safety as pedestrians. Therefore, the aim of this study was to explore parents' perspectives of the pedestrian risks faced by their children with DCD and/or ADHD.

Methods: Semi-structured interviews were conducted with 14 parents of primary school and early secondary school aged children with age range 7–17. The participants were divided into three groups based on their children's conditions: DCD group (10-17years, n=3), ADHD group (7-13years, n=5), and co-occurring group (7-16years, n=6). All parents confirmed an existing diagnosis and completed the SNAP-IV and DCDQ as screening tools. The interviews explored parents' perspectives regarding their children's pedestrian behaviors, parents' concerns and preventative measures taken to improve the pedestrian safety of their children with DCD and/or ADHD. Reflexive thematic analysis was undertaken to analyze the interviews, from which three themes were developed.

Results: The first theme related to the challenges experienced by children at the roadside; parents emphasized the significance of structured and controlled pedestrian crossing sites, underlining their preference for designated crossings as safer options due to their heightened perceptions of risk associated with other road-crossing locations. The second theme: parental concerns and influences on children's road safety referred to their children's performance and safety at the roadside, leading to increased monitoring and a more protective approach to road crossing. The third theme: road safety education related to various strategies parents implemented to mitigate risks, while balancing independence and prioritizing their safety.

Discussion: While there were commonalities in the challenges faced by children with DCD and/or ADHD at the roadside, there were also notable differences. Parents of children with DCD discussed challenges with spatial awareness and motor skills, whereas parents of children with ADHD discussed challenges with impulsivity and inattention. Parents of children with co-occurring DCD and ADHD described a complex interplay of these challenges. It is evident from the interviews that children with DCD and/or ADHD require a distinct approach to develop their pedestrian skills effectively and parents reported specific strategies they used to address the

risks associated with their children's roadside behavior. Promoting pedestrian safety for children with DCD and/or ADHD necessitates collaboration among parents, schools and local authorities to implement comprehensive measures ensuring their safety. These findings contribute to understanding parental experiences and needs, providing valuable guidance for targeted interventions and policies to enhance the road safety of children with DCD and/or ADHD.

KEYWORDS

pedestrians, attention deficit hyperactivity disorder, developmental coordination disorder, road crossing, risky behavior, child safety, executive function, parental concerns

Introduction

The ability to move around one's community and from one location to another by any mode of transportation such as walking, cycling, driving and public transport is the definition of community mobility (Scott and Tulloch, 2021). Community mobility is an integral occupational enabler for individuals across the lifespan which supports well-being and the participation in meaningful occupations including, but not limited to, education, social participation and leisure activities (Stav, 2014; Scott and Tulloch, 2021). Independent community mobility, particularly for children, plays a crucial role in their health and physical, social and mental development (Shaw et al., 2015). As children mature, their desire for independence grows, prompting them to seek autonomy in their mobility choices. Previous research has studied the growing desire for independent mobility for children across 14 different countries (Shaw et al., 2015). By age 11 years, most children in surveyed nations could cross main roads unaccompanied, by age 12 years, a majority had the freedom to travel within walking distance alone and by age 13 years, could navigate their way home from school independently or utilize local bus services (Shaw et al., 2015). Despite the importance of community mobility, approximately 1.35 million people die every year due to preventable Road Traffic Accidents (RTA) and the World Health Organization (2018) reports that road traffic accidents are the leading cause of death for children worldwide. Thus, moving around communities can be a hazardous activity especially for groups that are vulnerable at the roadside, such as child pedestrians (Tapiro et al., 2014). In the United Kingdom, the daily average of 1 death and 10 serious injuries including children and adults has remained relatively unchanged for more than 15 years and the overall estimated cost of road traffic accidents in Great Britain is £12 billion annually (Department for Transport, 2019, 2020). Thus, safe and accessible community mobility is crucial for individuals' well-being and meaningful engagement in activities, yet the persistent risks and high social and economic costs associated with road traffic accidents emphasizes the urgent need for effective measures to prioritize road safety.

Furthermore, previous studies have identified that children with Developmental Coordination Disorder (DCD) and/or Attention Deficit Hyperactivity Disorder (ADHD) are at additional risk of pedestrian injuries (Wilmut and Purcell, 2021; Tabibi et al., 2022). ADHD is a neurodevelopmental disorder characterized by inattention, hyperactivity and impulsivity beyond developmental norms that negatively impacts activities of daily living (Ramos-Quiroga et al.,

2009). Similarly, DCD is a neurodevelopmental disorder marked by significant motor coordination impairments, adversely affecting daily activities (Kirby et al., 2013). While ADHD affects 5-7% of children globally (Abdelnour et al., 2022), the worldwide prevalence of DCD is estimated to be 5% (Blank et al., 2019). A significant co-occurrence rate exists between these two disorders, with estimates suggesting a co-occurrence of 50%, which underscores the need for investigating these disorders separately and together in relation to pedestrian safety (Goulardins et al., 2015). Navigating busy roads presents a unique challenge for children with DCD and/or ADHD. Research reveals a significantly elevated risk of pedestrian injuries for these populations compared to typically developing children (Wilmut and Purcell, 2021; Tabibi et al., 2022). This heightened vulnerability can be attributed to several key factors associated with each disorder. The characteristics associated with ADHD, including reduced attention, impulsive behaviors and hyperactivity, can have a negative influence on pedestrian performance (Wilmut and Purcell, 2020; Tabibi et al., 2022). Inattention and hyperactivity are suggested to be associated with poor timing when deciding to cross (Parr et al., 2021), while impulsive behaviors could lead to unsafe road-crossing decisions (Tabibi et al., 2022). Similarly, DCD, characterized by motor coordination impairments, presents distinct challenges for safe pedestrian behavior (Kirby et al., 2013). Deficits in spatial awareness, visual processing such as looming sensitivity, which is the ability to perceive and respond to approaching objects or vehicles, and visualmotor ability can hinder their ability to navigate complex traffic situations effectively (Purcell et al., 2012, 2017). Children with DCD may struggle to accurately judge distances and gaps between vehicles or execute coordinated movements quickly and smoothly when crossing roads leading to an increased risk of pedestrian injuries (Kirby et al., 2013; Purcell et al., 2017). For example, Purcell et al. (2011) found that poor perceptual-motor coupling in DCD can impact selecting safe temporal crossing gaps leading to inadequate crossing decisions and increased risk of injury. Additionally, children with DCD were found to have poor visual-motor abilities leading to reduced sensitivity in identifying approaching vehicles and inadequate road crossing decision-making, contributing to a potential increased vulnerability to road traffic injuries (Purcell et al., 2012, 2017). When these challenges associated with DCD combine with the inattention, impulsivity and hyperactivity observed in ADHD, the vulnerability to pedestrian accidents can further increase, potentially leading to more frequent and severe road traffic injuries (Wilmut and Purcell, 2020). In relation to ADHD, Clancy et al. (2006) found that the risk of road

traffic injuries in adolescents could be attributed to inattention, although a study conducted by Stavrinos et al. (2011) found that executive dysfunction may be the primary underlying factor for the increased risk of pedestrian injuries in children with ADHD. Overall, while ongoing research continues to explore the underlying causes, there is a consensus that children with DCD and/or ADHD face an elevated risk of pedestrian road traffic accidents and injuries.

Despite the growing recognition of the risks associated with DCD and/or ADHD in relation to pedestrian safety, there is a dearth of knowledge regarding the experiences and perspectives of parents of children with these conditions in relation to pedestrian risks. Parents of these children can provide a unique perspective regarding the challenges faced in the context of road safety. Their close observation and intimate knowledge of their child's behavior and responses to the environment, uniquely position them to offer insights into the specific challenges faced by their children as pedestrians. While behavioral studies are essential, parental perspectives provide a contextual and nuanced understanding, shedding light on the practical implications of these challenges in real-world situations. However, few studies have highlighted parents' experiences of children with DCD and/or ADHD at the roadside. Brook and Boaz (2006) identified, through a questionnaire, that parents of adolescents, aged 16-17 years, with ADHD were more concerned about their child's involvement in roadside accidents compared to a typically developing control group. To prevent accidents, parents suggested repeated discussions about risks, increased supervision, avoidance of dangerous play and use of medication to enhance attention and behavior (Brook and Boaz, 2006). Furthermore, Wilmut and Purcell (2020) found a relatively similar result in relation to parents of children with DCD using a quantitative parent-reported questionnaire. These parents reported that reduced attentiveness while crossing, often due to underlying perceptual difficulties, is a major concern which could manifest as a lack of confidence and increased risk-taking behavior (Wilmut and Purcell, 2020). Wilmut and Purcell (2020) further stated that the presence of ADHD characteristics in DCD was associated with further reductions in attention and increased perceived risk-taking behaviors. While these studies shed light on parents' experiences of children with DCD and/ or ADHD, there is a need for a comprehensive investigation into parental perspectives on pedestrian safety for children with DCD and/or ADHD. Therefore, the aim of this study was to explore parents' perspectives of children with DCD and/or ADHD to gain a deeper understanding of the elevated susceptibility to pedestrian injuries among these children.

Materials and methods

Research aim and questions

The aim of this study was to explore the perspectives of parents of children with DCD and/or ADHD to gather their experiences of pedestrian risks. The following research questions were formulated to fulfil this aim.

 What are the perspectives of parents of children with DCD and/ or ADHD in relation to their children's ability to execute a safe road crossing?

- What, if anything, are parents of children with DCD and/or ADHD concerned about regarding their children's pedestrian safety?
- How do parents of children with DCD and/or ADHD help to prevent or minimize their child's involvement in pedestrian injuries?

Reflexivity

This research adopted an interpretive, reflexive stance (Braun and Clarke, 2022). Reflexive thematic analysis is an interpretative approach to qualitative data analysis prioritizing researcher reflexivity and acknowledging the subjective nature of knowledge construction (Braun and Clarke, 2023). Therefore, the authors' backgrounds and experiences had a profound influence on the study's design and interpretation. To bring and own our perspectives, the first author maintained post-interview notes which played a pivotal role during the reflective analysis process in helping to understand the participants' responses in the context of personal experiences and potential biases while serving as a valuable reference point for deeper discussion and analysis. Furthermore, ongoing discussions with the co-authors enriched insights and ensured the management of authors perspectives within the research process. Collaboration and reflection were integral aspects of our research journey, influencing various stages from design to discussion and paper editing.

Recruitment

A purposive sampling strategy was employed to recruit parents of children with DCD and/or ADHD, this ensured participants had specific knowledge or experience relevant to the research question, enabling the collection of richer and more insightful data (Etikan et al., 2016). As such, we determined the sample composition reflecting our knowledge and understanding regarding participants' characteristics relevant to addressing the research aim using pre-defined inclusion criteria (Thomas, 2022). This assisted in generating intensive data leading to an in-depth understanding of the experiences of children with DCD and/or ADHD as pedestrians from their parent's perspectives.

Between January and July 2022, participants were recruited via two main avenues: social media platforms and organizations working with children with DCD or ADHD. Careful selection and display of recruitment posts on social media platforms is crucial, as inconsistent recruitment outcomes using these platforms have been reported (Topolovec-Vranic and Natarajan, 2016). Therefore, marketing headlines that trigger curiosity without compromising privacy were used to facilitate the recruitment through social media (Bender et al., 2017; Arigo et al., 2018). Furthermore, non-profit organizations, schools and institutions in the United Kingdom working with children with DCD or ADHD were utilized for recruitment. This recruitment avenue was expected to maximize access to the parents of children with DCD and/or ADHD. Ethical approval for the study was granted by the School of Healthcare Sciences Research Ethics Committee, Cardiff University. Prior to participating in the study, a pre-interview

package including a participant information sheet and two screening tools were sent to potential participants.

Measures

All participants provided written informed consent and completed the Developmental Coordination Disorder Questionnaire (DCDQ; Wilson et al., 2009) and the Swanson, Nolan, and Pelham Rating Scale (SNAP-IV; Hall et al., 2020). The DCDQ and SNAP-IV were scored according to established scoring guidelines provided by their respective authors. The DCDQ is a parent-report questionnaire designed to assess the presence of motor coordination difficulties in children (Wilson et al., 2007). It provides insights into a child's motor ability to identify potential signs of DCD. The questionnaire includes 15 items scored on a 5-point scale, total scores range from 15 to 75 (Wilson et al., 2007). A cut-off total score of 57 or below indicates a greater possibility of motor difficulties (Wilson et al., 2007).

The SNAP-IV is a parent-report measure of ADHD and contains 26 items scored on a 4-point scale, with higher scores indicating a greater possibility of ADHD (Gau et al., 2008; Hall et al., 2020). Typically, a SNAP-IV cutoff score above 1.2 suggests an increased probability of ADHD, while a score above 1.8 is considered indicative of clinically significant ADHD (Bussing et al., 2008). These measures were utilized as part of the process to screen for the presence of DCD and/or ADHD to confirm participant eligibility for inclusion in the study. The full inclusion and exclusion criteria are summarized in Table 1 and were established to confirm that participants met the diagnostic criteria outlined in the DSM-5 for DCD and ADHD (American Psychiatric Association, 2013). Following confirmation of the presence of DCD and/or ADHD based on the pre-interview package and parental reports, interviews were scheduled at a mutually convenient time and conducted online using Microsoft Teams.

Procedure

Inclusion criteria

This study utilized an online semi-structured interview approach to gather data from participants. Prior to the interview, participants

Exclusion criteria

and/or the internet

TABLE 1 Inclusion and exclusion criteria.

were able to communicate in English

were able to provide

informed consent

Parents of children with DCD and/or Parents of children who ADHD who: · were less than 7 years of age or · were aged 7-17 years greater than 17 years of age · had DCD and/or ADHD had no DCD and/or ADHD characteristics based on the DCDQ characteristics based on the DCDO and SNAP-IV and SNAP-IV • lived in the UK · were unable to provide • navigated the community with informed consent their children were unable to access to a computer

were instructed to select a distraction-free environment with a reliable internet connection to ensure a smooth and uninterrupted interview process. The interview duration was typically 60 min. The interview questions were developed through an iterative process prior to conducting the interviews. In the initial stage, questions were identified by the researchers based on a review of the relevant literature pertaining to children with DCD and/or ADHD as pedestrians. Drawing on the understanding gained from in-depth reading of the topic, the questions were, then, refined, and additional questions were incorporated after discussion between the authors. Piloting was conducted to ensure that the questions were clear, comprehensible and would effectively elicit the desired information from participants. The piloting phase involved three participants, similar to the target population in terms of their roles and experiences as parents. The feedback and insights gained from the piloting were instrumental in refining and finalizing the interview questions such as rewording, enhancing their appropriateness and effectiveness for capturing the unique perspectives of parents. The final phase involved a series of iterative revisions, facilitated through discussions between authors (RF and CP), until a consensus was reached on the final set of questions that were utilized as a guide for the interviews. Examples of the interview questions are provided below.

- I would like you to walk me through your normal day while you are walking around the community with your child? What does it look like?
- Can you tell me more about your child's behavior at the roadside and when crossing a road?
- How do you feel about your child's performance at the roadside and when crossing a road?

The full semi-structured interview questions can be found in the Supplementary Material.

Coding and data analysis

To ensure trustworthiness of findings, a reflexive approach was taken to analyze the data collected from the interviews. The reflexive approach of the Thematic Analysis (TA) focuses on identifying and interpreting patterns or themes within the data through a dynamic interplay between the researcher and the research material. To achieve this, all interviews were digitally audio-recorded and transcribed verbatim to ensure accuracy and completeness of the data. The built-in audio recording and transcription features of Microsoft Teams were utilized for this purpose. The main author (RF) actively listened to the recorded interviews to ensure that the transcription was accurate correcting any inaccuracies. This approach enabled a comprehensive analysis of the data and facilitated the identification of nuanced themes.

The TA was chosen as the method of analysis, which is a widely used approach for identifying patterns and themes within qualitative data (Braun and Clarke, 2023). However, it should be noted that TA is a flexible method that can result in inconsistent and less cohesive themes if not conducted rigorously (Holloway and Todres, 2003). To mitigate this, a reflexive TA was conducted following the six phases outlined by Braun and Clarke (2023), while acknowledging the plurality of TA and recognizing that this process is recursive rather than strictly linear (Braun and Clarke, 2023). The analysis commenced with data familiarization, a first step involving an in-depth review and immersion in the data. The familiarization phase included repeated

listening to the audio-recordings, reading and rereading interview transcripts to gain a comprehensive understanding, along with making initial notes and recording key ideas and emerging patterns. During the coding phase, RF systematically organized the data, assigning descriptive codes to encapsulate essential elements and meaningful information. Subsequently, RF engaged in reflective discussions with CP to ensure that RFs personal stance was consistently examined and refined in light of the emerging insights from the data. This step was conducted meticulously and recursively to ensure that the codes accurately reflected the nuances within the dataset. The subsequent third phase centered on identifying potential themes, where codes were grouped together to form overarching themes. These themes were refined by collating relevant data extracts associated with each potential theme, ensuring that they accurately represented the dataset as a whole. The creation of a thematic map followed during reviewing themes phase, allowing us to visualize the intricate connections between codes and themes. This visual representation facilitated discussions and further refinements of the themes through an iterative process, sometimes leading to extensive re-coding and re-mapping until a consensus was reached. In the fifth phase, we defined and named each theme, providing clear, descriptive explanations that enhanced our understanding of both the specificities within each theme and the broader narrative that emerged. The final stage is completion of a report which presents a coherent synthesis of the analyzed data, offering a professional and insightful representation of the themes derived from pedestrian experiences of parents of children with DCD and/or ADHD.

Findings

A total of 14 parents of children with DCD and/or ADHD were recruited and interviewed. This included parents of 5 children with ADHD, 3 children with DCD, and 6 children with both conditions. The majority of the participants children were male, accounting for 71.4% of the sample, while 28.6% were female. Participants' children demographics are summarized in Table 2. This demographic information presented in Table 2 provides a more detailed view of the children's age range and specific diagnosis.

Table 3 provides an overview of the participants, including pseudonyms used for their names and children's characteristics.

The collected data on parents' perspectives of children with DCD and/or ADHD at the roadside were analyzed, and three distinct themes were developed. The first theme explored parents' observations of their children's roadside behavior and road-crossing performance, revealing unique challenges related to these conditions. The second

TABLE 2 Demographics of the participants children.

Primary Condition	Gender of the child		Children's age range	Total number of	
	Male	Female		participants	
ADHD	3	2	7–13	5	
DCD	1	2	10-17	3	
DCD and ADHD	6	0	7–16	6	
Total	10	4	7–17	14	

theme examined parental perceptions, unveiling their concerns and emotions about their children's pedestrian safety. The third theme highlighted parents' resourcefulness in crafting survival strategies to safeguard their children.

These themes shed light on parents' perspectives of children with DCD and/or ADHD concerning pedestrian roadside safety. A summary of the generated themes and sub-themes are presented in Figure 1. These will now be discussed.

Theme 1: challenges experienced by children at the roadside

This theme highlights descriptions parents of children with DCD and/or ADHD gave in terms of their children's roadside behavior and road crossing performance. Although many participants experienced similar pedestrian environments, parents from each group reported different pedestrian behaviors which related to whether their child had DCD and/or ADHD.

Environmental context for children's pedestrian challenges

To provide context for a deeper understanding of their children's pedestrian challenges, we begin by exploring the environmental context in which these challenges unfold. Among the 14 participants in the study, 10 resided in urban areas, while the remaining 4 lived in rural or village settings. However, participants from rural areas lived in close proximity to a town and were exposed to similar transport infrastructure. One participant, Sophia from the ADHD group, described living in a:

"...village or close to countryside but there are transport infrastructure busses, roundabout, signalized crossing, zebra crossing, and alleyway" (13-years old).

All participants agreed that zebra crossings, signalized crossings and human controlled crossings are safer crossing sites when compared to midblock crossing sites.

"For signalized crossing, he likes to press the button and waits" (Emily, 7-years old, from the ADHD).

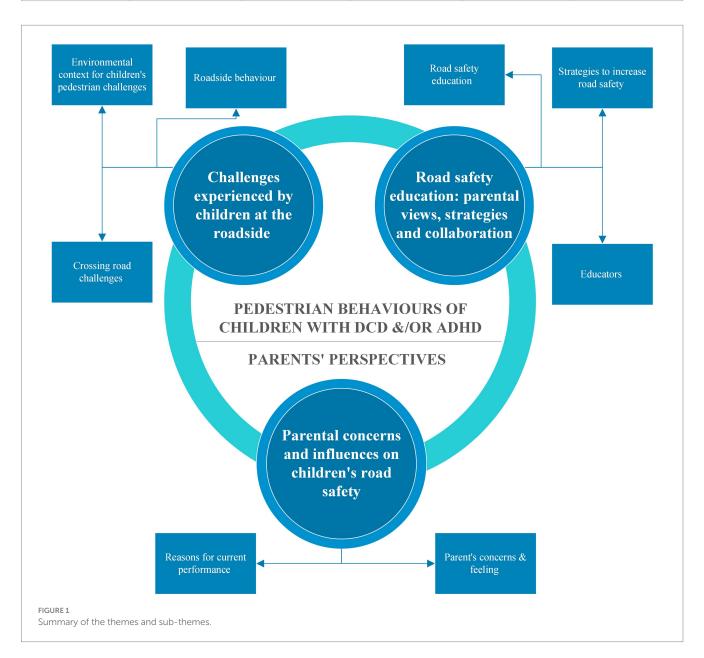
"those [referring to Zebra and signalized crossing], she's okay because all you have to do is wait for the cars to stop, do not you? And when you can clearly see that they have stopped, then you can go. So, the decision She's not having to judge" (Isabella, 17-years old, from the DCD group).

"He will wait by the lollipop lady and he knows because he's learning that rule" (Maryam, 10-years old, from the co-occurring group).

However, parents of children with ADHD expressed concern about their children's safety when crossing driveways due to previous near-miss incidents.

TABLE 3 Parents' information (pseudonyms used for names).

ADHD		DCD		ADHD + DCD	
Parents' names	Children info	Parents' names	Children info	Parents' names	Children info
1- Alex:	daughter 9 years old	1- Isabella:	daughter 17 years old	1- Maryam:	son 10 years old
2- Ava:	son 12 years old	2- Mia:	son10 years old	2- Charlotte:	son 16 years old
3- Olivia:	daughter 9 years old	3- Lily:	daughter 16 years old	3- Harper:	son 7 years old
4- Sophia:	son 13 years old			4- Amelia:	son 7 years old
5- Emily:	son 7 years old			5- Evelyn:	son 13 years old
				6- Harry:	son 8 years old



"The most important is the driveways as Emma runs across all those driveways." (Olivia, 9-years old, from the ADHD group).

that children with ADHD tend to exhibit risky behavior, such as standing at the edge of the pavement.

Parents in the ADHD group also reported that their children exhibit less dangerous behaviors when using zebra and signalized crossings as long as they are not distracted. However, parents reported

"He does stop, but what he does do is he stands at the very edge of kerb. So, he will really push the limit and be like, I am here." (Emily, 7-years old, from the ADHD group).

Conversely, parents reported that children with DCD showed a greater inclination to wait for others to cross with them at these crossing sites. Parents reported that they followed their parents, groups of pedestrians and the instructions of road crossing patrols.

"Because she's frightened of the traffic, she'll just follow other people [other pedestrians]" (Lily, 16-years old, from the DCD group).

According to parents, children with co-occurring DCD and ADHD also relied on others when using zebra crossings, signalized crossings and human controlled crossings, unless they were distracted by something on the other side of the road.

"He will wait by the lollipop lady and he knows because he's learning that rule, that he sits and waits there for me. So, he will never cross without me" (Maryam, 10-years old, from the co-occurring group).

In summary, parents highlighted similar pedestrian infrastructures in both urban and rural areas. However, they articulated a range of challenges experienced by their children with DCD and/or ADHD in relation to these infrastructures. While zebra crossings and signalized crossings were considered safer, concerns about driveways were common among parents of children with ADHD, highlighting potential attentional issues characteristic of ADHD. Children with ADHD tended to exhibit risky behaviors near pavements possibly due to difficulties associated with hyperactivity, whereas those with DCD showed a greater inclination to wait for other pedestrians or pedestrian signals leaving the crossing decision to other people or road architecture. This could indicate that children with DCD have less confidence in their perceptual and motor abilities. Children with co-occurring DCD and ADHD showed a combination of these behaviors such as relying on others for crossing and distracting easily.

Roadside behavior

Common roadside behaviors reported by parents across the three groups were difficulties when multitasking, such as talking and walking, which could exacerbate the problem and negatively affect their roadside safety. For example:

"When he is absorbed in what he is doing, e.g., talking to his brother, I need to grab his attention first by tapping his shoulder or the back of his neck, or I'll try and get near him and say, like, hey, John, just to try and cut through what's going on in his brain." (Emily, 7-years old, from the ADHD group).

"It's that concentration she cannot seem to multitask. She cannot talk and walk at the same time because it just takes her concentration away from what she's doing." (Lily, 16-years old, from the DCD group).

Furthermore, parents of children with ADHD described their children as very active, energetic and tending to run around, jump and engage in non-stop talking, making it difficult to focus on road safety. As a result, parents observed their children running or walking

without noticing the edge of the kerb, leading to walking or running on the road instead.

"But she's that energetic and she's that bouncy. She will run ahead and go straight across the driveway without even thinking that the pavements changed" (Olivia, 9-years old, from the ADHD group).

"They [John and his brothers] just like wander off the pavement into the road and start walking in the road instead of on the pavement. Especially John likes to walk right on the edge of the kerb." (Emily, 7-years old, from the ADHD group).

On the other hand, parents of children with DCD reported a lack of spatial awareness, which often resulted in them bumping into people and things. As parents report, they tend to rely on someone walking beside or in front of them. As a result, parents suggested that they lacked confidence in their children's ability for independent mobility and decision-making at the roadside. Examples from parents when discussing walking on the pavement include:

"She's always covered in bruises, where she's constantly bumping into things." (Lily, 16-years old, from the DCD group).

"She always tends to stay by the side of someone. She's never one to take the lead. If this only one person can fit through, she tends to stay back and follow." (Isabella, 17-years old, from the DCD group).

Parents of children with co-occurring DCD and ADHD reported a combination of these behavioral characteristics. While they were reported to have similar characteristics to the DCD group, including relying on others for decision making and exhibiting poor spatial awareness, leading to children bumping into people and things, those with co-occurring DCD and ADHD were described as often running, jumping and climbing. They were also observed to be unaware of the edge of the kerb, which increased their likelihood of walking on the road.

"So, I'm always reminding him to walk near the wall, not the edge of the road. All the time bumping into people. So, he's just got no concept of where his body is" (Maryam, 10-years old, from the co-occurring group).

"Because of the big thing with Malcom is his spatial awareness. He does not realize where his body is in the space. He is just sort of floating about and he would just not realize that the pavement had ended, and the road had start." (Amelia, 7-years old, from the co-occurring group).

This sub-theme highlighted distinct challenges faced by parents of children with DCD, ADHD and co-occurring DCD and ADHD regarding road behavior. Parents across the three groups commonly reported challenges with multitasking during roadside activities, which could worsen the situation and pose risks to roadside safety. Parents of children with ADHD noted their high activity levels, making them prone to running or walking on roads without noticing

hazards. Conversely, parents of children with DCD reported a lack of spatial awareness, leading to frequent collisions with objects/people. Those with co-occurring DCD and ADHD were reported by parents to display a combination of both behaviors, poor spatial awareness and high activity levels, emphasizing the complexity of pedestrian safety for these children.

Crossing road challenges

The behavior of children with DCD and/or ADHD when crossing roads was also reported by parents. Parents of children with ADHD discussed how their children tend to run or walk straight across the road without looking at oncoming vehicles or checking both ways. Parents believed that their attention is often directed toward their intended destination only, resulting in a disregard for the environmental cues and hazards in the immediate surrounding environment. This phenomenon was frequently referred to by parents as "tunnel vision."

"Not able to pay attention around about his surroundings, so he will literally just walk straight across the road without looking, he does not look" (Ava, 12-years old, from the ADHD group).

"When I went to pick him up from school, he ran into the road not looking." (Sophia, 13-years old, from the ADHD).

"She's just she tends to look at the point she wants to get to and it's almost like she gets tunnel vision. Nothing else is there. She just needs to get from where she is to that thing over there" (Olivia, 9-years old, from the ADHD group).

All parents of children with DCD, on the other hand, noticed their children move their head right and left as a visual scan before crossing but have difficulty in interpreting the visual cues to make an appropriate decision.

"She would turn her head like this. But she wasn't actually looking at the cars and making a judgment. She was doing the movement. And she would stand there, and she would do the head movement only." (Isabella, 17-years old, from the DCD group).

The parents further explained that they either rely on other pedestrians to make the decision or they make a random decision to cross the road, thereby increasing their vulnerability to accidents.

"She has not got the confidence and because she's frightened of the traffic, she'll just follow other people [other pedestrians], which sometimes is not a good thing because if they run out in front of something, she's trailing behind" (Lily, 16-years old, from the DCD group).

Parents reported observing their children either running across or looking down during road crossing. Parents suspect that children lack confidence in their decision-making ability and we assume that may contribute to the variation in their crossing styles. Moreover, they stated children with DCD lack the ability to judge the speed of

an approaching vehicle and cannot determine whether vehicles are far enough away to safely cross the road.

"She walks head down, she's like, I made my decision, and my head is down, and I am going. She does not run but she does walk very fast" (Isabella, 17-years old, from the DCD group).

"She cannot make that judgment. She cannot tell if that car is far enough away." (Lily, 16-years old, from the DCD group).

According to parents, children with both DCD and ADHD demonstrated relatively similar behaviors to the ADHD group and DCD group. Children may look both ways before crossing but face difficulties in processing the visual cues to make an appropriate decision.

"So, he was standing at the edge of the road, and he would turn his head. But he would not necessarily spot that the car was coming" (Maryam, 10-years old, from the co-occurring group).

Similar to the DCD group, parents of children with DCD and ADHD observe safe road crossing when among a group of pedestrians and tend to walk behind other people. Furthermore, children with DCD and ADHD were described as experiencing difficulties in judging the speed of approaching vehicles relative to their own speed, potentially leading to dangerous situations.

"I do not think he can judge how fast he's going. You know, if there's a car coming, if there's a car at the end of the road, he would not know that car was far enough away that you could cross." (Maryam, 10-years old, from the co-occurring group).

Parents also observed their children stopping in the middle of the road or expecting the car to stop for them, like at a zebra and signalized crossing.

"He does not really know how to react to traffic, so I'm trying to give an example of something that happened. We were crossing quite a busy road and as we were crossing the road, he stopped right in the middle of the road because there was a bus approaching. But it was a red light so the bus would have stopped but he stopped dead in the middle of the road" (Amelia, 7-years old, from the co-occurring group).

Parents of children with DCD and ADHD reported that their children might engage in impulsive behavior when crossing roads. For instance, they may exhibit a tendency to run across the road if they perceive something of interest on the other side or if they recognize someone they know.

"He might run across the road if he saw somebody he knew on the other side, or a dog that he wanted to speak to, he would not think it was a road." (Maryam, 10-years old, from the co-occurring group).

Overall, it is evident that visual-motor/attention challenges play a substantial role in the pedestrian safety of all children in the study. Whether due to issues related to attention or as described "tunnel vision" (in the case of ADHD), difficulties in processing visual cues (in

the case of DCD), or a combination of these factors (in the case of co-occurring ADHD and DCD). These challenges underscore the importance of addressing visual perception and attention in enhancing road safety for these children.

Theme 2: parental concerns and influences on children's road safety

Parental perceptions are critical aspects in understanding parents' perspectives regarding their children's road safety behavior and performance. This theme explores parents' concerns and feelings about their children's pedestrian safety and possible underlying causes of roadside performance through two subthemes.

Parent's concerns and feelings

The first subtheme revealed that parents of children with ADHD experience apprehension regarding their child's independent travel abilities, even when utilizing public transport from remote locations, such as busses.

"We've just been quite nervous about him doing that on his own and organizing himself to get on a bus that's 10 miles away to come home. Do not feel quite yet, he's ready to do that. So, I suppose we supervise him a lot and he does not really go anywhere on his own without us." (Sophia, 13-years old, from the ADHD).

These parents monitor their children closely and restrict them from going out unsupervised. They also frequently hold their child's hand, fearing potential traffic accidents.

"I probably held his hand a lot because I was very worried about him running off onto the road" (Sophia, 13-years old, from the ADHD).

Furthermore, the situation may be further complicated by the fact that the parent may be a single parent with the child and their siblings, making it even more challenging.

"I would assume that it is usually me with my three kids. So, it's not only me and John. So yeah, it it's kind of hard." (Emily, 7-years old, from the ADHD group).

Parents of children with DCD noted that road crossing may not be a priority initially, due to other pressing developmental issues, but as their children enter adolescence, the significance of safe road crossing becomes increasingly apparent. These Parents expressed concern about their children's ability to navigate roads safely.

"When she was very small, it wasn't something we thought about so much because of the range of problems that Claire had, it was not a top priority. We had other more pressing issues like skills, milestones that were late, that were more important than crossing the road. It really started to be an issue. I think when she got to be a teenager. So, by the time she was in secondary school, 12 could not cross the road and it wasn't even close to being able. When we attempted to teach her to cross the road. She wasn't even close to being able to do

it safely and to make safe decisions" (Isabella, 17-years old, from the DCD group).

"What happens as the children get older is that their independence is very restricted by this. Basically, a child that cannot cross the road cannot leave the house on their own. And you suddenly find that you have this child, who is 16, who is 5 feet tall who still needs their mum to take them." (Lily, 16-years old, from the DCD group).

These concerns can limit their children's independence leading to increased parental supervision when they wish to become independent. Moreover, parents stated driving children to school can hinder their development when they want their children to gain more road safety experience. Therefore, parental' concerns regarding their children's road safety are difficult when balancing children's independence with road safety. Based on insights shared by parents, effective strategies to teach their children how to cross roads safely while still allowing them to develop the independence they require for community mobility can facilitate this goal.

"I do not think it's helped because, we have had to take her back and forth to school every single day because of the distance of the school from where we live to where the school is and like I said, there's no way she could have walked safely back and forth. It's way too far and the roads are too busy, and I do not think it's helped her, and this is why I want to really encourage her to start going out with her friends now. So, because the more experience she's got on the road, the better it's going to be for her. She cannot live in a bubble with her parents walking behind her for the rest of her life." (Lily, 16-years old, from the DCD group).

Likewise, parents of children with co-occurring DCD and ADHD expressed concern and fear for their children's road safety. These concerns demonstrate the complexities and challenges of fostering independence in children while ensuring their safety. Parents recognized that their children's safety depends not only on their own actions but also on the actions of others, such as drivers and pedestrians.

"It's my biggest fear that Malcolm is going to get run over because it's so likely to happen. I can see it happening based on how I see him every single day near roads. As he gets older, and he becomes more independent, and he starts wanting to do more things independently, my fear grows." (Amelia, 7-years old, from the co-occurring group).

Additionally, parents were aware of the impact of their child's mood and fatigue levels on their judgment, making it essential to consider their emotional and mental well-being in addition to their physical safety.

"So generally, I'm quite nervous and scared, but some days he's good, really good and really receptive and really responsive. And then the other days are just because he gets fatigued in the afternoon because of how on-the-go it is in the afternoon, it's like a bit of a clouds formed his judgment and his mind. So, it depends on the time of day, and it depends on his mood." (Amelia, 7-years old, from the co-occurring group).

Overall, this sub-theme highlighted parents' concerns of children with DCD, ADHD and co-occurring DCD and ADHD in ensuring their child's safety during road crossing. Parents of children with ADHD expressed concern regarding their child's independent travel abilities, leading to increased parental supervision and a possible decrease in the opportunity for children to learn safe road crossing behaviors. Parents of children with DCD initially prioritized other developmental issues over road crossing but later express concerns about their child's ability to navigate roads safely. Parents of children with co-occurring DCD and ADHD also described their fear for their children's road safety, emphasizing the complexities of fostering independence while ensuring safety. Thus, all parents faced the demanding challenge of finding the balance between fostering their children's independence while prioritizing their pedestrian safety. The parents' expressions of fear and concern illustrate the importance of recognizing the complexities of the road crossing task and the need for effective strategies to teach children how to cross roads safely as they develop independence.

Reasons for current performance

Parents of children with ADHD perceive their children's poor performance as pedestrians to be linked to ADHD characteristics, which is affected by their mood and temperament. Specifically, parents reported that during episodes of bad mood or inability to self-regulate, their children's impulsiveness and inattention of their surroundings may lead to unsafe crossing behaviors, such as darting across roads without checking for oncoming traffic.

"Worse during the bad temperament, more like oppositional defiant disorder, leading to not stop and shoot across roads and I think he did not hear anything around." (Ava, 12-years old, from the ADHD group).

However, during periods of good mood, these parents reported that their child's road safety behavior could be adequate, highlighting the critical role of mood fluctuations in performance. Despite these challenges, parents expressed optimism about their children's ability to develop strategies as they grow older leading to improved pedestrian performance.

"If she's in a good frame of mind, she's like a professor of it. She will tell you exactly how you should cross the road. But if she's in a bit of a bad mood or whatever, it's like fight or flight response, and the matter of fact is that she can tell me perfectly she will just go." (Olivia, 9-years old, from the ADHD group).

Similarly, all parents of children with DCD believed that their child's poor pedestrian performance is related to their inability to judge distance and speeds accurately. They also reported that their children may struggle with spatial awareness, which can make it difficult to navigate around obstacles and people on the pavement. Additionally, fatigue, lack of confidence and forgetfulness can further impact their children's judgment and spatial awareness.

"She [referring to Isebella's daughter] cannot always tell where other people are properly. So, walking into people is a real problem. So, she constantly has this fear that people are gonna walk into her because she cannot tell where she is. So, she does not know where they are." (Isabella, 17-years old, from the DCD group).

"My feeling is that she cannot judge the speed of the car. So, you know if you or I look, you can tell and you learn through experience how fast the car is going. And is that car far enough, and have I got time to cross?" (Isabella, 17-years old, from the DCD group).

For parents of children with both DCD and ADHD, the DCD and ADHD are perceived to be contributing factors to their children's poor performance as pedestrians. They expressed concern about their child's lack of focus and impulse control, which can lead to unsafe behavior on the road. Moreover, parents acknowledged that their child's spatial awareness, concentration, attention difficulties and motor skills may affect their ability to judge distances and navigate their environment safely.

"If he's tired, if he's worried, you know? So, if he's anxious about something, then he's more likely to be dysregulated on edge and more bouncy, and all over the place, as we say" (Harry, 8-years old, from the co-occurring group).

"I think it's both [referring to ADHD and DCD]. So, the impulse bit is obviously ADHD, but because he has no idea of what his body is doing, you know he cannot stay upright. He does not know where he is in space, which is more of the DCD I think... When he's focused, he can cross the road safely. But you never know which day he's going to be focused, or you know which minute he'll be focused, and which minute he will not." (Maryam, 10-years old, from the co-occurring group).

Despite these challenges, some parents remained hopeful that with time and support, their child can improve their pedestrian performance. However, they currently would not feel comfortable allowing their child to walk to school independently or with friends, as they believe it would be unsafe.

"So next year, when he goes to secondary school, I will not let him walk to school on his own or even with a group of friends because I would not put them in that position where they have to keep him safe." (Maryam, 10-years old, from the co-occurring group).

In general, parents of children with ADHD link poor pedestrian performance to ADHD traits such as impulsivity and inattention which is affected by mood fluctuations. For those with DCD, spatial awareness, motor skills and fatigue pose challenges for appropriate pedestrian behaviors. Children with both conditions face a combination of limitations related to concentration, attention difficulties, motor skills and spatial awareness, leading to significant parental concerns about their road safety.

Theme 3: road safety education: parental views, strategies and collaboration

This theme explores parents' perspectives of current road safety education and the various strategies they developed themselves to ensure their children's safety. Despite concerns, parents have developed invaluable pedestrian safety strategies

that can be adopted to enhance the practical aspects of road safety education.

Road safety education

Parents of children with DCD and/or ADHD discussed their children's road safety education and also suggested some elements to facilitate the effectiveness of the education. Parents of children with ADHD believed that while their child received some road safety education, it may not be sufficient. They suggested that stories, which provide an emotional connection, may have a greater impact on their child than generic videos.

"I know they had that Bobby Colleran [campaign] that Slow Down for Bobby [Bobby Colleran is a local road safety campaign in the UK aimed at promoting safe travel to schools]. The family go to her school, and they were there a couple of weeks ago, going through everything again and sharing the books and we have got the books at home as well. We've read them... So, she's [Olivia is] quite compassionate, so that will make her think more than just watching a video of someone that she does not know." (Olivia, 9-years old, from the ADHD group).

"You know the old adverts that used to be on the television about if you did not buckle in... we had to show her things like that to make her understand the implications of those choices. From that point onwards, she wore a seat belt. No question. She gets straight in and buckles up." (Olivia, 9-years old, from the ADHD group).

Moreover, parents stressed the importance of teaching road safety in a way that their child can understand, with step-by-step instructions and minimal distractions. They reported that one-to-one or small group sessions can be most effective.

"Instructions need to be step by step, otherwise, his brain gets overloaded." (Emily, 7-years old, from the ADHD group).

Parents of children with DCD stated that they found ways to adapt to their children's needs. They were involved in the school's Kerbcraft program to reinforce road safety practices and took the initiative to teach their child about road safety during outings.

"Well, in school they do the same thing. They do Kerbcraft once from five or 6 years of age where they take them out in the community, and they cross busy roads." (Lily, 16-years old, from the DCD group).

They also mentioned a transition training program preparing their child for the transition from primary to secondary school, where road safety awareness is essential. Additionally, they emphasized the active role of parents in teaching their children road safety.

"They do a transition at the last year. So, when they go from year six to year seven and the road safety officer will go into the primary schools and speak to them and tell them, think how you are going to get to your new school when you start in September, plan your route, look for the safest route, do not look for the shortest route, it's gotta be the safest route. So, she did that as well." (Lily, 16-years old, from the DCD group).

While parents of children with both DCD and ADHD saw road safety education as crucial, their experiences revealed significant challenges in transferring theoretical knowledge into practical implementation. These parents, for example, reported that their children had received some road safety education at school and scout club. However, they agreed that their children might not put what they had learned into practice, particularly if they were not familiar with the roads.

"Theoretically, it does not work. For it to be slightly muscle memory, you need to do things physically. And that means practicing it, getting in the habit of doing it, and seeing it in practice." (Amelia, 7-years old, from the co-occurring group).

They further believe that simulation, one to one training and a training program with movement activities and visual materials would be more effective.

"Somehow exposing him to dangers in a safe way. Like through simulation, might help him understand and remember road safety lessons better. You can do visual social stories with him, but he does not relate to that if it is presented on a page, I think you know if you put him through some kind of road safety simulation, that would work because he'd remember it and he'd be in it." (Maryam, 10-years old, from the co-occurring group).

"I believe that learning should come from an angle where it's kind of like a visual, auditory, reading, and kinesthetic experience. So, it would involve a lot of movement, a lot of walking around, a lot of like activities, things that are visual, things that give context to the situation. You know, like when they do, first aid, you have gotta practice doing the CPR and stuff like that" (Maryam, 10-years old, from the co-occurring group).

"You're talking one teacher to 30-odd children, so Malcolm does not concentrate very well in school but he benefits greatly from one-on-one support." (Amelia, 7-years old, from the co-occurring group).

Some of them proposed that such training should be continuous, daily and revisited every few weeks or months with catchy campaigns and ads promoting road safety education. They also emphasized the importance of preparing their children for adulthood and for independent pedestrian mobility.

"Missed days with Malcom, it becomes out of his routine and then you just gotta start again. So doing things daily is really important. In school, it might be different, though. Say, if they were doing lessons on road safety in school, I think weekly would be fine if they got into a habit of it." (Amelia, 7-years old, from the co-occurring group).

In discussing road safety education, parents of children with DCD and/or ADHD highlighted various challenges specific to their respective group of children. Parents of children with ADHD emphasized the need for more impactful educational materials, such

as emotionally engaging stories, to supplement existing programs. They also stressed the importance of providing step-by-step instructions and minimizing distractions during training sessions. Parents of children with DCD described their efforts to adapt to their child's needs, including participation in school-based programs like Kerbcraft and transition training. They emphasized the significance of practical training and one-on-one support, as well as the need for continuous and frequent reinforcement of road safety concepts. Parents of children with co-occurring DCD and ADHD expressed similar viewpoints, underscoring the importance of personalized training approaches that incorporate visual and kinaesthetic elements. They also advocated for increased frequency and scope of road safety campaigns, along with real-life stories to highlight the risks associated with road traffic accidents. Overall, while parents across all groups recognized the importance of road safety education, they identified specific challenges and recommended tailored approaches to address the unique needs of their children.

Strategies to increase road safety

Parents of children with DCD and/or ADHD developed various strategies to mitigate the risk of pedestrian injuries. Table 4 contains a summary of the strategies used by parents. Parents of children with ADHD expressed the need for constant forward planning to reduce the risk of pedestrian injuries. They select quieter roads when possible and minimize the number of roads their child needs to cross. Some parents opted to drive their child to school and drop them off on quieter roads due to their child's behavioral characteristics.

"It's just having to constantly forward plan, even if it's just like a walk before bedtime, it's constantly forward planning because you can just never plan what she's gonna do." (Olivia, 9-years old, from the ADHD group).

"If there's a certain way, we'll walk, and we'll try and do it to the way where there's less roads to cross." (Alex, 9-years old, from the ADHD group).

To draw their child's attention to the road, parents often use verbal cues, such as talking through the situation and shouting ahead. Additionally, assigning responsibility to their child, such as asking the child to tell them when it is safe to cross the road, was reported to help children with ADHD to pay more attention. To increase safety, some parents use disability blue badges to park closer to their destinations. Crossing with peers and walking in the middle of the group were reported as tactics they use.

"you'd have to shout ahead. So, lots of talking it through, drawing his attention to look at the road." (Sophia, 13-years old, from the ADHD).

"I need to grab his attention first by tapping his shoulder or the back of his neck, or I'll try and get near him and say, like, hey, John, just to try and cut through what's going on in his brain" (Emily, 7-years old, from the ADHD group).

TABLE 4 Parents' strategies.

Group	Strategies mentioned by parents
ADHD	Constant forward planning
	Selecting quieter roads
	Driving to school
	Verbal cues and prompts
	Assigning responsibility
	Use of disability blue badges
	Crossing with peers
	Walking in the middle of the group
DCD	Repetition or repeated practice
	Familiarizing with environment and roads
	Crossing with peers
	Focus teaching on specific roads
	Avoiding traffic
ADHD + DCD	Verbal cues and prompts
	Hand gestures
	Physical guidance
	Repetition or repeated practice
	Familiarizing with environment and roads

"So, like if I ask him say like, you are gonna tell us when it's OK to cross the road, he will give more attention to the task" (Emily, 7-years old, from the ADHD group).

Parents of children with DCD emphasized the importance of repetition and familiarity in mitigating the risk of pedestrian injuries. They noted that their child would become overcautious in unfamiliar places and wait until there was no traffic before crossing the road. To address this, they identified the roads their child would need to cross and familiarized them with these roads, starting at a quiet time of day. One parent described how they repeatedly crossed the same main road with their child for about two weeks until the child was comfortable making the decision to cross safely.

"We went down to the main road and we just crossed it again and again and again. And all we did every day for like an hour a day, for about 2 weeks, was to just go to that road, cross to one side and then cross back again and then cross back again. But it was forcing her to make the decision. So, it was very time consuming." (Isabella, 17-years old, from the DCD group).

Due to their child's need for repeated learning, the parents emphasized the importance of repeating the practice several times to ensure their child was familiar with the route. Another parent mentioned that they would walk the route with their child several times to ensure they were comfortable and familiar with it before allowing their child to volunteer in that area.

"With her DCD, she needs to have repeated learning. So, you could not just do it once and then think she's OK. It needs to be repeated. So, four or five times because she's... for example, she's volunteering for play scheme to look after younger... So, we are gonna walk it about three or four times with her to make sure that she's familiar with the route." (Lily, 16-years old, from the DCD group).

Isabella further suggested implementing environmental changes such as adding more zebra crossings on main roads, particularly at roundabouts, to increase safety. She also suggested the use of indicators on the kerb to identify the safest place to cross. For example, these indicators could be a distinct marking on the pavement edge, clearly indicating the safest area for pedestrians to cross.

"I think the other thing that's helpful with zebra crossings and Pelican crossings is because it's like a set piece wherever you go... I think we need more Zebra crossings on main roads." (Isabella, 17-years old, from the DCD group).

However, parents noted that transferring skills from one road to another is challenging for their children. To address this issue, they suggested changing the teaching mindset from general life skills to specific roads that the child needs to cross.

"So that for every road that she's gonna need to cross, she has to work out a specific sort of skill set for that particular road." (Isabella, 17-years old, from the DCD group).

Finally, parents reported avoiding traffic whenever possible, for example, by driving to school earlier to park in a safer location.

"After the near misses. What I started doing was I started driving to the school 20 min earlier so that I could make sure I could park somewhere where she would not have to cross the car park to get to the car." (Isabella, 17-years old, from the DCD group).

"She'll only go at certain times of day, so she would not go when it was busy. So, sort of, you know, 4:30 she would not go because the road is too busy. So, she's picking times when it's not busy." (Isabella, 17-years old, from the DCD group).

Parents of children with both DCD and ADHD also discussed strategies to minimize the risk of pedestrian injuries. Verbal prompts were considered useful, as they help create awareness for the child, but constant reminders were necessary, especially when crossing the road.

"There is some awareness, but I always have to remind him to look both ways, because there's always that possibility that a car may not stop still. So, yeah, there's a lot of prompting... so I'll give him a heads up if we are going to go left or we are going to cross. So, sort of hand gestures." (Evelyn, 13-years old, from the co-occurring group).

Hand gestures were used to indicate the direction of movement and sometimes physical guidance was needed to help the child stay on track, especially when navigating unfamiliar environments. Repetition was also found effective, with one parent (Evelyn) noting progress over the past year by practicing crossing two specific roads to school every morning. Familiarity with the environment can also play a role, as the child may feel more confident and aware in familiar surroundings.

"Yeah, I've noticed progress definitely, especially over the last year in the two roads that we crossed to get to school with practice every single morning crossing those roads and when I'm with him, I'm telling him what to do and I'm watching him and trusting him to cross the roads safely. But it's gonna be a long time before we get to in being able to do anything like that on a busier road." (Evelyn, 13-years old, from the co-occurring group).

"It depends where he is. It depends whether he's familiar with that environment." (Charlotte, 16-years old, from the co-occurring group).

Overall, parents employed a range of strategies to ensure their child's pedestrian safety according to their needs. Children with ADHD thrive with forward planning, verbal prompts and attention management techniques, while those with DCD benefit from repetitive practice, familiar routines and environmental modifications. For the unique challenges posed by co-occurring DCD and ADHD, parents blend these approaches, adapting to unfamiliar situations, noting incremental progress.

Educators

The parents of children with DCD and/or ADHD discussed the responsibility of delivering road safety education for their children. Parents of children with ADHD believed that involving professionals such as the police and transport companies were crucial, in particular courses for bus drivers would be beneficial since busses are often used to transport children to and from school. Various delivery options were discussed, but parents believed that they ultimately had the most frequent opportunities to put education into practice and walk with their children every day, while teachers and Scout leaders could reinforce the message.

"Parents, teacher and Beaver's leader but the parents have the responsibility more than any other as they have more opportunity to put everything in practice and they walk everyday. Teacher and others should re-enforce." (Emily, 7-years old, from the ADHD group).

When it comes to children with DCD, the parents felt that the responsibility of delivering road safety education fell on them, as they knew their children best and could adapt to their needs accordingly. However, they also suggested that local authorities could play a role in delivering extra lessons in schools to supplement what parents were teaching at home, ensuring that children with DCD were equipped with the necessary road safety skills.

"This is our job, you know. You are the parent and yes, it takes longer with these children. But you know that's called being the parent of a dyspraxic child" (Isabella, 17-years old, from the DCD group).

"And with active travel, it's all very well put these roots in place, but if people are not learning their children to use them because the children got disabilities and they cannot use them, I think local authorities should help step in. I know the primary responsibility lies with parents at the end of the day, it is their children and they should make sure they are safe. But I think if the local authority can help by delivering extra lessons in school, it might be beneficial." (Lily, 16-years old, from the DCD group).

For children with ADHD and DCD, some parents believed that external professionals like the police would be more effective, while others suggested involving both parents and teachers. They also agreed that road safety education should be accessible to parents at home and delivered in schools by teachers, with the suggestion that teachers could incorporate it into the curriculum. While some parents believed that an external professional was needed, others felt a teacher with proper guidance documents could deliver a program effectively. Therefore, the parents felt that schools should take the lead in delivering road safety education, with parents and teachers working together to ensure their children's safety on the road.

"I think he probably responds better to external people. You know, if the police came and did it, he listens. You know the problem with parents doing things like that is you only have so much time in your day to do the things you need to do including therapy as well." (Maryam, 10-years old, from the co-occurring group).

"I think what would be really helpful if it's something that parents could access themselves, but also to be delivered in schools." (Harper, 7-years old from the co-occurring group).

"By default, they are probably say at schools, but I think if you can get into computer games, something where it's leisurely and does not feel like it's forced upon them. Because sometimes if you put it through, say, it's just for schools, it just schools teach it. It does not necessarily go in because it feels like you are forcing it in for information upon me instead of me understanding that it's valuable to me in life. So, it's getting that balance of doing it in a way that feels like it's fun." (Evelyn, 13-years old, from the co-occurring group).

Parents of children with DCD and/or ADHD discussed the responsibility of delivering road safety education for their children. Parents of children with ADHD felt that it was important to involve professionals like the police and transport companies, while also acknowledging their own role in daily practice. Parents of children with DCD felt primarily responsible but suggested local authorities supplement education. For children with co-occurring DCD and ADHD, parents had mixed views on involving professionals versus teachers, but agreed on the importance of accessible education delivered at home and in schools. Overall, ensuring road safety for children with DCD and/or ADHD was seen as a responsibility shared by parents, schools and local authorities. While parents saw themselves as playing a critical role in adapting education to their children's needs and reinforcing the message, schools can provide accessible education to supplement what parents teach at home. Additionally, local authorities can offer extra lessons and support to ensure children have the necessary skills to navigate the roads safely. By working together, parents, schools and local authorities can ensure the safety of all children on the roads.

Discussion

The aim of this study was to explore the perspective of parents of children with DCD and/or ADHD with the goal of gaining a better understanding of the pedestrian risks faced by their children. Semistructured interviews were conducted with parents and three main themes were generated, each aligning with a specific objective. In the first theme, parents' perspectives of the challenges faced by children at the roadside, addressed the objective of exploring the unique perspectives of parents of children with DCD and/or ADHD regarding their children's abilities and challenges in executing a safe road crossing., Additionally, the theme parental concerns and influences on children's road safety, sheds light on the worries and concerns parents have regarding their children's pedestrian safety covering the second objective. Finally, the objective of investigating the diverse strategies parents employ to minimize their child's involvement in pedestrian injuries was addressed in the road safety education: parental views, strategies, and collaboration theme. Although there was some overlap in the experiences shared by the participants, each parent provided a unique perspective and experience that contributed to a more comprehensive picture of the behavior of children with DCD and/or ADHD at the roadside. Importantly, the identified themes were not isolated from one another; they interacted and impacted the overall understanding of pedestrian behavior in this population to tell the everyday story of the parents of children with DCD and/or ADHD.

It is also important to note that the specific challenges and concerns varied depending on whether the child had DCD, ADHD or both DCD and ADHD. For example, parents of children with DCD primarily focused on difficulties with spatial awareness and motor skills, often struggling with judging distances and maneuvering safely around obstacles. Children with ADHD, on the other hand, faced challenges with impulsivity and inattention, which could lead to sudden dashes into traffic or difficulty focusing on potential dangers. Parents of children with co-occurring DCD and ADHD faced a complex interplay of these challenges, requiring constant vigilance and proactive measures to mitigate risks. This highlights the importance of tailoring interventions and support to the specific needs of each group, ensuring effective strategies that address their unique vulnerabilities and promote safe pedestrian behavior.

Challenges experienced by children at the roadside

The first theme uncovered important insights into the experiences of parents of children with DCD and/or ADHD in relation to roadside behavior and road crossing. Our findings suggest that parents of children with ADHD are concerned about their children's safety specifically when crossing driveways. Meanwhile, parents of children with DCD were more anxious about complex pedestrian environments like roundabouts. For parents of children with co-occurring DCD and ADHD, the concerns were compounded, encompassing both impulsivity at driveways and difficulties navigating complex environments. Notably, all three groups of parents (children with DCD, children with ADHD, and children with DCD and ADHD) shared a common agreement that zebra crossings, signalized crossings and road crossing patrols are perceived as safer options compared to midblock crossings. This finding emphasizes the significance of structured and controlled crossing sites for enhancing the safety of children with DCD and/or ADHD while navigating roads. This is consistent with previous research indicating that midblock crossings can pose greater risks because of the complexities involved in judging

distances, vehicle speeds, walking speeds and making accurate decisions (Purcell et al., 2011: Schwebel et al., 2014).

Regarding roadside behavior, children with ADHD exhibited high levels of activity and energy, which often led to a lack of attention to their surroundings and unintentionally walking or running on the road instead of the pavement. Conversely, a previous study conducted in an experimental setting by Stavrinos et al. (2011) found that children with ADHD-Combined Type demonstrated adequate pavement pedestrian behavior. The differences may be attributed to the inherent limitations of the laboratory setting, which may not fully replicate the complex and dynamic real-life environment characterized by a high volume of sensory stimuli as reported by parents in this study. This can be supported by Öhrström and Skånberg's (2004) exploration of the effects of traffic noise on sleep, where conflicting outcomes between field studies and laboratory experiments were highlighted, indicating potential limitations in the accuracy of results obtained solely from experimental settings. When discussing road crossing behavior, parents of children with ADHD reported their children's tendency to walk or run across the road, disregarding oncoming vehicles and environmental cues while crossing, which led to an increased possibility of engaging in unsafe crossings. Previous studies support this finding showing that children with ADHD are more likely to engage in unsafe road crossing behaviors, such as crossing when it is not safe, neglecting to look both ways before crossing and running across the road (Clancy et al., 2006; Stavrinos et al., 2011; Wilmut and Purcell, 2020). Furthermore, parents described their child as having "tunnel vision" in which children with ADHD focused solely on their intended destination. However, considering the characteristics associated with ADHD, including inattention and executive dysfunction, the concept of cognitive tunneling may provide a more accurate description (Briggs et al., 2016).

On the other hand, children with DCD struggled with spatial awareness and relied heavily on others for guidance, leading to reduced independence and decision-making at the roadside as reported by their parents. Furthermore, parents of children with DCD observed their children when crossing roads visually scanning before crossing but struggling to interpret the visual cues and make appropriate decisions. A study conducted by Purcell et al. (2012) found that children with DCD had lower looming detection thresholds compared to typically developing children, meaning they struggle to recognize an approaching object's potential threat as quickly as typical children, indicating weaker visual-motor processing skills that could lead to inaccurate crossing decisions. For example, children with DCD might misjudge a vehicles speed and assume a wider available traffic gap, potentially leading to risky situations. Therefore, the parents of children with DCD reported in their interviews that they often relied on other pedestrians or made random decisions, which increased their vulnerability to accidents.

According to parents, children with co-occurring DCD and ADHD displayed a combination of these behavioral characteristics, further increasing their vulnerability at the roadside and crossing roads. The lowered awareness in the DCD and co-occurring groups, but not in the ADHD group, is supported by Loh et al. (2011) who stated that impaired visual–spatial ability may be associated with DCD, while no similar association has been observed with ADHD. Parents spoke about the presence of behaviors attributed to ADHD. Parents of children with both DCD and ADHD reported that they engaged in more risky behavior and displayed significantly less

attention compared to those with DCD alone (Wilmut and Purcell, 2020). Thus, the presence of ADHD further exacerbates these difficulties, leading to a potential increase in their vulnerability to accidents.

Parental concerns and influences on children's road safety

The second theme captured parental understanding and emotional responses regarding their children's pedestrian safety, as well as their exploration of potential factors influencing their children's roadside performance. In this study, parents of children with DCD, ADHD and co-occurring DCD and ADHD expressed concerns about their children's roadside performance, which aligns with findings from Wilmut and Purcell (2020) who explored the lived experience of adults with DCD and parents of children with DCD using a self-report questionnaire. Brook and Boaz (2006) also found heightened concerns among parents of adolescents with ADHD compared to a typically developing control group. These findings have implications for road safety, as parents may be more inclined to limit their children's independent community mobility due to their concerns. Previous studies linked independent community mobility to enhanced physical health through active exploration, boosted mental well-being via cognitive development and independent play and stronger social bonds formed through peer interaction and community connection (Pacilli et al., 2013: Qiu and Zhu, 2017). In fact, parents of children with DCD and/or ADHD in this study reported closely monitoring their children's community movements, limiting active travel while relying on vehicles and limiting unsupervised outings because of these concerns. Moreover, the compromised coordination and communication between parents and children with ADHD when crossing, often exacerbated by anxiety and fear, can directly impact their attention and decision-making (O'Neal et al., 2022). These negative emotions, further heightened by situations where parents and children choose different crossing gaps, can impair children's ability to process information and make safe choices leading to increased collision risk and unsafe behaviors (O'Neal et al., 2022). Similarly, the emerging evidence indicating the effect of higher task-specific anxiety on motor behavior in children with DCD leading to poor gaze patterns, stepping behavior and gait, could increase the risk of accidents, further emphasizing the potential impact of emotional factors on roadside performance (Harris et al., 2022). Overall, these factors may result in parents adopting an overly protective approach toward their child, potentially leading to increased isolation and delayed development of independent mobility skills. Therefore, parents of children with DCD and/or ADHD were concerned about finding the balance between promoting their children's independence and prioritizing their safety and well-being as pedestrians.

Furthermore, parents believed that traits related to DCD and/or ADHD seemed to influence their children's pedestrian performance and road crossing behaviors. Parents of children with ADHD mentioned the influence of mood and temperament on their child's pedestrian safety. They observed that their child's impulsiveness, inattention and difficulty concentrating during negative mood episodes can contribute to engaging in unsafe crossing behaviors. Conversely, periods of improved mood and enhanced focus were associated with better performance in pedestrian tasks. Skirrow et al.

(2009) conducted a study suggesting that mood instability and ADHD traits may be interconnected and that mood instability could be considered a fundamental aspect of ADHD. However, Clancy et al. (2006) suggested that inattention may have a greater significance in predicting safety in the context of road crossing when compared to a typically developing control group. Further studies found a positive correlation between executive dysfunction and unsafe pedestrian crossings for both children with ADHD and typically developing children (Stavrinos et al., 2011; Tabibi et al., 2022, 2023). Additionally, Tabibi et al. (2023) stated that attentional abilities did not have a substantial impact on determining unsafe behaviors as measured by the Integrated Visual and Auditory Continuous Performance Test (IVA + Plus), a computerized test that assess different components of attention (Sanford and Turner, 1995). Generally, poor pedestrian performance among children with ADHD can collectively be attributed to a combination of ADHD characteristics, such as inattention, impulsivity and lowered concentration, which are influenced by mood and temperament, as well as executive dysfunction leading to the increased risk of unsafe crossing behaviors in children with ADHD. Therefore, the research findings that suggest that a combination of ADHD traits, executive dysfunction and mood and temperament can influence the pedestrian safety of children with ADHD, are supported by parents' perspectives in this study.

On the other hand, parents of children with DCD believed that their child's pedestrian performance is related to their inability to judge distance and speed accurately. This aligns with the research on looming sensitivity observed in the study by Purcell et al. (2012), as accurate perceptions of approaching vehicles to avoid collisions requires accurate judgment of the optical equivalent of distance and speed. This is further supported by the finding that children with DCD select insufficient temporal crossing gaps when presented with a virtual task simulating road crossing scenarios across different vehicle approach speeds, indicating difficulties in accurately judging and selecting appropriate gaps for safe crossing (Purcell et al., 2017) and demonstrating deficits in visuomotor adaptation skills (Bo and Lee, 2013). Additionally, this study's findings indicate that the reported difficulties with body position awareness further contribute to the challenges faced by these children in navigating obstacles and people on the pavement. The presence of fatigue, forgetfulness and diminished self-confidence can further influence the roadside behaviors of children with DCD as reported by parents in the study. Recent research using self-reported questionnaires found a decrease in confidence in road crossing skills among both adults and children with DCD (Wilmut and Purcell, 2020), supporting the findings from this study. However, earlier studies showed no difference in selfreported confidence regarding children aged 6 to 12 years with DCD's ability to independently and safely cross the road compared to their typically developing peers (Purcell et al., 2012; Purcell and Romijn, 2017). The differences in confidence levels between the current study and studies showing no difference in confidence may be attributed to the older age of participants included in this study. Therefore, developmental changes and the reliance on parent perspectives could contribute to the observed differences in confidence among individuals with DCD. Furthermore, while executive function deficits are also known in DCD (Sartori et al., 2020), their link to pedestrian performance remains unclear despite some associations with poor driving (Kirby et al., 2011). This suggests a potential role of executive functions in pedestrian safety, but more research is needed to understand its specific influence in this context. Overall, the challenges related to spatial awareness and accurate perception experienced by children with DCD could be the main contributors to the elevated likelihood of engaging in unsafe pedestrian behaviors.

For parents of children with co-occurring DCD and ADHD, understanding the underlying causes are more complex due to various factors related to the characteristics of ADHD and DCD. Parents reported that the pedestrian performance of their children can be influenced by inattention and impulsivity, which is related to inhibitory control, in addition to poor perceptual-motor skills and spatial awareness. Previous studies highlighted the presence of overlapping characteristics between DCD and/or ADHD (Bernardi et al., 2015; Harrowell et al., 2018; Wilson et al., 2020; Meachon et al., 2021). For instance, hyperactivity was identified as a co-occurring difficulty among children with DCD (Harrowell et al., 2018). Sartori et al. (2020) also found that children with DCD have poor performance in multiple executive functions including cognitive flexibility, working memory and inhibitory control. However, Meachon et al. (2021) explored the underpinning neurological mechanisms among individuals with DCD and/or ADHD in relation to inhibitory control and found that each group has a distinct executive mechanism despite the overt behavioral similarities. These findings suggest that although individuals with DCD and ADHD may present similar behaviors, including roadside behaviors, their underlying neurological mechanisms are distinct and should be addressed differently.

In summary, parents of children with DCD and/or ADHD in this study are facing the challenge of balancing between fostering children's independence and ensuring their safety as pedestrians. These parents attributed the current pedestrian performance of their children to the characteristics related to DCD and ADHD. However, emerging evidence indicates that executive dysfunction may serve as the underlying cause of their performance at the roadside. While the results are not conclusive, this implies that the road crossing behaviors of children with DCD and/or ADHD should be approached differently to ensure their road safety or when aiming to develop pedestrian skills. By recognizing the potential influence of executive dysfunction, interventions and strategies tailored to the specific needs of these children could be developed to optimize their road safety and pedestrian abilities.

Road safety education: parental views, strategies, and collaboration

The third theme explored the approaches adopted by parents and the importance of educational interventions in promoting pedestrian safety among children with DCD and/or ADHD. The findings revealed several key aspects in this domain.

Firstly, parents of children with DCD and/or ADHD expressed concerns about the inadequacy of current pedestrian safety programs in meeting their child's unique needs. They emphasized the importance of addressing additional needs in preparing their children for the transition to becoming an independent pedestrian through using tailored approaches and multiple modes of delivery. Parents suggested the implementation of customized programs that specifically address the distinctive traits associated with DCD and/or ADHD. For instance, in the case of children with DCD, previous studies identified specific elements that should be considered to enhance pedestrian safety. Notably, research conducted by Purcell

et al. (2012) revealed a lower looming detection threshold among children with DCD, which has implications for their ability to perceive and respond to approaching objects or vehicles. Additionally, Purcell and Romijn (2017) suggest that a multimodal approach, involving both allocentric (environment-centered) and egocentric (selfcentered) approaches, may be necessary to effectively be used by parents to teach road safety to children with DCD. These findings are closely linked to the reported difficulties in visual processing of perceived information and spatial awareness experienced by children with DCD. Repetition was also reported by parents as an effective element to improve the pedestrian safety of children with DCD and co-occurring DCD and ADHD. For example, parents in this study suggested that simulated environments, which can be virtual or physical, can provide safe repeated opportunities to learn pedestrian skills. Emerging evidence also suggests that virtual reality can be an effective approach for creating a safe environment that facilitates repetitive practice and improves pedestrian safety, benefiting both typically developing children and those with DCD and/or ADHD (Clancy et al., 2006; Purcell and Romijn, 2017; Morrongiello et al., 2018; Schwebel et al., 2018). By incorporating these insights and strategies into tailored programs, the specific challenges faced by children with DCD and/or ADHD could be effectively addressed.

Secondly, parents of children with DCD and/or ADHD implemented various strategies to enhance their children's pedestrian safety and decrease the risk of road crossing injuries. Despite efforts to provide pedestrian safety training, the alarming rate of fatalities among children on our roads remains a significant cause for concern and it is evident that implementing behavioral strategies can be costefficient and play a crucial role in enhancing safety (Schwebel et al., 2014). Furthermore, adopting these strategies to incorporate the distinct requirements of children can be effective in fostering their pedestrian skills and formulating tailored interventions. Therefore, it is imperative to understand the strategies used by parents to mitigate the risk of child pedestrian injuries. A summary of the parent strategies identified by each group is provided in Table 4. Common strategies used by parents of children with DCD and/or ADHD include constant forward planning, selecting quieter roads, driving to school, using verbal prompts, crossing with peers, walking in the middle of a group, repeated practice, avoiding traffic, using hand gestures, offering physical guidance and fostering familiarity with the environment. However, it is important to note that some of these strategies may not be feasible for all parents, depending on their individual circumstances. It is also worth considering that some of these strategies, while prioritizing pedestrian safety, may hinder the development of road crossing skills necessary for future independent mobility. For example, parents who consistently drive their children to school may not be able to provide an opportunity for their children to practice crossing the road safely in an unsupervised environment. Therefore, it is important for parents to identify strategies that are suitable for their children and their circumstances, while also considering the long-term impact of these strategies.

Moreover, the responsibility of ensuring pedestrian safety for children with DCD and/or ADHD extends beyond parents alone and involves collaboration among schools and local authorities. While parents of children with DCD and/or ADHD in this study face time constraints, they actively take on the responsibility of teaching their children pedestrian skills and ensuring their safety on the roads. These parental efforts align with previous research (Morrongiello and

Corbett, 2015; Ngu et al., 2016; Zare et al., 2019), which underscores the crucial role of parental involvement in enhancing pedestrian skills and mitigating the risk of accidents. Additionally, collaborative efforts involving schools and local authorities showed effective results in promoting road safety education. Studies investigating the impact of road crossing programs involving the active participation of schoolteachers and police officers in enhancing pedestrian skills (Schwebel et al., 2018; Zare et al., 2018; McLaughlin et al., 2019; Jiang et al., 2021) show the valuable contribution of educational institutions and local authorities in fostering pedestrian skills among typically developing children. Additional research is needed to further explore the collaborative efforts between these stakeholders specifically focusing on children with DCD and/or ADHD. Another crucial obstacle to consider when promoting pedestrian safety training for children with DCD, in particular, is the lack of widespread awareness and knowledge about the condition, even among medical and educational professionals (Hunt et al., 2021; Meachon et al., 2023). Therefore, fostering collaborative efforts must include raising awareness and providing targeted training for educators, healthcare professionals and local authorities, to equip them with the knowledge and skills necessary to support children with DCD as well as the other two groups. By recognizing the shared responsibility and fostering collaboration among parents, schools and local authorities, comprehensive and effective measures could be implemented to promote the pedestrian safety of children with ADHD and/or DCD.

The study provides findings into the lived experience of parents of children with DCD and/or ADHD in the context of road crossing. However, there are certain limitations to consider. One limitation to this study is that the sample size was relatively small, with only 14 participants, which may limit the variability and diversity of perspectives represented. Furthermore, the limited subgroup size of only three participants with DCD presents a specific challenge. Given the known complexity and heterogeneity of DCD, this small sample may not adequately capture the full range of experiences and challenges faced by this group within the context of road crossing. This limits the study's ability to confidently generalize findings to the broader population of parents of children with DCD and may mask potentially specific perspectives or concerns unique to this subgroup. Moreover, while the study aimed to understand parental perspectives across different age groups, the age ranges within each group varied slightly (DCD: 10-17 years, ADHD: 7-13 years, Co-occurring: 7–16 years). This variation may have influenced parental perspectives of age-related differences in pedestrian behaviors and limits the ability to draw qualitatively based conclusions about age-based trends from the current dataset. Nonetheless, the study offers valuable preliminary findings that warrant further investigation. Moreover, the study focused solely on parents' perspectives regarding road crossing, overlooking the child's perspective and the broader experiences and challenges parents face in other aspects of parenting. Future research should explore the perspectives of children with DCD and/or ADHD regarding road crossing and investigate additional dimensions of parenting challenges and examine the impact of DCD and/or ADHD on other daily activities beyond pedestrian safety. Additionally, it is worth noting that the semi-structured interviews were conducted online, potentially excluding individuals without internet access or those less comfortable with online communication. This could introduce bias, as those with different access or preferences may possess unique perspectives or experiences related to road crossing.

In conclusion, the findings revealed several key insights that represent parents' perspectives of children with DCD and/or ADHD regarding the pedestrian risks faced by their children. Firstly, the importance of structured and controlled pedestrian crossing sites was emphasized by parents. Secondly, parents expressed heightened concerns about their children's performance and safety at the roadside, leading to increased monitoring and a more protective approach. Addressing these concerns is essential to promote the independence and well-being of these children. Additionally, while the underlying causes are not yet fully understood, it is evident that the reported road crossing behaviors of children with DCD and/or ADHD require a distinct approach to better develop their pedestrian skills effectively. Furthermore, parents implemented various strategies to mitigate the risks associated with roadside activities, but it is important to balance independence and the development of pedestrian skills. Lastly, promoting pedestrian safety for children with DCD and/or ADHD will require collaboration and shared responsibility between parents, schools and local authorities to implement comprehensive measures to ensure their safety and well-being. These findings contribute to the understanding of the perspectives of parents and provide valuable guidance for the development of targeted interventions and policies to promote the road safety of children with DCD and/or ADHD.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

Ethics statement

The studies involving humans were approved by the School of Healthcare Sciences Research Ethics Committee. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

RF: Writing – review & editing, Writing – original draft, Visualization, Methodology, Investigation, Formal Analysis, Data curation, Conceptualization. KW: Writing – review & editing,

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

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Lawrence M. Parsons,
The University of Sheffield, United Kingdom

REVIEWED BY Manfred Klöbl, Medical University of Vienna, Austria Logan Thomas Trujillo, Texas State University, United States

*CORRESPONDENCE
Emily J. Meachon

☑ Emily.Meachon@unibas.ch
Georg W. Alpers
☑ alpers@uni-mannheim.de

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EEG spectral power in developmental coordination disorder and attention-deficit/hyperactivity disorder: a pilot study

Emily J. Meachon^{1,2*}, Marlene Kundlacz¹, Kate Wilmut³ and Georg W. Alpers^{1*}

¹School of Social Sciences, University of Mannheim, Mannheim, Germany, ²Faculty of Psychology, University of Basel, Basel, Switzerland, ³Centre for Psychological Research, Faculty of Health and Life Sciences, Oxford Brookes University, Oxford, United Kingdom

Developmental coordination disorder (DCD) and attention-deficit/hyperactivity disorder (ADHD) overlap in symptoms and often co-occur. Differentiation of DCD and ADHD is crucial for a better understanding of the conditions and targeted support. Measuring electrical brain activity with EEG may help to discern and better understand the conditions given that it can objectively capture changes and potential differences in brain activity related to externally measurable symptoms beneficial for targeted interventions. Therefore, a pilot study was conducted to exploratorily examine neurophysiological differences between adults with DCD and/or ADHD at rest. A total of N = 46 adults with DCD (n = 12), ADHD (n = 9), both DCD + ADHD (n = 8), or typical development (n = 17) completed 2 min of rest with eyes-closed and eyes-open while their EEG was recorded. Spectral power was calculated for frequency bands: delta (0.5-3 Hz), theta (3.5-7 Hz), alpha (7.5-12.5 Hz), beta (13-25 Hz), mu (8-13 Hz), gamma (low: 30-40 Hz; high: 40-50 Hz). Within-participants, spectral power in a majority of waveforms significantly increased from eyesopen to eyes-closed conditions. Groups differed significantly in occipital beta power during the eyes-open condition, driven by the DCD versus typically developing group comparison. However, other group comparisons reached only marginal significance, including whole brain alpha and mu power with eyes-open, and frontal beta and occipital high gamma power during eyesclosed. While no strong markers could be determined to differentiate DCD versus ADHD, we theorize that several patterns in beta activity were indicative of potential motor maintenance differences in DCD at rest. Therefore, larger studies comparing EEG spectral power may be useful to identify neurological mechanisms of DCD and continued differentiation of DCD and ADHD.

KEYWORDS

electroence phalography, neuro development a l disorders, dyspraxia, oscillations, resting state

1 Introduction

Developmental Coordination Disorder (DCD) and Attention-Deficit/Hyperactivity Disorder (ADHD) are neurodevelopmental disorders, each affecting about 5% of the population (Thomas et al., 2015; Blank et al., 2019). Despite their unique diagnostic specifications in the DSM-5, DCD and ADHD overlap in many secondary symptoms, including motor and executive functioning difficulties, and can be challenging to disentangle (American Psychiatric Association, 2013; Kaiser et al., 2015; Meachon et al., 2022). In addition, DCD and ADHD co-occur in about 50% of cases and it is not clear if co-occurrence is driving the symptom overlaps or vice versa (Blank et al., 2019). In previous studies, surmounting evidence shows that adults in particular do not have significantly different objective task performance when their symptoms are engaged, but can differ in underlying mechanisms observed at the neural level (e.g., via inhibition: MacLaren et al., 2007; Meachon et al., 2021). Therefore, it is important to differentiate DCD and ADHD by examining neural mechanisms of one or both conditions.

Some studies have shown there are functional differences in the brain between children with DCD and/or ADHD during cognitive tasks (e.g., McLeod et al., 2014) and at rest (e.g., McLeod et al., 2016; Rohr et al., 2021). Furthermore, neural differences are often particular to individuals with co-occurring DCD and ADHD as opposed to just one condition or those of typical development (McLeod et al., 2014; Langevin et al., 2015; Thornton et al., 2018). However, the detection of differences in neural activity between DCD and ADHD has been mixed: some studies found key distinctions (McLeod et al., 2014, 2016) and others noted subtle to no differences (Thornton et al., 2018; Rinat et al., 2020). In addition, it is still unclear if individuals with DCD and/or ADHD have different baseline activity in the resting state and, to our knowledge, this has not been explored among adult groups (Tallet and Wilson, 2020; Meachon et al., 2021). Therefore, the present study exploratorily investigated oscillatory resting state electrical brain activity (i.e., alpha, beta, theta, delta, gamma, and mu) via electroencephalography (EEG) in adults with DCD and/or ADHD.

We first provide an overview of resting state measurement with EEG, describe the existing evidence of resting state activity in DCD or ADHD, and detail the known symptomatic overlaps of DCD and ADHD. While the resting state is often equated to measurement of the default mode network in imaging and connectivity research (Mak et al., 2017), for clarity, we use the term "rest state" throughout the paper to describe EEG measurement not explicitly intended to assess connectivity.

1.1 Resting state measurement with EEG

EEG is used to measure the electrical impulses of axonal activity in near-surface regions of the brain (Teplan, 2002). Numerous experiments have used EEG to gain insights into the underlying neural activity in relation to various disorders in biological and psychological science (e.g., Michel et al., 1993; Luck, 2014). EEG is considered an essential tool in several research fields (e.g., attention and speech development), in diagnosis (e.g., epilepsy and sleep disorders; O'Sullivan et al., 2006), and it provides highly accurate

temporal resolution not achievable with other neurophysiological measures (e.g., NIRS and MRI).

EEG is often used to capture spectral power through specific frequencies of electrical activity, also known as oscillations. Oscillatory activity can be used to infer general information about one's conscious state and can be used to discriminate various disorders of consciousness and to predict several cognitive functions such as attention and fluid intelligence (White and Siegel, 2016; Corchs et al., 2019; Rogala et al., 2020). There are four major forms of wavelengths observed in EEG, including: alpha, beta, theta, and delta which occur within unique ranges (in Hertz: Hz) and have different associations. The alpha band has a range of around 8-12 Hz and signifies a relaxed state characterized by medium to large amplitudes (10-150 microV; Klimesch, 1999) theorized to reflect underlying inhibition and cognition relevant to attention (Klimesch, 2012). Beta frequencies occur in a range around 14-25 Hz and indicate focused wakefulness, characterized by small amplitudes (<25 microV; Lubar et al., 1995) which may reflect the presence or absence of maintaining one's cognitive or sensorimotor state (Engel and Fries, 2010). The theta band is often considered a marker of attentional control and has a range of about 4-7 Hz with large amplitudes (>50microV; Cavanagh and Frank, 2014). The range of delta waves and their implications are generally more variable than other frequency bands, but occur during slow-wave sleep (Dijk et al., 1990) and may interfere with one's ability to complete a cognitive task (Harmony, 2013).

An even more ambiguous frequency is the gamma band which occurs at frequencies of 25 Hz or greater in wakeful and sleep states (Mably and Colgin, 2018). Gamma activity is thought to be non-specific in function but can occur in response to sensory stimuli and higher order cognitive processing (Başar, 2013; Mably and Colgin, 2018). Gamma frequency bands were among the least reported frequencies in studies examining resting state electrophysiology in psychiatric conditions (Newson and Thiagarajan, 2019). Finally, the mu frequency which occurs around 8-13 Hz, is thought to reflect cognitive processing when paired with beta bands and motor processing, motor imagery, perception, and/or action in combination with alpha bands (Pineda, 2005; Pfurtscheller et al., 2006; Démas et al., 2020). While the higher end of the mu frequency spectrum is thought to be specific to central region and motor-related activity, the higher end of the spectrum is considered to be generalized in both aspects (Thorpe et al., 2016). Some have suggested mu is specific to sensorimotor regions whereas the overlapping alpha frequency is more relevant to the occipital cortex, they are highly difficult to distinguish (e.g., Garakh et al., 2020). For example, when measured with in adults EEG alone, mu rhythms can distribute more widely and should be considered across the cortical surface (e.g., Thorpe et al., 2016).

In typical resting state EEG measurement, participants sit still with open or closed eyes for several seconds to minutes at a time. Rest state activity can differ substantially between conditions with eyesopen and eyes-closed, especially because when participants' eyes are open, they are naturally exposed to more visual stimuli associated with arousal levels, e.g., via skin conductance (Barry et al., 2007, 2009; Alba et al., 2016). In the eyes-open resting condition, amplitudes in all four major frequency bands are typically reduced compared when one's eyes are closed and the alpha band in particular is highly relevant to the resting state because it primarily indexes resting state-related arousal rather than activation indicative of visual processing changes

from eyes-closed to eyes-open conditions (e.g., Barry et al., 2007; Barry and De Blasio, 2017).

ADHD or useful for its distinction (Newson and Thiagarajan, 2019).

1.2 Resting state brain activity in DCD and ADHD

Several studies have examined differences in resting state neurophysiological activity in children with DCD alone compared to typically developing children (De Castelnau et al., 2008), or ADHD alone compared to typically developing children (e.g., Liechti et al., 2013; Buyck and Wiersema, 2014; Alba et al., 2016). However, the neurophysiological evidence surrounding DCD in the resting state is particularly limited. It has been theorized that symptoms of DCD are related to a difference in frequency band coherence in the brain during motor tasks (Tallet and Wilson, 2020). Accordingly, DCD has been dubbed a "disconnection syndrome" due to alterations in connectivity between different areas of the brain among individuals with DCD (Tallet and Wilson, 2020). One EEG study examined spectral coherence during different motor tasks among children with DCD, with motor tasks varying from simple to difficult (De Castelnau et al., 2008). The alpha and beta frequency bands showed increased coherence in children with DCD compared to typically developing controls, which was likely related to sensorimotor activation (De Castelnau et al., 2008). In this case, higher coherence is considered to be more dysfunctional and increased with heightened task difficulty (De Castelnau et al., 2008). These results provided evidence that children with DCD have a higher cognitive load while performing motor tasks and reduced connectivity during these tasks. Furthermore, Keating et al. (2023) recently reported that children with DCD showed reduced synchronization of mu oscillations during movement and reduced mu power while observing a moving kaleidoscope pattern compared to typically developing children. However, no differences in mu and alpha activity were detected at rest between groups or between eyes-open and eyes-closed trials (Keating et al., 2023). While this suggests a role of mu in movement relevant to DCD, it is unclear why activity did not differ between eyes-open and eyes-closed trials.

Contrary to the limited evidence for DCD, the resting state has been examined in a plethora of EEG studies about ADHD (e.g., Barry et al., 2011; González et al., 2013; Buyck and Wiersema, 2014; Alba et al., 2016; Rommel et al., 2017; Clarke et al., 2020). It is considered a robust finding that absolute delta power is increased in individuals with ADHD during eyes-closed compared to typically developing individuals (Newson and Thiagarajan, 2019). The theta-beta ratio has also been suggested as a biomarker for ADHD, however, a review of 65 studies of the resting state in ADHD showed this result is inconsistent in adults and likely dependent on age (Newson and Thiagarajan, 2019). This was confirmed in several studies, such as Kiiski et al. (2022), who identified numerous features of absolute and relative spectral power relevant to predicting ADHD in adults which did not include the theta/beta ratio. For example, increased power in delta and theta spectral power, could successfully classify those with ADHD from typically developing individuals (Kiiski et al., 2022). Furthermore, it is possible differences in theta activity and the theta-beta ratio can also occur in other disorders, such as epilepsy, dementia, alcoholism, and schizophrenia, and is not specific to

1.3 Current study

First, we expected the level of electrical activity in the brain would generally decrease (i.e., in average activity of all participants) from the eyes-closed to eyes-open condition as has been observed in previous studies (e.g., Barry et al., 2007; Barry and De Blasio, 2017). Second, we hypothesized that theta and delta frequencies will be increased in the ADHD group compared to the control group replicating the results of Kiiski et al. (2022) and the collective findings reviewed by Newson and Thiagarajan (2019) in parietal-occipital regions and overall. Third, based on the existing evidence that beta bands are linked to sensorimotor activation (e.g., De Castelnau et al., 2008), we expect increased power in frontal and central beta frequencies will be present indicating impairment among those with DCD compared to typically developing adults. Considering the findings for alpha frequencies are mixed, we will also examine if in frontal and central alpha are increased in DCD in line with de Castelnau et al. (2008), or if there are no between-group differences in alpha power in line with Keating et al. (2023). As there are no existing studies to indicate the general frequency band patterns in participants with DCD + ADHD or to examine differences at rest between DCD and ADHD groups, we exploratorily compare frequency band activity between all groups (i.e., DCD, ADHD, DCD+ADHD, typically developing) by brain region. Exploratory correlation analyses will also be conducted to compare symptom severity to spectral power.

2 Methods

2.1 Participants

A total of N = 46 adults were included in the present study. Among them, n = 12 had a diagnosis of DCD, n = 9 had a diagnosis of ADHD, n = 8 were diagnosed with both DCD and ADHD (DCD + ADHD), while n = 17 were typically developing, with no known mental or physical health conditions. Participants identified as women (n = 35), men (n=10), and transgender (n=1). In addition, a majority were right-handed (n = 37). They were, on average, 25.8 years old (SD = 7.85;Range: 19-53). As adults with DCD in particular can be difficult to recruit, combining test locations is a common approach in DCD research to gather larger sample sizes while the condition remains under-recognized (e.g., Meachon et al., 2021; Miller et al., 2021). Therefore, participants were tested in Germany (n = 26) and the UK (n=20). Aside from the language in which the study session was conducted, demographic factors (i.e., age, gender, handedness) neither differed between study groups nor based on test site (see Supplementary material).

Several participants in the clinical groups had co-occurring mental health conditions, including autism spectrum disorder (n=4), dyslexia (n=2), learning difficulties (n=2), and anxiety or depression (n=2). All participants with ADHD were asked not to take ADHD medication for 24 h before the testing session. The study was approved by the local ethics committees at both sites.

TABLE 1 Group classification and testing location comparisons.

Groups: overall (N = 46)	Sample size (n)	Average ADC score (SD)	Average ASRS v.1 score (SD)	Median MABC-2 percentile
DCD	12	108.2 (22.5)	41.5 (8.8)	N/A
ADHD	9	89.1 (14.5)	59.3 (8.8)	N/A
DCD+ADHD	8	112.0 (18.8)	59.0 (13.4)	N/A
Control	17	69.8 (13.8)	44.4 (11.4)	N/A

Participants from Germany (n = 26)	Sample size	Average ADC score	Average ASRS v.1 score	Median MABC-2 Percentile
DCD	2	85.5 (47.4)	43.0 (7.1)	N/A
ADHD	6	91 (15.4)	61.3 (10.5)	N/A
DCD+ADHD	3	118.7 (24.9)	69.0 (8.7)	N/A
Control	15	72.0 (13.1)	47.8 (6.5)	N/A

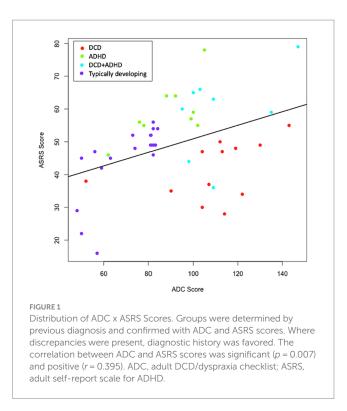
Participants from UK (n = 20)	Sample size	Average ADC score	Average ASRS v.1 score	Median MABC-2 percentile
DCD	10	113.9 (14.9)	41.2 (9.5)	3rd
ADHD	3	85.3 (14.5)	55.3 (0.6)	25th
DCD+ADHD	5	108.0 (16.0)	53.5 (12.5)	5 th
Control	2	53.5 (5.0)	19 (4.2)	55 th

Overall group scores on the ADC and ASRS v.1 were compared via a one-way ANOVA. There was a significant effect of group on ADC score [F(3,42)=16.61,p<0.001]. Tukey's post hoc test revealed group comparisons between the control group versus the DCD and DCD+ADHD groups were significant (p<0.001). There was also a significant effect of group on ASRS v.1 scores [F(3,42)=8.13,p<0.001]. Tukey's post hoc test revealed significance (p<0.01) in all group comparisons except for the DCD versus typically developing groups and the ADHD versus DCD+ADHD groups. ADC scores were based on a scale with responses scored from values 1 to 4 as opposed to 0–3. Therefore, cutoff scores are higher than recommendations for >/= 65, instead at 98 and over. ADC, adult DCD/dyspraxia checklist; ASRS, adult self-report scale for ADHD; MABC-2, movement assessment battery

2.2 Screening and group classification

All participants were screened in line with the DSM-5 diagnosis and current gold standard assessment for adults with DCD (American Psychiatric Association, 2013; Blank et al., 2019). Criterion A for DCD (indicating motor skill acquisition and executive are below peers; American Psychiatric Association, 2013) was confirmed in participants recruited in the UK using the Movement Assessment Battery for Children version 2 (MABC-2; Henderson et al., 2007; see Table 1). In addition, criterion B and C (B: motor skills interfere with daily life in several domains, C: symptoms began in childhood; American Psychiatric Association, 2013) were addressed with the Adult DCD/Dyspraxia Checklist (Kirby et al., 2010; American Psychiatric Association, 2013). Furthermore, presence of ADHD symptoms was assessed with the Adult Self-Report Scale for ADHD v.1 (Kessler et al., 2005; see Table 1).

Groups were classified based on previous diagnosis and confirmed to differ in expected directions based on self-reported symptoms of DCD and/or ADHD (see Table 1). As visualized in Figure 1, some clear group distinctions can also be observed. In some cases,



participants had borderline values in self-reported DCD and ADHD symptoms (see Supplementary material), however, they are highly consistent by study group based on previous diagnosis (see Figure 1).

2.3 Procedure

Resting state trials included 2 min with eyes-open, and 2 min with eyes-closed, respectively. For eyes-open trials, participants were told to relax and look at a fixation cross in the middle of the screen while preventing head or eye movements as much as possible. For eyes-closed trials, participants were instructed to remain relaxed and awake but still. Participants had the opportunity to take a break and move between eyes-open and eyes-closed trials. The data is a subset of participants who completed resting tasks in the middle of a broader pilot study which included a detailed questionnaire and executive functioning tasks [see Meachon et al. (2021)].

2.4 EEG measurement

The EEG measurement took place in a soundproof booth with absence of phones or other technology aside from the study equipment. Participants were seated in a comfortable chair at a viewing distance of approximately 150 cm to the screen, with a visual angle of about 15 degrees, and had a keyboard in front of them to advance between trials during the break via a mouse click. Measurement of the rest state task lasted 4 min total, plus an openended break between eyes-open and eyes-closed trials. A black fixation cross was presented in the middle of a gray screen (visual angle: 43° in Germany and 67° in UK). The EEG systems at both sites had 64 electrodes which followed the international 10–20 system (Brain Products GmbH) with a ground electrode at FpCz and

Reference at FCz. The impedance of the electrodes was monitored closely as to not exceed 15 k Ω .

2.5 EEG pre-processing

Electroencephalography data were recorded at or adjusted to a sampling rate of 500 Hz for consistency between tests sites. All data used band pass filters of 0.5 and 50 Hz in line with similar resting state EEG studies (De Castelnau et al., 2008; Van Dongen-Boomsma et al., 2010; Woltering et al., 2012; Liechti et al., 2013; Buyck and Wiersema, 2014; Alba et al., 2016; Rommel et al., 2017). The reference was computed using the average of all electrodes. When individual electrodes were substantially noisy and/or lack of signal reception was suspected, a topographical interpolation was performed. This was computed in n=5 participants for an average of 1.4 electrodes each. An independent component analysis (ICA) was conducted for each subject to detect and remove eyeblink and movement artifacts where relevant. Finally, artifact rejection was performed to automatically remove noisy epochs for both trial types, leading to a removal of small amounts of data in n = 17 participants. The range of artifact removal was from 0.4s (i.e., 0.003% of the data in one condition for one participant) to 124s (i.e., 1.1% of the data in one condition for one participant) with a median of 2.1 s. The eyes-open and eyes-closed conditions were pre-processed separately, and each divided into 2 s epochs without overlap. All pre-processing was conducted with Brain Vision Analyzer 2.0 (Brain Products, Germany).

2.6 EEG analysis

At each electrode and using a windowing approach, amplitudes were measured for alpha (7.5–12.5 Hz), beta (13–25 Hz), theta (3.5–7 Hz), and delta (0.5–3 Hz), bands in line with existing resting state studies measuring resting states in participants with ADHD (Van Dongen-Boomsma et al., 2010; Woltering et al., 2012; Liechti et al., 2013; Buyck and Wiersema, 2014; Alba et al., 2016; Rommel et al., 2017). We also included two ranges of gamma bands from 30 Hz to 40 Hz, reported in this paper as "low gamma," and 40–50 Hz reported in this paper as "high gamma." The mu band was also estimated from 8 Hz to 12 Hz based on existing studies about the motor system (e.g., Perry et al., 2011). We note that the mu and alpha ranges are nearly equivalent, and may be better denoted by region rather than frequency.

Spectral power was computed at all frequency bands in the frontal (Fp1, Fp2, F1, F2, F3, F4, F5, F6, F7, F8, Fz, AF3, AF4, AF7, AF8, FC1, FC2, FC3, FC4, FC5, FC6, FT7, and FT8), centroparietal (C1, C2, C3, C4, C5, C6, CPz, CP1, CP2, CP3, CP4, CP5, CP6, P1, P2, P3, P4, P5, P6, P7, and P8), and occipital (Oz, O1, O2, POz, PO3, PO4, PO7, and PO8) regions during 2 min for each of the eyes-closed and eyes-open conditions. Whole-brain analyses are defined as frequency bands which included all valid electrodes across the scalp, as opposed to specific regions (i.e., frontal, centroparietal, and occipital). Due to a higher degree of noise in some temporal electrodes, as well as some differences in measurement systems between sites (e.g., Iz only used in UK EEG), a temporal region was not assessed. For a cautious approach, these electrodes (electrodes FCz, AFz, Fpz, FT9, FT10, PO9, PO10, P9, P10, TP9, TP10, and Iz) were also removed from the analyses of overall brain activity. In addition, sequences with electric

potentials above 100 mV were rejected during data processing. A frequency extraction was performed for each frequency band by specifying windows at the following frequencies: delta: 0.5–3 Hz; theta: 3.5–7 Hz; alpha: 7.5–12.5 Hz; beta: 13–25 Hz; mu: 8–13 Hz; low gamma: 30–40 Hz; high gamma: 40–50 Hz. Arithmetic means were computed for resulting absolute spectral power values during eyesopen and eyes-closed conditions. For a full picture of activity in this exploratory study, this was considered for the whole brain, as well as frontal, centroparietal, and occipital regions. Data was then extracted from Brain Vision Analyzer for statistical comparison.

2.7 Statistical analysis

Removal of outliers was performed liberally as to not exclude potential meaningful clinical differences. Therefore, values were removed when they were above three standard deviations from the mean and cross-checked with Q–Q plots. This resulted in the removal of 9 values across all participants for all spectral power averages in the eyes-open condition, and 27 values across all participants for all spectral power averages for the eyes-closed condition.

For within-subject comparisons between eyes-open and eyes-closed conditions, a paired-samples t-test was conducted. Given that there are just two conditions (k=2), a Bonferroni correction was not needed [i.e., at 5% significance: 0.05/c; where c=k(k-1)/2, is equal to significance p<0.05]. Group differences in average frequency band activity were calculated with one-way ANOVAs and to determine the more specific group differences, Tukey's $post\ hoc$ test is reported to account for multiple comparisons. Levene's test of unequal variance was performed to account for small group sizes in the nature of the present pilot study. When unequal variances were found, Welch statistic corrections are reported and the Games-Howell $post\ hoc$ tests, which do not assume unique variances, were used.

Given the possibility for intracranial variance (i.e., Hagemann et al., 2008) or other potential confounding factors, we have also included difference scores between eyes-open and eyes-closed conditions (see Supplementary material).

Associations between symptom severity and spectral power were conducted with Pearson correlations. Analyses were conducted in IBM SPSS Statistics, Version 26.0.

3 Results

3.1 Eyes-open versus eyes-closed

By majority, there was a significant increase in spectral power for all frequency bands and in overall activity from the eyes-open condition to the eyes-closed condition (see Table 2).

3.2 Whole-brain spectral power

During the eyes-open condition, alpha and mu frequency bands reached only marginal significance by group [alpha: F(3, 38) = 2.83, p = 0.052, $\eta^2 = 0.182$; Mu: F(3, 38) = 2.68, p = 0.060, $\eta^2 = 0.175$]. Tukey's post hoc test revealed the difference in alpha activity was driven by significantly (p = 0.047) higher values in the DCD group (M = 20.55,

TABLE 2 Frequency band activity compared between conditions of eyes-open and eyes-closed.

Frequency band	Eyes-open <i>M (SD)</i>	Eyes-closed <i>M (SD)</i>	N	Significance value (p)
Alpha	12.38 (9.96)	27.8 (25.14)	39	<0.001
Beta	6.18 (3.00)	8.11 (4.14)	40	<0.001
Delta	20.70 (16.25)	27.14 (17.69)	41	0.002
Theta	10.64 (6.40)	14.44 (8.73)	39	<0.001
Gamma low	4.83 (2.63)	6.60 (3.82)	40	<0.001
Gamma high	5.18 (2.70)	6.01 (3.28)	41	0.011
Mu	12.20 (9.85)	26.74 (23.95)	41	<0.001
Overall	10.49 (7.83)	19.97 (13.59)	40	<0.001

Activity across all participants is reported. Gamma low refers to signals between 30 and 40 Hz, gamma high includes 40-50 Hz.

SD = 15.82) than the ADHD group (M = 6.50, SD = 3.82). There were no significant *post hoc* comparisons for the Mu frequency band. Furthermore, there were no significant differences between groups for alpha, beta, theta, delta, low gamma (30–40 Hz), high gamma (40–50 Hz), mu, and overall brain activation during eyes-open and eyes-closed conditions.

3.3 Frontal cortex

There was a marginally significant group difference for beta activity in the frontal cortex in the eyes-closed condition $[F_{Welch}(3,15.6)=3.11,\ p=0.057]$. The Games-Howell *post hoc* test revealed this effect was driven by a difference (p=0.043) between the DCD (M=14.52,SD=9.11) and typically developing groups (M=5.64,SD=4.19). All other frequency bands across eyes-open and eyes-closed conditions were not significantly different between groups.

3.4 Centroparietal cortex

There were no significant between-group differences for the eyesopen or eyes-closed conditions across all spectral power bands in the central region.

3.5 Occipital cortex

Between-group differences were present in occipital electrodes for beta $[F_{Welch}(3,16.5)=6.86,\ p=0.003]$ and marginally significant for high gamma $[F_{Welch}(3,18.3)=3.01,\ p=0.057]$ spectral power for the eyes-open condition. Differences in beta power were primarily driven by the comparison (p=0.001) between the DCD $(M=4.04,\ SD=1.69)$ and typically developing groups $(M=8.64,\ SD=3.62)$. For high gamma power, Games-Howell post hoc tests showed no group differences. There were no significant group differences in spectral power in the occipital cortex during the eyes-closed condition.

3.6 Symptom severity and spectral power

Several correlations were present between severity of DCD symptoms, via ADC scores, or ADHD symptoms, via ASRS scores

across all participants. However, all correlations were non-significant when p-value adjustments for multiple comparisons were applied. Furthermore, the correlations were not significant when examined with Spearman correlations as a follow-up procedure (see Supplementary material).

4 Discussion

Overall, the present study preliminarily demonstrated that individuals with DCD and/or ADHD as well as typically developing adults may exhibit only a few noteworthy differences or trends in neural activity that may relate to symptoms of one or both conditions. When considering differences by task condition, we confirmed there is a generally consistent pattern of increased spectral power from eyesopen to eyes-closed comparisons in our sample. These findings are in line with previous studies of typically developing individuals (e.g., Barry et al., 2007). This result primarily reflects validity of the data and neurophysiological activity during each condition. Furthermore, significant group differences were present in the occipital region (eyesopen: beta) while only marginally significant differences were present in whole brain activity (eyes-open: alpha, mu), frontal beta with eyesclosed, and occipital high gamma with eyes-open. Some of these differences were driven by specific group comparisons which could reflect baseline differences at rest.

4.1 Increased activity from eyes-open to eyes-closed

We confirmed most frequency bands were consistent with our first expectation that spectral power frequencies would increase from eyes-open to eyes-closed conditions. These findings are in line with some previous studies of typically developing adults (e.g., Barry et al., 2007; Barry and De Blasio, 2017).

4.2 Group differences in resting state activity

There were several noteworthy group differences in spectral power which could relate to unique features of DCD and/or ADHD. Given the preliminary nature of this study, we discuss

potential explanations for both significant and marginally significant findings. While we cannot claim marginally significant findings are true or robust effects, we urge future research to consider their potential and retest these findings to reveal if these are trends toward or away from statistical significance.

While we could not confirm our hypothesis regarding increased theta and delta frequencies in the ADHD group, an interesting trend was present for alpha power. The marginally significant difference in whole brain alpha power during the eyes-closed condition was driven by the DCD versus ADHD group comparison, indicating a potential for overall alpha power to distinguish DCD and ADHD. The role of alpha power is generally dominant and often reduced in individuals with ADHD compared to typically developing participants (Barry and Clarke, 2013; Deiber et al., 2020; Debnath et al., 2021), but we could not replicate this pattern in the present study. Given that alpha power comparisons between adults with and without ADHD have been linked to both hypoactivation and hyperactivation, it is challenging to interpret this result (Deiber et al., 2020). Nonetheless, the relevance of alpha spectral power should be continuously explored in adults with ADHD, DCD, and for its potential as a marker to differentiate DCD and ADHD.

Next, we expected that fronto-central beta activity would significantly differ for the DCD and typically developing groups in particular and aimed to explore the alpha activity given that previous results are few and conflicting (De Castelnau et al., 2008; Keating et al., 2023). Alpha activity only differed marginally and for the DCD versus ADHD group comparison. Therefore, our results do not support that there is a difference in alpha power at rest between those with DCD and typical developing adults, in line with patterns also observed in children (Keating et al., 2023). However, alpha could be relevant in the context of distinguishing DCD and ADHD and should be tested further to determine the possibility.

Furthermore, we found a marginally significant group difference in whole-brain mu activity. Given that (a) no group differences could be found via *post hoc* tests and (b) mu waveforms overlap with alpha and can be challenging to disentangle (Garakh et al., 2020), these results should be interpreted with caution. Future research should examine the mu and alpha waveforms and their potential for regionally specific roles in DCD and/or ADHD.

In addition, we found group differences in line with our expectations such that occipital beta power was significantly increased in DCD compared to typically developing participants but this pattern was only observed at marginal significance for frontal beta power. The latter trend is in line with relevance of frontal beta to DCD noted by de Castelnau et al. (2008). Notably, the previous associations between some cases of ADHD and greater frontal beta rhythms (Kropotov, 2016), are also not identified in the present study. Beta waves broadly reflect a wakeful state with mental activity taking place but can also be related to motor initiation and termination as well as motor planning and inhibition (Kropotov, 2016; Heinrichs-Graham et al., 2017; Barone and Rossiter, 2021).

While the differences in beta found in this study should be interpreted with caution and tested further, there are several explanations we theorize might be linked to beta differences in DCD that should be tested further in future research. In general, beta rhythms have been noted to increase after movement, potentially as a result of the motor system regaining balance, adaptation, or regulation (Heinrichs-Graham et al., 2017). Among typically developing

individuals, increases in post-movement beta activity were greater when movement was stopped suddenly compared to slowly (Heinrichs-Graham et al., 2017). This pattern should also be tested in individuals with DCD. In addition, substantial increases or decreases in post-movement beta activity can reflect motor learning taking place (Barone and Rossiter, 2021). Therefore, it is possible the elevated beta level in DCD could reflect some degree of novelty of the resting state task specific to this group or a unique motor modulation in line with motor difficulties known to coincide with DCD. Furthermore, the beta frequency may reflect the presence or absence of maintaining one's cognitive or sensorimotor state (Engel and Fries, 2010). This could indicate a potential difficulty in the transition from movement to rest or maintenance of rest in the DCD group and required less effort from the typically developing group, who likely find it more natural to sit still or fluidly control their posture than those with DCD (e.g., Geuze, 2005; Miller et al., 2019). Thus, resting state activity may originate in structural differences, functional differences of neural networks or different (cognitive) activity during quiet sitting [also see Wilhelm et al. (2001)].

In our study, it is possible that the chairs at different testing locations could support participants in balancing to different degrees, given that more participants in the DCD group were tested in the UK and more of the typically developing group in Germany. However, both explanations would be supported by a consistent beta difference in the DCD+ADHD group (primarily tested in UK), which was not found in this study. As there are several plausible explanations for the observed increases in beta power in DCD at rest which can only be speculated upon in the present paper, future studies should test various contexts of rest and activity in DCD to support determining whether beta could be a potential biomarker for DCD.

Finally, when considering occipital activity, a significant difference in beta in the eyes-open condition was driven by comparisons between the DCD and typically developing groups, and a marginally significant difference was found in high gamma activity with eyes-open. The differences between the DCD and typically developing groups could potentially reflect a difference in visual attention related to the widely known role of the occipital cortex (Gola et al., 2013). Beta and gamma power are often indicative of wakeful and mentally active states, potentially related to higher order cognitive processing (Başar, 2013; Mably and Colgin, 2018). It is possible that participants with DCD needed to modulate their motor activity in posture and to sit still. This might have resulted in a unique recruitment of occipital beta and gamma power, potentially linked to increased effort and/or attention (Gola et al., 2013).

Overall, there are far more similarities in resting state electrical activity than there are significant differences in the present study. It is possible that differences between DCD and ADHD as well as DCD and typically developing participants observed at the external, behavioral, or subjective levels are often subtle in neurophysiology, especially in adults. By adulthood, symptoms of DCD and ADHD could have already been managed in treatment or compensated for on an individual level (Wilmut, 2017). Therefore, it is not surprising that adults with DCD and ADHD have many similarities in the context of a simpler task, but it is all the more remarkable that several key group differences and potential trends were observed in the present study, especially between DCD and typically developing adults.

4.3 Limitations and future directions

The present study is considered a pilot study and by nature is under-powered. As studies with comparisons of spectral power DCD and/or ADHD have not been previously conducted, the present study also provides a baseline for effect sizes in future related studies in calculating a minimum sample size and general direction for selecting relevant spectral power bands. Furthermore, within the pilot and exploratory context of the study, we provided conservative corrections and tested across multiple sites to increase the sample size for DCD and DCD+ADHD groups in particular. We considered potential group differences between test sites and while demographics were consistent, it should be noted that more participants with DCD were recruited in the UK sample. Our challenge recruiting individuals with DCD in the German sample is suspected to be due to underrecognition of DCD in German clinicians (Meachon et al., 2023). Nonetheless, we replicated the set-up as closely as possible. Furthermore, some differences between sites and groups recruited are inevitably different. For example, intracranial variance could differ between participants (Hagemann et al., 2008) and coincidentally between groups. Although difference scores did not significantly differ between the groups, it could be assumed the groups had comparable change from eyes-open to eyes-closed conditions (see Supplementary material). However, given the small sample size, replication of the present study is necessary to conclusively determine if resting state differences between DCD and/or ADHD are robust.

Another limitation is the order of the tasks consistently beginning with eyes-open trials and ending with eyes-closed in between measurement of other executive functioning task. While other studies have indicated the increase in power from eyes-open to eyes-closed can be found even when the eyes-closed and eyes-open conditions are repeated many times within one study (e.g., Barry et al., 2007), this should still be considered with a randomized design for future studies on DCD and ADHD. In addition, all participants completed resting state trials in an enclosed room without the experimenter present. Therefore, it is possible some participants moved during the task which could not be detected in EEG artifact analysis alone. While there were no demographic differences based on test site and as many features as possible were kept consistent, it is still possible the different testing locations (e.g., chairs) could have had a minor influence on comfort during the rest task.

Finally, causal links cannot be drawn between specific patterns of electrical brain activity and symptoms of DCD and/or ADHD in this study and should be examined in future research. This could be particularly important in future steps toward determining which endophenotypic features are unique to co-occurring versus single-occurring DCD and ADHD.

5 Conclusion

This study provides a foundation for determining the potential for overlap and differentiation of DCD and ADHD through resting state electrical brain activity. Several group differences could be noted in adults with DCD and typical development during seated rest with some potential differences between DCD and ADHD. This suggests that there might be a few fundamental

significant baseline differences unique to DCD which are not present in co-occurring DCD+ADHD or ADHD alone. We theorize, but cannot confirm, that resting state behavior can still engage symptoms in DCD, potentially requiring additional motor load to maintain a seated position. Furthermore, numerous overlaps were observed between groups such that spectral power values were not significantly different in the resting state more often than differences were found. Therefore, it is likely the neural mechanisms between DCD and/or ADHD are generally similar at baseline. This is important for the future assessment of DCD, direction of differentiation of DCD and ADHD, and the interpretation of the resting state in general.

Data availability statement

The datasets presented in this study can be found in online repositories. The names of the repository/repositories and accession number(s) can be found at: https://osf.io/zjws3/ and https://madata.bib.uni-mannheim.de/451/.

Ethics statement

The studies involving humans were approved by University of Mannheim Ethics Committee; Oxford Brookes University Ethics Committee. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

EM: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Software, Validation, Visualization, Writing – original draft, Writing – review & editing. MK: Data curation, Investigation, Methodology, Software, Writing – original draft, Writing – review & editing. KW: Conceptualization, Investigation, Methodology, Resources, Supervision, Writing – original draft, Writing – review & editing. GA: Conceptualization, Investigation, Methodology, Project administration, Resources, Supervision, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2024.1330385/full#supplementary-material

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REVIEWED BY
Giulia Purpura,
University of Milano Bicocca, Italy
Emily Kilroy,
University of Southern California,
United States

*CORRESPONDENCE

Jill G. Zwicker

☑ jill.zwicker@ubc.ca

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Cortical grey matter volume differences in children with developmental coordination disorder compared to typically developing children

Myrah Malik¹, Alexander Weber^{2,3}, Donna Lang^{2,4}, Tamara Vanderwal^{2,5} and Jill G. Zwicker^{2,3,6}*

¹Graduate Programs in Rehabilitation Science, University of British Columbia, Vancouver, BC, Canada, ²Brain, Behaviour, & Development Theme, BC Children's Hospital Research Institute, Vancouver, BC, Canada, ³Department of Pediatrics, University of British Columbia, Vancouver, BC, Canada, ⁴Department of Radiology, University of British Columbia, Vancouver, BC, Canada, ⁵Department of Psychiatry, University of British Columbia, Vancouver, BC, Canada, ⁶Department of Occupational Science & Occupational Therapy, University of British Columbia, Vancouver, BC, Canada

Introduction: The cause of Developmental Coordination Disorder (DCD) is unknown, but neuroimaging evidence suggests that DCD may be related to altered brain development. Children with DCD show less structural and functional connectivity compared to typically developing (TD) children, but few studies have examined cortical volume in children with DCD. The purpose of this study was to investigate cortical grey matter volume using voxel-based morphometry (VBM) in children with DCD compared to TD children.

Methods: This cross-sectional study was part of a larger randomized-controlled trial (ClinicalTrials.gov ID: NCT02597751) that involved various MRI scans of children with/without DCD. This paper focuses on the anatomical scans, performing VBM of cortical grey matter volume in 30 children with DCD and 12 TD children. Preprocessing and VBM data analysis were conducted using the Computational Anatomy Tool Box-12 and a study-specific brain template. Differences between DCD and TD groups were assessed using a one-way ANOVA, controlling for total intracranial volume. Regression analyses examined if motor and/or attentional difficulties predicted grey matter volume. We used threshold-free cluster enhancement (5,000 permutations) and set an alpha level of 0.05. Due to the small sample size, we did not correct for multiple comparisons.

Results: Compared to the TD group, children with DCD had significantly greater grey matter in the left superior frontal gyrus. Lower motor scores (meaning greater impairment) were related to greater grey matter volume in left superior frontal gyrus, frontal pole, and right middle frontal gyrus. Greater grey matter volume was also significantly correlated with higher scores on the Conners 3 ADHD Index in the left superior frontal gyrus, superior parietal lobe, and precuneus. These results indicate that greater grey matter volume in these regions is associated with poorer motor and attentional skills.

Discussion: Greater grey matter volume in the left superior frontal gyrus in children with DCD may be a result of delayed or absent healthy cortical thinning, potentially due to altered synaptic pruning as seen in other neurodevelopmental disorders. These findings provide further support for the hypothesis that DCD is related to altered brain development.

KEYWORDS

developmental coordinator disorder, motor skills disorder, children, MRI, brain structure, voxel-based morphometry, grey matter

1 Introduction

Developmental Coordination Disorder (DCD) is defined by motor abilities that are below expectations for the child's chronological age in the absence of any underlying neurological, visual, or intellectual condition that could better explain the motor difficulties (American Psychiatric Association, 2013). The motor deficit significantly affects activities of daily living, school, work, leisure, and play and can have an adverse impact on mental health and quality of life (Zwicker et al., 2012; Caçola et al., 2016; Zwicker et al., 2017; Izadi-Najafabadi et al., 2019; Karras et al., 2019). The motor difficulties and secondary consequences of DCD often persist into adulthood (Kirby et al., 2014). Children with DCD are more likely than typically developing (TD) children to have attentional difficulties, with over 50% of children with DCD having a co-occurring ADHD diagnosis (Dewey et al., 2002; Fliers et al., 2010).

Neurodevelopmental disorders such as DCD are a heterogenous group of conditions which are thought to be due to impaired growth, development, or function of the central nervous system (CNS) (American Psychiatric Association, 2013). This has led researchers to try to identify brain-based differences in DCD through functional and structural brain imaging studies (Brown-Lum and Zwicker, 2015; Biotteau et al., 2016). Multiple functional studies have identified group-level differences in parietal and frontal regions (Kashiwagi et al., 2009; Zwicker et al., 2010, 2011; McLeod et al., 2014), although these findings have not been consistent. Fewer studies have investigated differences in brain structure. There have been reports of thinner right medial orbitofrontal cortices alongside greater clustering coefficient alterations in the structural connectome of the lateral orbitofrontal cortex in children with DCD (Langevin et al., 2014; Caeyenberghs et al., 2016), but empirical volumetric evidence is sparse in this population. A structural neuroimaging study conducted by Reynolds et al. (2017) found that children with DCD showed significant decrease in grey matter in the frontal lobe of the right hemisphere, and a recent study showed decreased grey matter volume in parts of the cerebellum (Gill et al., 2022). Overall, the number of structural studies in DCD is low, and heterogeneity in sample ages, inclusion criteria, and methodologies used mean there is still much to be learned about structural morphology in children with DCD.

The parietal and frontal lobes have been proposed as one of the correlates of motor impairments in DCD, mainly due to their respective roles in visuospatial information and higher-order cognitive functions (e.g., working memory, organizing/planning). The purpose of this study was to test for potential grey matter volume differences using voxel-based morphometry in children with DCD compared to TD children. We also examined correlations between grey matter volume and clinical measures of motor function and attention difficulties. We hypothesized that children with DCD would have: (1) lower grey matter volume in parietal and frontal regions compared to TD peers; and (2) positive correlations between grey matter volume and motor function and attentional performance.

2 Materials and methods

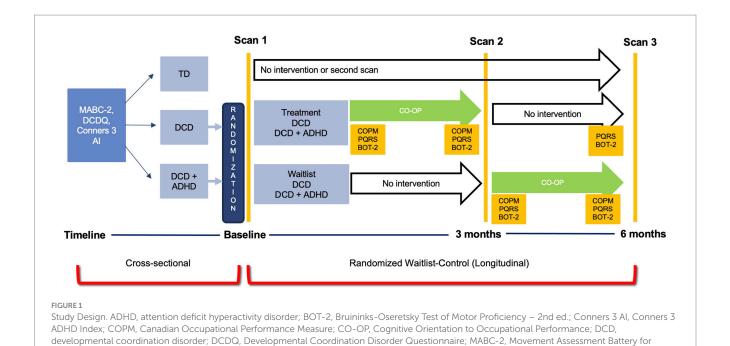
2.1 Study design

The current investigation was part of a larger cross-sectional study and randomized waitlist-control trial that used multiple brain imaging modalities (ClinicalTrials.gov ID: NCT02597751). For the purpose of this analysis, data collected as part of the cross-sectional study were used to investigate differences in grey matter volume in three groups of children: DCD, DCD and co-occurring attention deficit hyperactivity disorder (DCD+ADHD), and TD children (Figure 1). Approval was obtained by UBC Children's and Women's Research Ethics Board (#H14-00397). After screening and recruitment, parents or legal guardians provided written consent and children assented to participate in the study.

2.2 Participants

A convenience sampling method was used to recruit 8-12-years old participants. Children with DCD and DCD+ADHD were recruited from Dr. Zwicker's research-integrated DCD Clinic at Sunny Hill Health Centre for Children, BC Children's Hospital ADHD Clinic, caseloads of occupational and/or physical therapists from Sunny Hill and the Vancouver Regional Pediatric Team, and the community. TD children were recruited through advertisements in Vancouver schools and community centres, and by word-of-mouth.

Children were assessed by a registered occupational therapist or trained graduate student to ensure they met the inclusion criteria of the study. Children with DCD were identified according to the Diagnostic Statistical Manual 5th edition (DSM-5) diagnostic criteria (American Psychiatric Association, 2013): (1) a score ≤ 16th percentile on the Movement Assessment Battery for Children - 2nd edition (MABC-2) (Henderson et al., 2007); (2) a score in the suspected or indicative range on the DCD Questionnaire (DCDQ) (Wilson and Crawford, 2007); (3) parent-reported motor difficulties from a young age; and (e.g., cerebral palsy, criteria, intellectual disability) (4) no other medical condition that could explain motor difficulties as per parent-report, clinical observation, and/or medical exam. For the DCD+ADHD group, all the above were met in addition to parent report of an ADHD diagnosis. Given that attention difficulties are common in children with DCD even without ADHD (Dewey et al., 2002), the Conners 3 ADHD Index was used to measure ADHD symptomatology in all participants (Conners, 2009). The control group (TD children) included children 8-12-years old with no history of motor difficulties and a MABC-2 score ≥ 25th percentile. Exclusion criteria included being born preterm (gestational age<37 weeks) or diagnosed with any other neurodevelopmental disorder, such as autism spectrum disorder. Children assigned to the TD group were excluded if they were diagnosed with ADHD. Additionally, children



with metal in their bodies (e.g., braces) or with a history of claustrophobia were excluded from participation in the study.

Children- 2nd ed.; PQRS, Performance Quality Rating Scale; TD, typically developing children.

2.3 Clinical measurements

The following measures – MABC-2, DCDQ, and Conners 3 ADHD Index – were used to describe the characteristics of the sample for each group.

2.3.1 Movement Assessment Battery for Children2nd edition (MABC-2)

The MABC-2 is designed for children (ages 3 to 16 years old) (Henderson et al., 2007) and is the most widely used measure to identify children with DCD (Blank et al., 2019). The MABC-2 assesses a child's performance in eight motor tasks in three areas of motor performance: (1) manual dexterity; (2) aiming and catching; and (3) balance (Henderson et al., 2007). Raw scores are translated to age-related percentile norms where a lower score indicates greater motor difficulties. The MABC-2 has an internal consistency of α =0.90, excellent test–retest reliability (ICC=0.97) and good factorial and construct validity (Schulz et al., 2011; Wagner et al., 2011; Wuang et al., 2012; Psotta and Abdollahipour, 2017). The assessment takes about 30 min to administer and can be administered by any trained individual.

2.3.2 Developmental Coordination Disorder Questionnaire (DCDQ)

The DCDQ (Wilson and Crawford, 2007) is a parent-completed questionnaire that is used to identify motor impairments in children 5 to 15 years old. Parents compare their child's abilities in 15 activities relative to their TD peers in three different categories: (1) control during movement; (2) fine motor/handwriting; and (3) general coordination. A higher score indicates better motor performance on

a scale of 15 to 75. In this study, age-specific cut-off scores were used as specified in the DCDQ manual. The DCDQ has high internal consistency (α =0.94) and adequate sensitivity (85%) (Wilson et al., 2000; Cairney et al., 2008; Wilson et al., 2009). The DCDQ is the recommended screening tool for DCD according to the international guidelines for identification of children with DCD (Blank et al., 2019).

2.3.3 Conners 3 ADHD Index (Conners 3 AI)

The Conners 3 ADHD Index is parent-completed questionnaire that aids health care professionals in determining whether a child does or does not have ADHD symptoms (Conners, 2009). This norm-referenced assessment is based on a large North American sample. It is one of the most commonly used screening tools to assess ADHD symptoms in both research and clinical settings (Conners, 2009). A score over 70 indicates clinically significant attentional difficulties. The Conners 3 ADHD Index has high internal consistency (α =0.90), high predictive value, and mean test–retest reliability of 0.83 (Morales-Hidalgo et al., 2017). For the purpose of this study, the Conners 3 ADHD Index was used to quantify the degree of attentional difficulties; higher scores indicate poorer attentional performance.

2.3.4 Sociodemographic questionnaire

A socio-demographic questionnaire was used to collect information regarding participant demographics such as age, sex, history of therapy interventions, medications, and additional diagnoses.

2.4 Neuroimaging measures

2.4.1 MRI data acquisition

All brain images were acquired at the Magnetic Resonance Imaging (MRI) Research Facility at BC Children's Hospital Research Institute in Vancouver, Canada. All children participated in an MRI

safety screening and an MRI simulator session to familiarize themselves with the scanning environment (noise, confined space, and head coil). They were also provided with strategies from the research team to help reduce potential anxiety. High resolution isotropic structural scans were obtained on a 3-Tesla General-Electric Discovery MR750 MRI scanner. A T1-weighted 3D structural scan was acquired with the following parameters: three-dimensional spoiled gradient recalled acquisition in steady state (3D SPGR), echo time = 30 ms, repetition time = 3,000 ms, FOV = 256, matrix size = 256 \times 256, flip angel = 12°, number of slices = 256, slice thickness = 1 mm, interleaved with no gaps (voxel size 0.9375 \times 0.9375 \times 1 mm). T1-weighted scans were ascertained to permit reliable segmentation of tissues (grey matter, white matter, and cerebrospinal fluid) and reliable identification of underlying regions (Lerch et al., 2017).

2.4.2 Image quality control

All scans were visually inspected for truncation, motion, aliasing-related and other artifacts by trained raters (Krupa and Bekiesińska-Figatowska, 2015; Reuter et al., 2015). Specifically, image quality was assessed for head coverage, wrapping artifact, radiofrequency noise, signal inhomogeneity, susceptibility artifact, and ringing artifact (Reuter et al., 2015). An ordinal score was given to each image based on motion artifacts and image quality (pass, questionable, or fail) using standardized methodology (Harvard Center for Brain Science, 2014). Two trainees assessed the scans independently; the level of agreement for the categorization of each scan assessed by each trainee was 96%. Only scans that passed the final quality check from both trainees were included in the analysis.

Additionally, quantitative measures of motion were calculated using the software package MRIQC (Esteban et al., 2017). In particular, we measured coefficient of joint variation (CJV), where higher values are related to the presence of heavy head motion and large intensity non-uniformity (Ganzetti et al., 2016).

2.4.3 Voxel-based morphometry

2.4.3.1 Image pre-processing

Data were converted from DICOM (Digital Imaging and Communications in Medicine) to NIfTI (Neuroimaging Informatics Technology Initiative) using the dcm2nii tool from MRIcron.¹ T1 images were processed using voxel-based morphometry (VBM), a computational technique that measures differences in grey matter volume through a voxel-wise comparison (Ashburner and Friston, 2000; Whitwell, 2009). All pre-processing and VBM data analysis were carried out using the Computational Anatomy Tool Box (CAT12, v1742, The Structural Brain Mapping Group, Jena, Germany, http:// dbm.neuro.uni-jena.de/cat12/), through Statistical Parametric Mapping 12 software (SPM12, v7771, The Wellcome Centre for Human Neuroimaging, London, United Kingdom, https://www.fil. ion.ucl.ac.uk/spm/) in MATLAB R2020b (Mathworks, Natick, Massachusetts, United States). For image pre-processing, all T1 images were manually registered to the anterior commissure at the origin of the Montreal Neurological Institute (MNI) coordinate system (Jahn, 2019). The co-registered images were then segmented into grey matter (GM), white matter (WM), and cerebrospinal fluid (CSF). As the images were from a pediatric sample, the tissue probability maps of GM, WM, and CSF were obtained using the Template-O-Matic Toolbox (TOM8, http://dbm.neuro.uni-jena.de/software/tom/). All images were included if their weight average Image Quality Rating (IQR) was greater than 80%, corresponding to a "good" image quality. Mean correlations between all volumes were visualized through CAT12. Volumes with a correlation below two standard deviations from the sample mean were again visually inspected for artifacts.

Next, good quality affine-registered white and grey matter tissue segments were extracted to construct a customized Diffeomorphic Anatomical Registration Through Exponentiated Lie Algebra (DARTEL) study-specific template registered to the MNI-International Consortium for Brain Mapping (ICBM) space. This alternative to the adult-based template provided by CAT12 was used to achieve a more accurate inter-participant registration to improve the realignment of small inner structures for an overall better segmentation (Good et al., 2001; Yassa and Stark, 2009). This additional step was based on pediatric VBM studies done in other neurodevelopmental disorders that created a study-specific average template for their sample (Reynolds et al., 2017; Wang et al., 2017; Sáenz et al., 2020). Individual images were corrected for bias-field inhomogeneities and segmented into GM, WM, and CSF. The images were then normalized using affine spatial normalization and a further modulation was applied to convert the voxel values of tissue concentration (density) to measures of volume. Finally, the normalized GM maps were smoothed with an isotropic Gaussian kernel (full width at half maximum = 6 mm). Total intracranial volume (TIV) was calculated from the GM, WM, and CSF images for each participant using CAT12 module "Total intracranial volume." Figure 2 provides a schematic of the modified VBM pipeline.

2.4.3.2 Computational anatomy toolbox (CAT12)

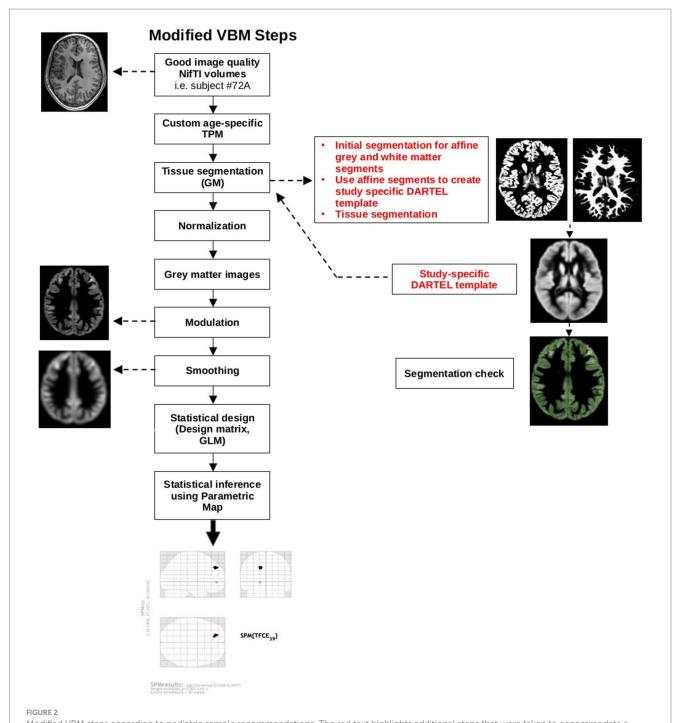
The Structural Brain Mapping Group at the University of Jena (Jena, Germany) designed the automatic and easy-to-use toolbox CAT12 as an extension to the SPM software. CAT12 follows a standard VBM analysis pipeline similar to VBM8. Since our sample's IQR ranged from 80 to 90%, we used segmentation through SPM's extension CAT12 rather than FreeSurfer or FSL as SPM produces a more robust segmentation for those with limited image quality (Fellhauer et al., 2015). When compared to previous toolboxes, CAT12 provided a more accurate and robust volumetric analysis (Farokhian et al., 2017) and advanced segmentation tool (Tavares et al., 2020). It has also been used in neurodevelopmental disorders that commonly co-occur with DCD (Wang et al., 2017; Mei et al., 2020; Sáenz et al., 2020) where the workflow was adapted to accommodate a pediatric population as recommended for VBM analysis.

2.5 Statistical analysis

2.5.1 Participant characteristics

Participant characteristics were analysed using Jeffreys's Amazing Statistics Program (JASP https://jasp-stats.org/). The Chi-squared test was used to compare sex distribution between groups. To compare group differences in age, TIV, MABC-2 (motor measure), Conners 3 ADHD Index (attentional difficulties measure), and DCDQ, we used two-tailed Mann–Whitney-U test since Levene's test and

¹ https://www.nitrc.org/projects/mricron



Modified VBM steps according to pediatric sample recommendations. The red text highlights additional steps that were taken to accommodate a pediatric sample. DARTEL, Diffeomorphic Anatomical Registration Through Exponentiated Lie Algebra; GM, grey matter; GLM, general linear model; NIFTI, Neuroimaging Informatics Technology Initiative; SPM, Statistical Parametric Map; TPM, tissue probability map.

Shapiro–Wilk indicated violation of assumptions of equal variance (p<0.001) and normality (p<0.001), respectively.

2.5.2 VBM statistical analysis

All statistical models were designed with general linear modeling through SPM. Individual participant smoothed grey matter volumes were entered into a second level analysis to estimate differences between DCD vs. TD group using a one-way Analysis of Variance

(ANOVA) design. TIV (centered to overall mean) was used as a covariate/nuisance variable as recommended in VBM analysis to account for inter-participant differences. While a two-sample t-test was inputted into the statistical design, the output was an ANCOVA, with TIV entered as a covariate. No significant differences between age (p=0.40) or sex (p=0.15) were observed between groups. Subsequently, these variables were not included as covariates in the analysis to conserve degrees of freedom. Threshold-Free Cluster

Enhancement (TFCE) thresholding was conducted using the TFCE Toolbox Version $r214^2$ with 5,000 permutations (Smith method) with unequal variance (DCD vs. controls) with an E=0.5 and H=0.2. Structural images were analyzed using TFCE due to its increased sensitivity compared to voxel-or cluster-based statistics (Smith et al., 2009; Salimi-Khorshidi et al., 2011; Radua et al., 2014). Statistical significance was assessed with the permutation test included in SPM.

Initially, we had planned a regression analysis to examine if MABC-2 and Conners 3 ADHD Index scores predicted grey matter volume; however, MABC-2 and Conners 3 ADHD Index scores were moderately negatively correlated (r = -0.66, p < 0.001). Instead, two independent regression analysis were used to examine the relationship between grey matter volume and clinical measures of motor function (MABC-2) and attention difficulties (Conners 3 ADHD Index), respectively, while controlling for the effect of intracranial volume.

TFCE and an alpha level of 0.05 were used to help account for type 1 errors. All results are reported with TFCE thresholding; however, they are uncorrected for multiple comparisons (no $p_{FDR-corrected}$ or $p_{FWE-corrected}$) due to the small sample size. Results are presented at p < 0.001 with cluster size threshold at 50 voxels. Cluster size threshold was based on current literature regarding cluster thresholding. Given our N < 50, we opted for a more stringent cluster threshold of 50 compared to lower thresholds of 10 (Lieberman and Cunningham, 2009; Woo et al., 2014). This is also comparable to previous publications of cerebellar VBM with samples of children with neurodevelopmental disabilities (D'Mello et al., 2015).

3 Results

3.1 Final sample

This study recruited 115 children (TD=35; DCD=80), from whom 73 were excluded because they either declined to participate (n=4), were later determined to have exclusionary diagnoses or to have been born preterm (n=11), did not meet inclusion criteria (n=1), or had insufficient data quality for VBM analysis (n=57) (Figure 3). Due to the smaller than anticipated sample size, the DCD (n=15) and DCD+ADHD (n=15) groups were combined; they did not differ significantly in terms of age, sex distribution, MABC-2 subtest and total scores, or Conners 3 ADHD Index scores (all p>0.05). Our final sample included 30 children with DCD [mean (SD) age: 9.9 (1.5) years] and 12 TD children [mean (SD) age: 10.3 (1.5) years]. The majority of participants (74%) were male (Table 1).

Children (both TD and DCD) whose data were excluded due to motion had an average CJV of 0.73 (\pm 0.13 SD), while those that were kept had an average CJV of 0.60 (\pm 0.09 SD). These values were significantly different [p <0.001; 95%CI=(0.09, 0.16)]. Of the participants that were included for analysis, there was no difference in CJV between the TD and DCD cohorts. Furthermore, for the children included for analysis, no correlation was found between CJV and MABC-2 scores [95%CI=(-0.26, 0.06)].

3.2 Participant characteristics

Demographic and behavioral characteristics of the sample are shown in Table 1. As expected, the mean total MABC-2 score was significantly lower in children with DCD compared to the typically developing group, indicating significant motor impairments. In addition, the DCD group had significant attentional difficulties (poorer attentional performance) as indicated by a mean score over 70 on the Conners 3 ADHD Index. This finding is consistent with the literature which suggests children with DCD have significant attentional difficulties and high rates of ADHD (Dewey et al., 2002; Kadesjö and Gillberg, 2008; Goulardins et al., 2015; Lange, 2018). Lastly, our DCD sample included 24 males (80%), which aligns with DCD having a higher prevalence in males compared to females (American Psychiatric Association, 2013).

3.3 Grey matter differences between TD vs. DCD

Compared to typically developing children, children with DCD had significantly greater grey matter [cluster size (k) >50, $p_{uncorrected} \le 0.001$] in the left superior frontal gyrus (Table 2; Figure 4). There were no regions where children with DCD had lower grey matter volume compared to typically developing children [cluster size (k) <50].

3.4 Grey matter correlates: motor function and attentional performance

MABC-2 scores were negatively correlated [cluster size (k) >50, p_{uncorrected} \le 0.001] with grey matter volume in the left superior frontal gyrus, left frontal pole, and right middle frontal gyrus (Table 3 and Figure 5). Lower MABC-2 scores were related to greater grey matter volume. There were no regions where children with DCD had greater grey matter volume with higher MABC-2 scores [cluster size (k) <50]. The additional clusters (left frontal pole and right middle frontal gyrus) did not overlap with the DCD>TD contrast mentioned above.

The Conners 3 ADHD Index T-score was positively correlated with grey matter volume in the left superior frontal gyrus (cluster size (k) >50, p_{uncorrected} < 0.001) (Table 4 and Figure 6), indicating that higher Conners 3 ADHD Index T-scores (greater attentional difficulties/poorer attentional performance) were related to greater grey matter volume. There were no regions where children with DCD had lower grey matter volume with higher Conners ADHD index T-score [cluster size (k) <50]. The additional clusters (left superior parietal lobe and left precuneus) did not overlap with the DCD > TD contrast.

4 Discussion

This study examined grey matter differences in children with DCD compared to typically developing children. Contrary to our hypothesis, we found that children with DCD had greater grey matter volume compared to TD children. This difference was only found in the left superior frontal gyrus. This result may be clinically significant, as lower MABC-2 scores were significantly correlated with greater

² http://dbm.neuro.uni-jena.de/tfce/

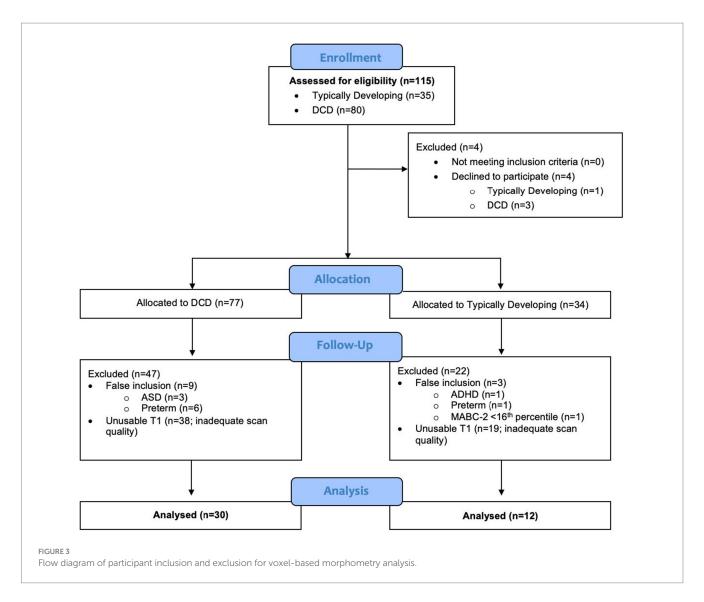


TABLE 1 Description of Cohort (N = 42).

Clinical characteristics	DCD (N = 30) N (%) or Mean (SD)	TD (N = 12) N (%) or Mean (SD)	p-value
Male	24 (80)	7 (58)	0.15
Age at MRI (years)	9.9 (1.5)	10.3 (1.5)	0.40
MABC-2 (percentile)	6.1 (7.4)	64.2 (25.5)	< 0.001
Conners 3 ADHD Index (t-scores)	84.8 (9.7)	56.5 (11.7)	<0.001
Total intracranial volume (L)	1.53 (0.17)	1.52 (0.08)	0.98

ADHD, attention deficit hyperactivity disorder; DCD, developmental coordination disorder; L, litres; MABC-2, Movement Assessment Battery for Children -2nd edition; MRI, magnetic resonance imaging; SD, standard deviation.

grey matter volume in this region, and the same relationship was identified in left frontal pole and right middle frontal gyrus. Greater grey matter volume was also significantly correlated with higher Conners 3 ADHD Index in several regions of the left hemisphere: superior frontal gyrus, superior parietal lobe, and precuneus. These results indicate that greater grey matter volume in these regions is

TABLE 2 MNI coordinates for significantly greater grey matter volume in children with developmental coordination disorder compared to typically developing children.

Location	Х	Y	Z	TFCE	p _{uncorrected}	Cluster size
Left superior	-16	55	22	666.1	0.001	51
frontal gyrus	-16	63	16	599.8	0.001	

associated with poorer motor skills and worse attentional problems. Our findings do not align with previous structural MRI studies in DCD. Langevin et al. (2014) reported thinner cortex in the right temporal pole and Reynolds et al. (2017) identified smaller grey matter volume in the right frontal lobe, specifically the middle, medial, and superior frontal gyri in children with DCD. These disparate findings may be due to methodological differences between studies. For example, we used a robust VBM analysis (CAT12) and modified pipeline to accommodate a pediatric sample (e.g., Wang et al., 2017). Likewise, Langevin et al. (2014) included participants aged 8 to 17 years, which was a broader age range compared to this study and may have different results due to more variance from brain development across such a broad age range. To discuss our results,

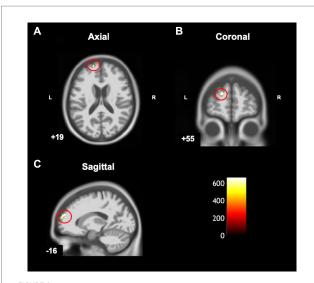


FIGURE 4 Statistical parametric map superimposed on CAT T1 IXI555 template shows between-group differences with significantly greater grey matter volume (yellow-white region) in children with developmental coordination disorder in comparison to typically developing children (ρ < 0.001 uncorrected). Left superior frontal gyrus in (A) Axial; (B) Coronal; and (C) Sagittal view. Color bar shows t-values post threshold-free cluster enhancement analysis.

TABLE 3 MNI coordinates for correlations between grey matter volumes and MABC-2 percentile scores.

Location	Х	Y	Z	TFCE	p _{uncorrected}	Cluster size
Left superior frontal gyrus	-14	64	16	665.63	0.001	85
Left frontal pole	-22	69	16	654.00	<0.001	
Right middle	35	46	28	504.03	0.001	81
frontal gyrus	31	39	24	358.29	< 0.001	

we will first highlight typical brain development and then interpret our findings in that context.

Cortical thickness, surface area, and volume are all generally shown to increase from early infancy (Gilmore et al., 2011; Lyall et al., 2014) until late childhood/early adolescence (Giedd et al., 1999; Lenroot et al., 2007; Wierenga et al., 2014; Tamnes et al., 2017). Following this developmental period, normal brain development is characterized by reductions in cortical thickness, grey matter, and surface area, with further thinning and decreases throughout adolescence (Tamnes et al., 2009; Gogtay and Thompson, 2010; Walhovd et al., 2016; Tamnes et al., 2017). Cortical thinning, which has region-specific trajectories, is a hallmark of brain development and evolution (Sowell et al., 2007; Amlien et al., 2014; Tamnes et al., 2017). It is defined as "the decline in thickness of outer layers of the brain that are most evolutionarily advanced in humans and are thought to play particularly important roles in higher levels of information processing and orchestrating actions" (Spear, 2013, p. 3). Tamnes et al. (2017) suggest that cortical thinning is the primary contributor to cortical volume reductions, as surface area exhibits relatively smaller decreases with age. Synaptic pruning and myelination are considered to be two contributors to the complex process of cortical thinning of grey matter that occurs in healthy brain development (Tau and Peterson, 2009; Spear, 2013).

Knowledge about typical brain development is needed to understand brain development in neurodevelopmental disorders, which are characterized by impaired growth, development, or function of the central nervous system (American Psychiatric Association, 2013). Abnormalities in cortical volume and thickness have been reported in a number of neurodevelopmental disorders, including ADHD and ASD (Castellanos et al., 2002; Makris et al., 2006; Nakao et al., 2011; Ha et al., 2015; Lange et al., 2015; Khundrakpam et al., 2017; Liu et al., 2017; Boedhoe et al., 2020; Sáenz et al., 2020). A delay or dysfunction in cortical thinning might explain the anomalies in surface area, volume, and thickness seen in these other disorders (Shaw et al., 2007, 2011; Khundrakpam et al., 2017). Here, we observed higher regional brain volumes in DCD, which we interpret along these lines, and as a dysfunction in cortical thinning. We would also posit that when combined with previous findings, synaptic pruning is the more likely underlying factor in this population. Recent diffusion tensor imaging studies reported white matter differences in DCD relative to typically developing peers (Brown-Lum et al., 2020). The authors found no accompanying differences in radial diffusivity, leading them to conclude that the differences in DCD were unlikely to be related to disrupted myelination (Brown-Lum et al., 2020). Kilroy et al. (2022) also reported no differences in radial diffusivity in DCD compared to TD children. Since typical brain development is associated with increased myelination and synaptic pruning, the combined evidence from the current study (i.e., greater cortical volume in a specific region) and diffusion tensor imaging studies (i.e., unlikely disruption in myelination) (Brown-Lum et al., 2020; Kilroy et al., 2022) suggest that the delay in cortical thinning in children with DCD is likely due to dysfunction or delay in mechanisms responsible for synaptic pruning. Synaptic pruning, which happens between early childhood to adulthood, is defined as the targeted elimination of less functional or extra synapses to improve connections in the brain and is necessary for normal brain development (Lichtman and Colman, 2000; Tessier and Broadie, 2009; Paolicelli et al., 2011; Navlakha et al., 2015; Sakai, 2020). The more a particular synapse is used, the stronger it becomes, which decreases the likelihood of it being eliminated; weaker connections are more susceptible to synaptic pruning (Lichtman and Colman, 2000; Tessier and Broadie, 2009; Paolicelli et al., 2011; Navlakha et al., 2015; Sakai, 2020).

Greater grey matter volume was located in the frontal lobe in children with DCD, specifically in the left superior frontal gyrus. The left superior frontal gyrus is involved in activities that support higher cognitive functions, such as the processing of sensory and motor information (Exner et al., 2002), executive function (e.g., working memory, planning) (Hoffmann, 2013), and spatial cognition (Hopfinger et al., 2000; du Boisgueheneuc et al., 2006; Harms et al., 2013). These functions are consistent with the difficulties reported in children with DCD (Wilson and McKenzie, 1998; Alloway and Temple, 2007; Leonard et al., 2015; Fong et al., 2016; Wilson et al., 2020). Though we were somewhat surprised by the focal nature of this finding, the known functions of the region relate strongly to the clinical picture in DCD. Greater grey matter volume in the left

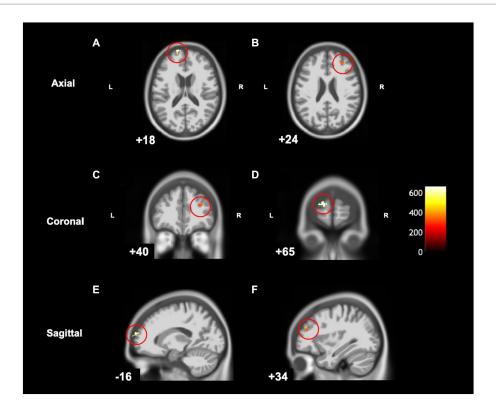


FIGURE 5

Statistical parametric map superimposed on CAT T1 IX1555 template shows significant negative correlations (yellow and orange) between grey matter and MABC-2 total percentile scores (p<0.001 uncorrected). (**A**) Left superior frontal gyrus and frontal pole; (**B**) Right middle frontal gyrus; (**C**) Right middle frontal gyrus; (**D**) Left frontal pole; (**E**) Left superior frontal gyrus and frontal pole; (**F**) Right middle frontal gyrus. Color bar shows t-values post threshold-free cluster enhancement analysis.

TABLE 4 MNI coordinates for correlations between grey matter volume and Conners 3 ADHD Index T-Score.

Location	Х	Y	Z	TFCE	P _{uncorrected}	Cluster size
Left superior	-16	43	37	1192.72	< 0.001	663
frontal gyrus	-25	60	24	1018.14	< 0.001	
	-29	51	37	935.97	< 0.001	
Left superior parietal lobe and left precuneus	-13	-65	46	721.47	<0.001	61

superior frontal gyrus could reflect altered brain development in this specific region, perhaps due to a delay or disruption of synaptic pruning as discussed above; however, future studies are required to confirm this hypothesis.

In addition to the left superior frontal gyrus, motor function was correlated with the right middle frontal gyrus and left frontal pole. The right middle frontal gyrus is suggested to play an important role in re-orientating attention to different environmental stimuli (Japee et al., 2015), where attention plays an important role in motor learning (Song, 2019). The frontal pole cortex, also known as Brodmann area 10, is important in monitoring the outcomes of movements/actions (Tsujimoto et al., 2011). These findings are consistent given the

attentional (Dewey et al., 2002; Fliers et al., 2010) and motor learning/planning difficulties in children with DCD (Wilson et al., 2012). A delay or dysfunction in cortical thinning (through decreased pruning) may underlie the greater grey matter in these regions associated with lower MABC-2 scores.

The findings regarding attentional scores follow a similar explanation. Worse attentional symptomatology was correlated with greater grey matter volume in the superior frontal gyrus (discussed above), as well as the superior parietal lobe and precuneus. The superior parietal lobe is involved in manipulating information in working memory (Koenigs et al., 2009) and sensorimotor integration (Wolpert et al., 1998). The precuneus is part of the parietal cortex and is involved in a wide variety of cognitive processes, including internally guided attention and shifting attention in motor imagery tasks (Cavanna and Trimble, 2006). The greater grey matter volume in these regions may be a result of decreased synaptic pruning. In addition to structural differences, a recent study by Rinat et al. (2020) reported altered functional connectivity between the sensorimotor network and the posterior cingulate cortex and precuneus in children with DCD, providing further evidence that these regions are implicated in DCD. Greater grey matter volume has also been observed in ASD (Tang et al., 2014), a common co-occurrence with DCD. In addition, the precuneus has been implicated in other neurodevelopmental disorders that have difficulty with attention (Nakao et al., 2011; Sáenz et al., 2020). Animal models suggest that the consequences of excessive synaptic connections due to a failure of synaptic pruning impairs

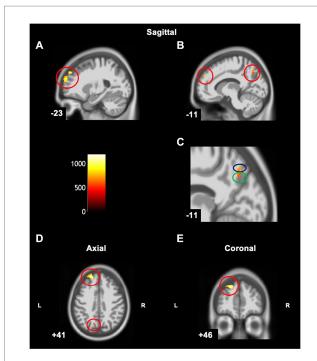


FIGURE 6 Statistical parametric map superimposed on CAT T1 IXI555 template shows significant positive correlations (yellow and orange) between grey matter and Conners ADHD Index T-Score (p < 0.001 uncorrected). (A) Left superior frontal gyrus; (B) Left superior frontal gyrus, superior parietal lobe, and precuneus; (C) Zoomed image of B: Left superior parietal lobe (blue) and left precuneus (green); (D) Left superior frontal gyrus; (E) Left superior frontal gyrus. Color bar shows t-values post threshold-free cluster enhancement analysis.

learning new spatial re-orientations (Afroz et al., 2016); these findings suggest that too many brain connections (synapses) may put limitations on learning potential (Eltokhi et al., 2020) which is consistent with the difficulties presented in children with DCD (Alloway and Archibald, 2008; Tsai et al., 2012).

We would also point out that the ADHD-related findings in this DCD group do not align with many studies in "stand-alone" ADHD, which show greater cortical thinning in prefrontal and frontolimbic regions (e.g., Francx et al., 2016). The findings here may thus be specific to the combined circuitry affected in individuals with both ADHD and DCD, and needs replication. This is also interesting from a transdiagnostic research perspective, perhaps illustrating that across some disorders (and possibly even at some ages), the neural correlates associated with some symptom domains may be unique and not transdiagnostic.

There are several limitations in this study. First, our sample size was much smaller than anticipated so we were unable to control for multiple comparisons. After applying exclusion criteria and stringent quality checks of the 111 scans, our final sample was relatively small (N=42) and unequal (DCD was nearly double the size of the non-DCD cohort). However, only the highest quality scans were included which increases confidence in the findings and generalizability of the results (Sáenz et al., 2020). Further, we had intended to analyze children with DCD and children with DCD+ADHD separately, but due to the smaller than anticipated sample size, we combined the children into one group. However, the

majority of children in our sample had clinically significant ADHD symptoms (regardless of diagnosis), which may have minimized the anticipated group differences. We noted that 5/15 (33%) of children with DCD+ADHD were taking stimulant medication which may have confounded the results, particularly if they had been taking the stimulants for long periods of time (Nakao et al., 2011). In addition, there were some limitations regarding volume-based measures. Since grey matter includes surface area and thickness, each of which have their own developmental trajectories, the interpretation of grey matter volume becomes difficult without examining surface area or thickness individually (Frye et al., 2010). Future studies should continue to explore differences in grey matter volume in children with DCD but in a larger sample and over time to examine if maturation differs from typically developing children. In addition, exploring cortical thickness and volume in the same study would provide more insight into the structural morphology associated with DCD. Likewise, results could be stratified by age, sex and/or medication use to provide further insights (Caviness et al., 1996; De Bellis et al., 2001; Spencer et al., 2013). More longitudinal studies from childhood through adolescence evaluating cortical thickness, volume, and surface area in this population are needed to better delineate structural morphology in DCD. Lastly, to further explore mechanisms of synaptic pruning, animal and molecular studies should be conducted to examine the underlying behavioral and neurological consequences of altered synaptic pruning in this population.

In conclusion, we found that children with DCD had greater grey matter volume in the left superior frontal gyrus, and that greater grey matter volume in this region and other frontal and parietal regions was associated with poorer motor and attentional skills. These findings support the conceptualization of DCD as a neurodevelopmental disorder, as in general, cortical thinning is associated with healthy development and advances in skills and aptitudes. We hypothesize that the greater grey matter volume in superior frontal gyrus may reflect a delay or absence of healthy cortical thinning in DCD, potentially due to altered synaptic pruning as seen in other neurodevelopmental disorders. This study adds to growing evidence that DCD may be related to altered brain development. Additional new research will be needed to determine what factors influence brain development in children with DCD, and which risk factors may be modifiable to potentially prevent this common motor disorder.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by Children's and Women's Health Centre/University of British Columbia Research Ethics Board. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin and participating children assented to take part in this study.

Author contributions

MM: Formal analysis, Visualization, Data curation, Validation, Writing – original draft. AW: Formal analysis, Visualization, Writing – review & editing. DL: Formal analysis, Writing – review & editing. TV: Formal analysis, Writing – review & editing. JZ: Formal analysis, Visualization, Conceptualization, Funding acquisition, Methodology, Project administration, Resources, Supervision, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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EDITED BY Emily J. Meachon, University of Basel, Switzerland

REVIEWED BY
Christian Hyde,
Deakin University, Australia
Deirdre R. Dawson,
University of Toronto, Canada
Brittany Travers,
University of Wisconsin-Madison,
United States

*CORRESPONDENCE
Jill G. Zwicker

☑ jill.zwicker@ubc.ca

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Changes in cortical grey matter volume with Cognitive Orientation to daily Occupational Performance intervention in children with developmental coordination disorder

Myrah Anum Malik¹, Alexander Mark Weber^{2,3}, Donna Lang^{2,4}, Tamara Vanderwal^{2,5} and Jill G. Zwicker^{2,3,6}*

¹Graduate Programs in Rehabilitation Science, University of British Columbia, Vancouver, BC, Canada, ²Brain, Behaviour, and Development Theme, BC Children's Hospital Research Institute, Vancouver, BC, Canada, ³Department of Pediatrics, University of British Columbia, Vancouver, BC, Canada, ⁴Department of Radiology, University of British Columbia, Vancouver, BC, Canada, ⁵Department of Occupational Science and Occupational Therapy, University of British Columbia, Vancouver, BC, Canada

Introduction: Cognitive Orientation to daily Occupational Performance (CO-OP) is a cognitive-based, task-specific intervention recommended for children with developmental coordination disorder (DCD). We recently showed structural and functional brain changes after CO-OP, including increased cerebellar grey matter. This study aimed to determine whether CO-OP intervention induced changes in cortical grey matter volume in children with DCD, and if these changes were associated with improvements in motor performance and movement quality.

Methods: This study is part of a randomized waitlist-control trial (ClinicalTrials. gov ID: NCT02597751). Children with DCD (N=78) were randomized to either a treatment or waitlist group and underwent three MRIs over 6 months. The treatment group received intervention (once weekly for 10 weeks) between the first and second scan; the waitlist group received intervention between the second and third scan. Cortical grey matter volume was measured using voxel-based morphometry (VBM). Behavioral outcome measures included the Performance Quality Rating Scale (PQRS) and Bruininks-Oseretsky Test of Motor Proficiency-2 (BOT-2). Of the 78 children, 58 were excluded (mostly due to insufficient data quality), leaving a final N=20 for analyses. Due to the small sample size, we combined both groups to examine treatment effects. Cortical grey matter volume differences were assessed using a repeated measures ANOVA, controlling for total intracranial volume. Regression analyses examined the relationship of grey matter volume changes to BOT-2 (motor performance) and PQRS (movement quality).

Results: After CO-OP, children had significantly decreased grey matter in the right superior frontal gyrus and middle/posterior cingulate gyri. We found no significant associations of grey matter volume changes with PQRS or BOT-2 scores.

Conclusion: Decreased cortical grey matter volume generally reflects greater brain maturity. Decreases in grey matter volume after CO-OP intervention were in regions associated with self-regulation and motor control, consistent with

our other studies. Decreased grey matter volume may be due to focal increases in synaptic pruning, perhaps as a result of strengthening networks in the brain via the repeated learning and actions in therapy. Findings from this study add to the growing body of literature demonstrating positive neuroplastic changes in the brain after CO-OP intervention.

KEYWORDS

developmental coordinator disorder, motor skills disorder, children, CO-OP, rehabilitation, MRI, brain structure, voxel-based morphometry

1 Introduction

Developmental coordination disorder (DCD) is classified as a neurodevelopmental disorder in the Diagnostic and Statistical Manual 5th edition (DSM-5). This motor disorder affects approximately 450,000 Canadian school-aged children (American Psychiatric Association, 2013; Statistics Canada, 2021). The related gross and fine motor difficulties affect important childhood activities such as tying shoelaces, printing, or riding a bicycle (Kirby and Sugden, 2007; Zwicker et al., 2012; Blank et al., 2019). Early intervention is important, as children with DCD typically continue to experience motor difficulties well into adolescence and adulthood if adequate intervention is not provided throughout childhood (Kirby et al., 2013).

Traditionally, interventions have been process-oriented and focused on addressing the sensorimotor dysfunction that was thought to contribute to their motor impairments (Polatajko et al., 2001; Mandich et al., 2002; Polatajko and Mandich, 2004). Newer approaches leverage current theories of cognitive and motor learning and advocate for problem-solving focused intervention (Sugden, 2007). One such intervention, the Cognitive Orientation approach to daily Occupational Performance (CO-OP), was developed by occupational therapists in Canada (Polatajko et al., 2001). This task-specific intervention is a cognitive-based, problem-solving approach that uses verbal mediation and identifies strategies to support motor skill acquisition (Polatajko et al., 2001). Several systematic reviews have been conducted that further demonstrate the effectiveness of CO-OP intervention for children with DCD (Smits-Engelsman et al., 2012, 2018; Scammell et al., 2016; Yu et al., 2018), making CO-OP one of the recommended treatments in the international clinical practice guidelines for DCD (Blank et al., 2019).

While CO-OP has been deemed effective, the underlying mechanisms or neural bases for clinical improvements were unknown. Our research group recently used magnetic resonance imaging (MRI) to investigate brain changes associated with CO-OP intervention. In a study that focused on the cerebellum, we showed increases in grey matter volume in the brainstem and in cognitive (right crus II) and motor regions (right and left lobule VIIIb and lobule IX) of the cerebellum following the intervention (Gill et al., 2022). Improvements in actual movement performance predicted the increases in cerebellar grey matter volume. In addition, increased functional connectivity in the default mode network and right anterior cingulate cortex were observed after CO-OP intervention (Izadi-Najafabadi et al., 2022b), as well as improved white matter microstructure in several regions, including the bilateral anterior thalamic radiations, bilateral sensorimotor

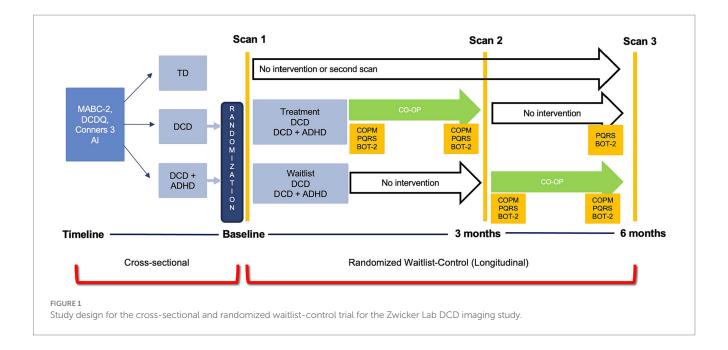
tracts, bilateral cingulum, and corpus callosum (Izadi-Najafabadi and Zwicker, 2021). These brain regions are associated with attention, self-regulation, motor planning, and inter-hemispheric communication (Izadi-Najafabadi and Zwicker, 2021).

Cortical brain structures undergo developmental changes during childhood (Giedd et al., 1999; Wierenga et al., 2014; Gilmore et al., 2018), and some therapies (e.g., behavioral, medications) have been associated with changes in cortical volume in children with neurodevelopmental disorders (Spencer et al., 2013; Sterling et al., 2013). To date, it is unknown whether CO-OP induces neuroplastic change in cortical brain structure. The aims of this study were to determine: (1) whether CO-OP intervention induces changes in cortical grey matter volume of children with DCD; and (2) if any grey matter volume changes are associated with improvements in motor performance and movement quality. We hypothesized that following CO-OP intervention, we would find: (1) increased cortical grey matter volume in regions of the brain associated in coordinating motor and executive functioning skills (i.e., parietal and frontal lobe); and (2) positive associations between changes in grey matter volume, motor performance, and movement quality improvements.

2 Materials and methods

2.1 Study design

This study was a part of a randomized waitlist-control trial using multiple neuroimaging modalities to assess brain changes with CO-OP intervention (ClinicalTrials.gov ID: NCT02597751). For the purpose of this study, structural MRI data were collected before and after CO-OP intervention to investigate changes in grey matter volume in children with DCD and in children with DCD and co-occurring ADHD (DCD+ADHD). Participants received treatment either after the first MRI (treatment group) or after a 3-month waiting period (waitlist group). A statistician randomized participants using computer-generated sequential blocks of 4 to 6. The randomization codes, either treatment or waitlist, were sealed in opaque envelopes until study enrollment. After screening and recruitment, all parents or legal guardians provided written consent and children assented to participate in the study. The study design (randomized waitlist-control trial) for the purpose of this paper is shown in Figure 1. All aspects of the study were approved by the Children's and Women's Health Centre/University of British Columbia Research Ethics Board (#H14-00397).



2.2 Participants

A convenience sampling strategy was used to recruit participants between September 2014 to July 2019. The following sources were used to recruit participants for the intervention: (1) Dr. Zwicker's researchintegrated DCD clinic at Sunny Hill Health Centre for Children; (2) BC Children's Hospital ADHD Clinic; (3) from caseloads of occupational and/or physical therapists from Sunny Hill and the Vancouver Regional Pediatric Team who service schools in the Vancouver and surrounding districts; and (4) the community. Community recruitment was done by using bulletin boards at BC Children's Hospital, UBC, and Vancouver schools. TD children were recruited through advertisements in Vancouver schools and community centres, and by word-of-mouth.

Inclusion criteria were based on the DSM-5 diagnostic criteria (American Psychiatric Association, 2013) and international clinical practice recommendations for DCD diagnosis (Blank et al., 2019) as follows: (1) scores ≤16th percentile on the MABC-2 (Henderson et al., 2007); (2) score in the suspected or indicative range on the Developmental Coordination Disorder Questionnaire (DCDQ) (Wilson and Crawford, 2007); (3) parent-reported motor difficulties from a young age; and (4) no other medical condition that could explain motor difficulties as per parent-reports, clinical reports and/or medical examination. Participants were excluded if they were born preterm (gestational age week <37 weeks) or diagnosed with other neurodevelopmental disorders, such as autism spectrum disorder.

The control group (TD children) included children 8–12-years old with no history of motor difficulties and a MABC-2 score ≥25th percentile. Exclusion criteria included being born preterm (gestation week <37 weeks) or diagnosed with any other neurodevelopmental disorder, such as autism spectrum disorder. Children assigned to the TD group were excluded if they were diagnosed with ADHD.

2.3 Procedure

Prior to enrollment, all participants were administered the Movement Assessment Battery for Children (MABC-2) to quantify the

level of motor impairment (Henderson et al., 2007) and whether the participants met the inclusion criteria for the study. In addition, the DCDQ parent-completed questionnaire was used to identify motor impairments of the participants in comparison to their peers (Wilson and Crawford, 2007). Lastly, the Conners 3 ADHD Index parent form was used to assess for attentional performance (Conners, 2009). Scores 70 and above are considered to be clinically significant (poorer attentional performance indicates greater attentional difficulties).

Both scanning and intervention took place at BC Children Hospital Research Institute. All children participated in a magnetic resonance imaging (MRI) safety screening and were informed about the MRI procedure. An MRI simulator session was done to familiarize the children with the scanning environment (noise, confined space, and head coil). They were also provided with strategies from the research team to help reduce potential for anxiety. The simulator session helped to answer the child's and/or parent's questions and inform the research team about the child's ability to remain still in the MRI scan, as the scans are sensitive to motion (Zaitsev et al., 2015).

After the first MRI session, children were randomly assigned to either the treatment or waitlist group, so that the research team was blinded to group allocation until after the first MRI. Children in both groups had three MRI sessions: (1) scan 1 occurred at enrollment; (2) scan 2 was conducted 3 months after the first scan (to measure treatment effect in the treatment group and maturation in the waitlist group); and (3) scan 3 occurred 6 months after the first scan (to assess follow-up 3 months after intervention in the treatment group and to measure the treatment effect of the waitlist group). Following the first MRI session, children in the treatment group received CO-OP intervention (led by an occupational therapist) once weekly for 10 weeks; they then had a post-intervention scan, and another follow-up scan 3 months later. Children in the waitlist group waited for 3 months for their second MRI and then began CO-OP intervention for 10 weeks; they had a third MRI after intervention (Figure 1). Caregivers were encouraged to attend treatment sessions so that therapists could instruct them how to facilitate strategy use between treatment sessions. Prior to intervention, children selected three functional motor goals (e.g., printing, tying shoes, performing

sport-related movements) that they wanted to achieve over the 10 weeks of therapy; each session lasted an hour. Outcomes measures included the Canadian Occupational Performance Measure (COPM) (Law et al., 2014), Performance Quality Rating Scale (PQRS) (Martini et al., 2014), and Bruininks-Oseretsky Test of Motor Proficiency-2 (BOT-2) (Bruininks and Bruininks, 2005). The COPM and BOT-2 were administered by an occupational therapist not involved in the intervention. The PQRS was video recorded by the treating therapist before and after intervention but was scored by the assessing therapist who was blinded to pre-test/post-test status.

2.4 Clinical outcome measures

2.4.1 Canadian Occupational Performance Measure

The COPM (Law et al., 2014) is a client-centered questionnaire that was administered by an occupational therapist before and after the completion of the 10-week CO-OP intervention. It allows the individual to rate their performance and satisfaction for each of their self-chosen goals on a scale of 1 to 10, where a higher score indicates increased levels of performance and satisfaction with their self-chosen goals (Law et al., 2014). A two-point change is considered clinically meaningful (Carswell et al., 2004; Law et al., 2014). The COPM is considered a valid, reliable, and responsive outcome measure (Carswell et al., 2004; Dedding et al., 2004; Eyssen et al., 2011).

2.4.2 The Performance Quality Rating Scale

The PQRS is a 10-point performance rating scale to evaluate changes in observed movement quality during task performance; higher scores indicate better movement quality (Miller et al., 2001; Polatajko and Mandich, 2004). The PQRS has moderate to substantial inter-rater reliability, excellent test-retest reliability, and good internal responsiveness (Miller et al., 2001; Martini et al., 2014). Before and after CO-OP intervention, children were video-recorded performing their chosen goals. An occupational therapist who was not engaged in the delivery of the intervention and was blinded to the pre/post assessment sessions scored the videos. The child's actual performance quality was rated on a scale of 1 to 10 (1 being "cannot do the skill at all" and 10 being "does the skill very well") (Martini et al., 2014). An increase of three points is considered clinically significant (Martini et al., 2014). The PQRS complements the COPM by measuring the actual, rather than perceived, performance of the child's selfchosen goals.

2.4.3 Bruininks-Oseretsky Test of Motor Proficiency-2

The short form of BOT-2 (Bruininks and Bruininks, 2005) was completed for this study. This short form consists of one or two items from each of the eight areas: bilateral coordination, balance, running speed and agility, strength, fine-motor precision, fine-motor integration, manual dexterity, and upper extremity coordination (Bruininks and Bruininks, 2005). The BOT-2 is a standardized, norm-referenced assessment that measures motor performance (Deitz et al., 2007), where a higher percentile scoring indicates better motor performance. This assessment is reported to have moderate to strong inter-rater/test-retest reliability (Deitz et al., 2007), excellent concurrent validity with other motor measures, and adequate

construct and content validity (Slater et al., 2010). The total percentile scores of the BOT-2 short-form were used for analysis.

2.5 Neuroimaging measures

2.5.1 MRI data acquisition

All brain images were acquired at the MRI Research Facility at BC Children's Hospital Research Institute in Vancouver, Canada. MRI scans were obtained on a 3-Tesla General-Electric Discovery MR 750 scanner. T1-weighted 3D structural scans were acquired with the following parameters: three-dimensional spoiled gradient recalled acquisition in steady state (3D SPGR), echo time = 30 ms, repetition time = 3,000 ms, FOV = 256, matrix size = 256 \times 256, flip angel = 12°, number of slices = 256, slice thickness = 1 mm, interleaved with no gaps (voxel size 0.9375 \times 0.9375 \times 1 mm). Using T1 weighted scans allows for reliable segmentation of tissues (grey matter, white matter, and cerebrospinal fluid) and permits reliable identification of underlying regions (Lerch et al., 2017).

2.5.2 Image quality control

All scans were visually inspected for truncation, motion, aliasingrelated and other artifacts (Krupa and Bekiesińska-Figatowska, 2015; Reuter et al., 2015). Specifically, image quality was assessed for head coverage, wrapping artifact, radiofrequency noise, signal inhomogeneity, susceptibility artifact, and ringing artifact (Reuter et al., 2015). An ordinal score was given to each image based on motion artifacts and image quality (pass, questionable, or fail) using standardized methodology (Harvard Center for Brain Science, 2014). Two trainees assessed the scans independently; the level of agreement for the categorization of each scan assessed by each trainee was 96%. Only scans that passed the final quality check from both trainees were included in the analysis. Additionally, quantitative measures of motion were calculated using the software package MRIQC (10.1371/journal. pone.0184661). In particular, we measured coefficient of joint variation (CJV), where higher values are related to the presence of heavy head motion and large intensity non-uniformity (10.3389/ fninf.2016.00010). Forty-five participants with DCD with significant motion artifact or poor grey to white matter differentiation were excluded from the larger sample to produce the final dataset of 20 participants with two good quality scans before and after intervention.

Twenty-two TD participants with similar data quality artifacts were excluded, resulting in nine participants with good quality scans acquired 3 months apart.

2.5.3 Voxel-based morphometry

2.5.3.1 Image pre-processing

Data were converted from DICOM (Digital Imaging and Communications in Medicine) to NIfTI (Neuroimaging Informatics Technology Initiative) format using the dcm2nii tool from MRIcron.¹ T1 images were processed using voxel-based morphometry (VBM), a computational technique that measures differences in grey matter volume through a voxel-wise comparison (Ashburner and Friston,

¹ https://www.nitrc.org/projects/mricron

2000; Whitwell, 2009). VBM uses T1-weighted MRI scans and performs a voxel-by-voxel statistical analysis across each image to identify volume differences between patients and controls (Ashburner and Friston, 2000). All pre-processing and longitudinal VBM data analysis were carried out using the Computational Anatomy Tool Box (CAT12, v1742, The Structural Brain Mapping Group, Jena, Germany, http://dbm.neuro.uni-jena.de/cat12/), through Statistical Parametric Mapping 12 software (SPM12, v7771, The Wellcome Centre for Human Neuroimaging, London, United Kingdom, https://www.fil.ion.ucl.ac.uk/spm/) in MATLAB R2020b (Mathworks, Natick, Massachusetts, United States).

For image preprocessing, all T1 images were manually registered to the anterior commissure at the origin of the Montreal Neurological Institute (MNI) coordinate system (Jahn, 2019). Initially in longitudinal VBM analysis, intra-participant co-registration was performed on the pre- and post-intervention images. The co-registered images were then realigned across participants and bias-corrected with reference to the mean images computed from each participant's pre- and post-intervention images. The images were then segmented into grey matter, white matter, and cerebrospinal fluid (CSF) with the customized pediatric tissue probability maps from Template-O-Matic Toolbox (TOM8 https://neuro-jena.github.io/software.html#tom) as an initial estimate. All images were included if their weight average Image Quality Rating (IQR) was greater than 80%, corresponding to a "good" image quality. Mean correlations between all volumes were visualized through CAT12. Volumes with a correlation below two standard deviations from the sample mean were again visually inspected for artifacts. Next, good quality pre- and post-average affine-registered white and grey matter tissue segments were extracted to construct a customized Diffeomorphic Anatomical Registration Through Exponentiated Lie Algebra (DARTEL) study-specific template registered to the MNI-International Consortium for brain Mapping (ICBM) space. This alternative to the adult-based template provided by CAT12 was used to achieve a more accurate interparticipant registration to improve the realignment of small inner structures for an overall better segmentation (Good et al., 2001; Yassa and Stark, 2009). Likewise, this additional step is similar to VBM studies done in other pediatric neurodevelopmental disorder studies that created a study-specific average template for their sample (Reynolds et al., 2017; Wang et al., 2017; Sáenz et al., 2020). The images were then normalized using an affine spatial normalization and a further modulation was applied to convert the voxel values of tissue concentration to measures of volume. Finally, the normalized grey matter maps were smoothed with an isotropic Gaussian kernel (full width at half maximum = 6 mm). Total intracranial volume (TIV) was calculated for the pre and post grey matter, white matter, and CSF images for each participant using CAT12 module intracranial volume."

2.5.3.2 Computational Anatomy Toolbox

The Structural Brain Mapping Group at the University of Jena (Jena, Germany) designed the automatic and easy-to-use toolbox CAT12 as an extension to the SPM software. CAT12 follows a standard VBM analysis pipeline similar to VBM8. We used segmentation through SPM's extension CAT12 rather than FreeSurfer or FSL as SPM produces a more robust segmentation for those with limited image quality (Fellhauer et al., 2015). This decision was further supported through a comparison to previous toolboxes, with CAT12

providing a more accurate and robust volumetric analysis (Farokhian et al., 2017) and advanced segmentation tool (Tavares et al., 2020). It has also been used in neurodevelopmental disorders that commonly co-occur with DCD (Wang et al., 2017; Mei et al., 2020; Sáenz et al., 2020) where the workflow was adapted to accommodate a pediatric population as recommended for VBM analysis.

2.6 Statistical analysis

2.6.1 Participant characteristics

Jeffreys's Amazing Statistics Program (JASP https://jasp-stats.org/) was used to summarize participant characteristics [age, sex, TIV, MABC-2 (motor ability), DCDQ (motor function), and Conners 3 ADHD Index (ADHD symptomatology)] and pre-post intervention outcome measures. The behavioral data for the entire cohort have been reported by Izadi-Najafabadi et al. (2022a). Here, we report motor outcome data for the sub-sample in this paper. The Wilcoxon Signed-Rank Test (non-parametric) was used to compare the before and after effect of CO-OP intervention on average COPM performance and satisfaction scores, average PQRS total actual performance scores, and BOT-2 motor percentile ranks. The alpha level was set to 0.05 with Bonferroni correction used to correct for multiple comparisons when comparing pre- post values to avoid type 1 errors.

2.6.2 Longitudinal VBM statistical analysis

All statistical models were to be set up using general linear modeling through SPM. Initially, our goal for the analysis was to conduct a treatment vs. waitlist comparison; however due to the smaller than anticipated sample size for each group (n=7 treatment, n = 13 waitlist), we combined scans 1 and 2 of the treatment group and scans 2 and 3 of the waitlist group to examine grey matter volume differences before and after intervention. Paired participant smoothed grey matter volumes were entered into a second level analysis using the "Flexible factorial" module in CAT12. To estimate differences in pre-post grey matter in children with DCD, a repeated measure analysis of variance (ANOVA) design with 5,000 permutations with an alpha level of 0.05 was used, with whole and within exchangeability blocks. Threshold-Free Cluster Enhancement (TFCE) thresholding was conducted using the TFCE Toolbox Version r2142 with 5,000 permutations (Draper-Stoneman method) with equal variance (patients) with an E=0.5 and H=0.2. TIV was mean-centered and used as a covariate/nuisance variable as recommended in VBM analysis to account for intra-individual differences. In order to conserve degrees of freedom, age, attentional performance, and sex were not included as covariates in this analysis. Initially, a regression analysis was proposed to examine the relationship between grey matter volume and COPM, PQRS, and BOT-2. However, PQRS and COPM were significantly positively correlated (r = 0.39, p = 0.046) for this sample (Hinkle et al., 2003). Instead, we used a regression analysis with only grey matter volume, BOT-2 (motor performance), and PQRS (actual performance quality), controlling for the effect of intracranial volume. PQRS was used instead of COPM as it is a more objective measure, despite COPM being our primary outcome. Structural

² http://dbm.neuro.uni-jena.de/tfce/

images were analyzed using TFCE due to its increased sensitivity compared to voxel- or cluster-based statistics (Smith and Nichols, 2009; Salimi-Khorshidi et al., 2011; Radua et al., 2014). We assessed statistical significance with the permutation test included in SPM.

All results are reported with TFCE thresholding uncorrected for multiple comparisons (no $p_{FDR-corrected}$ or $p_{FWE-corrected}$) but corrected for the number of planned comparisons (pre > post, pre < post). Results are presented at p < 0.001 with cluster size threshold at 50 voxels. Cluster size threshold was based on current literature regarding cluster thresholding. Given our N < 50, we opted for a more stringent cluster threshold of 50 compared to lower thresholds of 10 (Lieberman and Cunningham, 2009; Woo et al., 2014). This is also comparable to previous publications of cerebellar VBM with samples of children with neurodevelopmental disabilities (D'Mello et al., 2015). Uncorrected results (p < 0.001) minimize false negative results but do increase the false-positive rate (Durnez et al., 2014).

3 Results

3.1 Final sample

This study recruited 80 children with DCD+/-ADHD from which 60 were excluded because they either had co-occurring ASD or preterm birth (n=9), declined to participate in the intervention (n=2), discontinued intervention (n=4), or had insufficient data quality for both scans (before and after intervention) to conduct VBM analysis (n=45) (Figure 2). In order to preserve power, the DCD and DCD+ADHD group were combined. Our final sample for voxel-based morphometry analysis after quality checks comprised of 20 children with DCD [mean (SD) age=9.9 (1.6) years], of which 70% were males.

Similarly, 35 TD children were initially recruited, from which 26 were excluded because they either declined to participate (n=1), had ADHD (n=1), were born preterm (n=1), had a MABC-2 \leq 16th percentile (n=1), or had insufficient data quality for both scans (3 months separation) to conduct VBM analysis (n=22).

No difference was found between children with DCD with good or bad quality T1 images based on BOT2 or PQRS scores. Difference in Connors T-score was close to significant [p=0.086, CI95%=(-8.67, 0.57)]. The loss of children due to poor image quality was similar between the TD and DCD cohorts—71 and 70%, respectively—suggesting that getting quality T1 images from children this age is difficult in general.

Children with DCD whose data was excluded due to motion had an average CJV of 0.68 (± 0.13 SD), while those that were kept had an average CJV of 0.61 (± 0.09 SD). These values were significantly different [p = 0.002; CI95%=(0.03, 0.11)]. Of the participants that were included for analysis, there was no difference in CJV between the TD and DCD cohorts. Furthermore, for the children with DCD included for analysis, no correlation was found between CJV and MACB-2 scores [CI 95%=(-0.36, 0.06)].

3.2 Participant characteristics

Table 1 presents demographic and behavioral characteristics of the sample. Our final sample included 20 participants with DCD

(Figure 2), of which 19 (95%) had attentional difficulties as indicated by a score of 70 or greater on the Conners ADHD Index. This is further supported by current literature which suggests a greater than 50% overlap between the DCD and ADHD (Kadesjö and Gillberg, 1998; Goulardins et al., 2015; Lange, 2017). Lastly, our sample of DCD included 14 males (70%), which aligns with previous literature indicating high prevalence of DCD in males (American Psychiatric Association, 2013). We also included 9 TD participants to investigate if any grey matter changes were due to maturation over a 3 months period.

3.3 Motor outcomes

The Wilcoxon Signed-Rank Test (non-parametric) was used to compare the before and after effect of CO-OP intervention. Participants showed statistically significant improvements (p<0.001) in their motor goals (COPM), movement quality (PQRS), and motor skills (BOT-2) after CO-OP intervention (Table 2).

3.4 Grey matter volume changes following intervention

When comparing pre-post scans, children with DCD had significantly decreased grey matter [cluster size (k) > 50, $p_{\text{uncorrected}} < 0.001$] in the right hemisphere in the following regions: middle cingulate gyrus, posterior cingulate gyrus, and the superior frontal gyrus following CO-OP intervention (Table 3 and Figure 3). There were no regions where there was increased grey matter when comparing pre- intervention to post-intervention—the inverse contrast [cluster size (k) < 50]. A follow-up analysis with age as a covariate can be found in Supplementary Figure S1.

When looking at TD children over the same period of time, no changes were found in the grey matter [cluster size (k) >50, $p_{\text{uncorrected}}$ <0.001].

3.5 Relationship of motor performance and performance quality to changes in grey matter volume

There were no significant positive or negative (inverse contrast) association between overall motor performance on the BOT-2 percentile scores and grey matter volume changes [cluster size (k) <50]. In addition, there was no significant positive or negative (inverse contrast) association between actual performance quality on motor goals after 10 weeks of intervention (as measured by PQRS scores) and grey matter volume changes [cluster size (k) <50].

4 Discussion

In this randomized waitlist-control study, we found that, following CO-OP intervention, children with DCD had decreased grey matter volume in the right superior frontal gyrus, right middle cingulate gyrus, and right posterior cingulate gyrus. We reiterate that due to sample size, we did not correct for multiple comparisons, and further

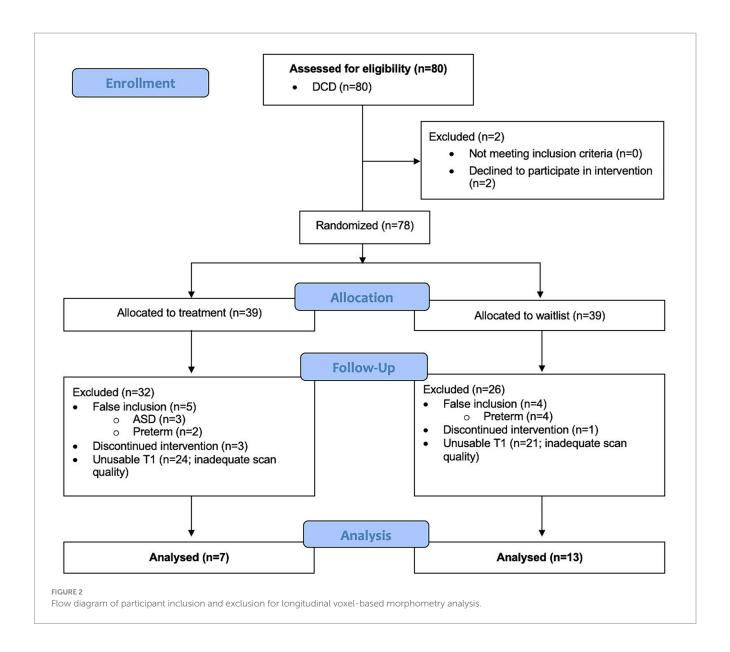


TABLE 1 Description of cohort (N = 20).

Participant characteristics	DCD <i>N</i> (%) or mean (SD)	TD <i>N</i> (%) or mean (SD)
Male	14 (70)	4 (44)
Age at MRI (years)	9.9 (1.6)	10.4 (1.5)
MABC-2 (percentile)	5.5 (8.5)	61.7 (27.5)
Conners ADHD Index (T-scores)	87.0 (5.8)	57.1 (12.5)
DCDQ in suspected or indicative range	20 (100)	1 (11)
Total intracranial volume (L)	1.50 (0.18)	1.51 (0.09)

ADHD, attention deficit hyperactivity disorder; DCD, developmental coordination disorder; DCDQ, Developmental Coordination Disorder Questionnaire; L, litres; MABC-2, Movement Assessment Battery for Children-2nd ed.; SD, standard deviation.

work and replication of these findings is needed. The regions that showed volumetric changes are different from those identified in our previous work that investigated brain volume differences in DCD

TABLE 2 Clinical outcomes before and after CO-OP intervention.

Clinical outcome measure	Pre-test mean (SD)	Post-test mean (SD)	р
COPM performance	3.1 (1.1)	6.3 (1.3)	<0.001
COPM satisfaction	2.5 (1.5)	7.2 (1.4)	<0.001
PQRS	3.1 (1.3)	6.2 (1.4)	<0.001
BOT-2 percentile	13.4 (11.7)	22.0 (18.4)	<0.001

BOT-2, Bruininks-Oseretsky Test of Motor Proficiency-2nd edition; CO-OP, Cognitive Orientation to daily Occupational Performance; COPM, Canadian Occupational Performance Measure; PQRS, Performance Quality Rating Scale; SD, standard deviation.

versus typically developing children using the pre-treatment scans (Malik et al., 2023). There, we showed that children with DCD had greater grey matter volume in left STG at baseline. The post-treatment findings in the right hemisphere reported here may reflect the lateralization of the brain in the early stages of learning to problem solve, or it might relate to emotional regulation (Shobe, 2014; Blais et al., 2017), both of which could be modulated via the CO-OP

TABLE 3 MNI coordinates for pre-post intervention differences in grey matter volume.

Location	Χ	Υ	Z	TFCE	$oldsymbol{ ho}_{uncorrected}$	Cluster size
Right middle cingulate gyrus	8	-27	31	495.2	0.001	
Right posterior cingulate gyrus	10	-47	27	466.2	<0.001	114
Right posterior cingulate gyrus	10	-38	33	463.4	<0.001	
Right superior frontal gyrus	11	24	54	494.7	< 0.001	
Right superior frontal gyrus	8	45	45	449.9	0.001	102
Right superior frontal gyrus	10	37	52	445.8	<0.001	

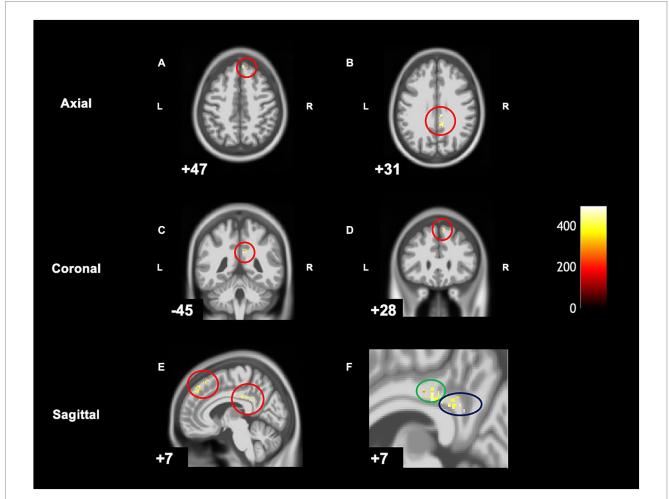


FIGURE 3
Within-group differences show significantly decreased grey matter in children with DCD after intervention on CAT T1 IXI555 GS. (A) Right superior frontal gyrus. (B) Middle and posterior cingulate gyrus. (C) Right middle and posterior cingulate gyrus. (D) Right superior frontal gyrus. (E) Right superior frontal gyrus, middle and posterior cingulate gyrus. (F) Zoomed image of E: middle (green) and posterior cingulate gyrus (blue).

intervention. In other words, the intervention changes did not "normalize" the previously observed group-based or diagnostic differences in volume, though they may relate to important changes and learning functions implicated in the therapy. To contextualize and interpret the results reported in this study, we will discuss motor learning theories and background literature on the frontal and parietal regions that were found to have decreased grey matter volume.

According to general principles of motor learning and neuroplasticity, interventions that involve people in active, repetitive training can not only increase motor function (Salbach et al., 2004; Michaelsen et al., 2006; Arya et al., 2011) but can also lead to

neuroplastic changes (Takeuchi and Izumi, 2013; Maier et al., 2019). Compared to TD children, children with DCD use different strategies and brain regions to improve motor performance and motor learning (Zwicker et al., 2010, 2011; Biotteau et al., 2016). Children with DCD also tend not to improve their motor skills with practice alone (Schoemaker and Smits-Engelsman, 2015), but rather benefit from using cognitive strategies and problem-solving skills to facilitate motor skill acquisition (Mandich et al., 2003; Jokić et al., 2013). CO-OP is a task-oriented intervention that combines both motor learning theories with cognitive approaches (i.e., problem-solving and self-evaluation) (Polatajko et al., 2001; Jackman et al., 2018).

Additional studies underscore the self-regulation processes. For example, self-regulation is thought to be a mediator to improve motor skills (Jokić et al., 2013; Green and Payne, 2018). Brain imaging data to date seem to support this hypothesis, as brain regions associated with self-regulation showed greater structural (most findings were in default mode network) (Izadi-Najafabadi and Zwicker, 2021) and functional connectivity (default mode network and the right pregenual anterior cingulate cortex) (Izadi-Najafabadi et al., 2022b) after CO-OP intervention. Two of the brain regions that showed decreased grey matter volume after intervention, the right posterior cingulate gyrus and the right superior frontal gyrus, further support the hypothesis that changes in self-regulation may mediate improved motor function.

The posterior cingulate gyrus is one of the most densely connected regions in the brain, and has been discussed as one of the "least wellunderstood regions of the cortex" (Leech and Smallwood, 2019, p. 73). It functions as a hub for the default mode network (DMN), arguably one of the most complex networks in the brain (Alves et al., 2019; Buckner and DiNicola, 2019). The DMN has been implicated in a wide array of higher-order functions, most of which rely on internally constructed information (Buckner et al., 2008; Leech and Sharp, 2014; Pan et al., 2018). Perhaps the most relevant DMN functions in the current context would be network suppression during active tasks and learning combined with self-referential processing and self-regulation (Brewer et al., 2013). During CO-OP intervention, children with DCD are guided to use self-regulatory skills such as goal setting, planning, and self-monitoring to address their motor performance difficulties (Hyland and Polatajko, 2012; Jokić et al., 2013). As self-regulation is thought to mediate motor skill improvements observed with CO-OP intervention (Jokić et al., 2013; Green and Payne, 2018), it makes clinical sense that neuroplastic changes would be observed in brain regions associated with self-regulation. We propose that CO-OP intervention may reinforce synaptic connections while pruning less efficient pathways, resulting in a decrease in grey matter volume. While speculative, our interpretation is consistent with theories and findings relating to experience-dependent neuroplasticity (Giedd et al., 1999; Lenroot et al., 2007; Kleim and Jones, 2008; Wierenga et al., 2014).

The right superior frontal gyrus also showed decreased grey matter volume after CO-OP intervention. Activation of the right superior frontal gyrus is thought to modulate inhibitory control (Hu et al., 2016). Inhibitory control is defined as the suppression of behavior in response to internal or external influences (Morasch and Bell, 2011). It is a cognitive function that plays an important role in tasks such as riding a bike, where it is often necessary to prevent an action from being performed inappropriately (Coxon et al., 2007). As children with DCD have difficulty with inhibitory control of attention and executive function (Wilson et al., 1997; Wilson and Maruff, 1999; Mandich et al., 2003; Tsai, 2009; Wilson et al., 2020), impairment in inhibitory control might play a role in underlying motor coordination problems (Tsai, 2009). Our finding of decreased grey matter in the frontal region is consistent with neurodevelopmental and lesion neuroimaging studies that have identified dorsolateral and medial prefrontal cortices (responsible for unwanted response) (Rubia et al., 2005) and the right inferior frontal gyrus and basal ganglia (for cancellation of prepared or ongoing movements) (Aron et al., 2003; Chambers et al., 2006; Chikazoe et al., 2008) are involved in inhibitory control. Inhibitory control tends to improve with active intervention (Tsai, 2009), which may be associated with decreased grey matter

volume in the superior frontal gyrus. As above, we speculate that this may be due to synaptic pruning of pathways that were reinforced during intervention.

The middle cingulate gyrus is known by two names: (1) dorsal anterior cingulate cortex (Palomero-Gallagher et al., 2008, 2009; Apps et al., 2013); or (2) middle cingulate cortex, which is further split into an anterior and posterior middle cingulate cortex (Apps et al., 2013; Brewer et al., 2013; Vogt, 2016). The different terminology may lead many studies to inaccurately discuss the functional properties of the middle cingulate cortex (Apps et al., 2013). Here, we adopt the more recent nomenclature of the middle cingulate gyrus, which itself is divided into dorsal, middle and posterior subsections. It has extensive connections with cognitive (e.g., lateral prefrontal) and motor-related (e.g., premotor and primary motor) areas of the cortex (Vogt and Morecraft, 2009; Stevens et al., 2011). Based on the seminal study conducted in nonhuman primates by Paus (2001), the most dorsal portion of the middle cingulate gyrus is important for the execution of voluntary motor control through its cognitive and motor connections and processing of abstract thinking and intention in motor execution. In humans, the dorsal middle cingulate gyrus is activated with motorrelated tasks (Beckmann et al., 2009) and during the internal generation of movements (pre-frontal, pre-motor, parietal, basal ganglia) (Deiber et al., 1999; Cunnington et al., 2002; Debaere et al., 2003; Lau et al., 2004; van Eimeren et al., 2006; Jankowski et al., 2009). The posterior middle cingulate cortex is part of the caudal cingulate premotor area which is involved in multisensory orientation of the individual in space and in sensing the force and direction of movements in space (Vogt, 2016). Reflecting on the methodology of the intervention, to achieve the self-chosen motor goals, CO-OP uses cognitive-based strategies during task-specific intervention to facilitate motor skill acquisition. The problem-solving aspect of the intervention promotes the thought process of "what" and "how" to do a particular action. This thinking, in addition to the cognitive connections of middle cingulate gyrus may have promoted a decrease in grey matter (through increased synaptic pruning) in the middle cingulate gyrus and cognitive (posterior cingulate gyrus) and motor regions (superior frontal gyrus) found in this study.

Previously, we reported that children with DCD had greater grey matter volume in the left superior frontal gyrus compared to typically developing children (Malik et al., 2023). This is posited to be a result of aberrant brain maturation, specifically a delay or dysfunction in cortical thinning through the mechanisms (synaptic pruning) described earlier. It is thought that similar to other neurodevelopmental disorders, dysfunction of synaptic pruning may result in increased grey matter as the neural connections are not maturing as expected. This leads to difficulties in working memory and higher order cognitive functions. After having undergone 10 weeks of CO-OP intervention, the decrease in grey matter volume in the right middle and posterior cingulate gyrus and right superior frontal gyrus may be a result of increased synaptic pruning.

The short form of BOT-2 percentile (measure of motor performance) significantly increased after intervention with this sub-set of participants; however, this was not the case in the larger sample (Izadi-Najafabadi et al., 2022a). We did not see any association between grey matter changes and this measure of overall motor performance in this small sample. One possible explanation for this has to do with the nature of the intervention and neuroplasticity. CO-OP is a task-oriented intervention that focuses on using a problem-solving based framework to acquire specific motor skills; it does not address the underlying motor

impairment (Polatajko et al., 2001; Blank et al., 2012; Smits-Engelsman et al., 2012). Given the principles of neuroplasticity (Kleim and Jones, 2008), we would only expect to see brain changes associated with the specific target actions, not overall motor performance; thus, it is not surprising that we did not observe a relationship between grey matter volume changes and BOT-2 scores. However, we also investigated the association between PQRS and grey matter volume changes in the brain. The PQRS is a measure of actual performance quality for the child-chosen goals that were addressed in therapy over 10 weeks. We also did not find a relationship between movement quality and grey matter volume. Given our small sample size, we are likely under-powered to detect this expected relationship.

Several limitations are present in this study. First, our sample size was limited. Of the 80 participants scanned, several scans were lost due to insufficient quality for VBM analysis at either or both time points. After stringent quality checks and exclusion criteria, the final sample was small (N=20). As mentioned by Sáenz et al. (2020), this may lead to a biased sample as the significant head motion can be associated with clinical traits and the scans of the most severely impaired participants therefore might have been excluded; the exclusion of poor-quality scans also limits the generalizability of results (Sáenz et al., 2020). Relatedly, we did not correct for multiple comparisons in the analyses, and thus the results are potentially more vulnerable to false positives. The fact that the results are sensible (i.e., the identified brain regions relate to key aspects of the intervention) suggests the results may be valid, but replication is needed. Second, there are no standardized quantification guides to measure degree of motion artifact in T1 scans. We used visual inspection by two trained, independent raters based on established guidelines (Harvard Center for Brain Science, 2014), ensuring that only highquality scans that were deemed acceptable by both raters were included. Third, while we intended to analyze children with DCD and DCD+ADHD separately, our sample for VBM analysis was smaller than expected and included children with DCD and co-occurring ADHD (n=6) could have confounded our findings. However, the sample had similar Connors ADHD Index scores, suggesting the children with diagnosed ADHD were more similar than different to the DCD group. We were also unable to compare the treatment group with the waitlist group, which would have controlled for maturation, nor were we able to examine follow-up effects in the treatment group. Fourth, we did not control for age, sex, or medications, in order to conserve power. Finally, there are some limitations regarding volume-based measures. Since grey matter includes surface area and thickness, each of which have their own developmental trajectories, the interpretation of grey matter volume becomes difficult without examining surface area or thickness individually (Frye et al., 2010).

Future studies should continue to explore intervention-induced changes in grey matter volume in children with DCD but in a larger sample of children and measured overtime to see if these changes are maintained. Exploring cortical thickness and volume in one study longitudinally would provide more insight into the brain's structural morphology in this disorder. Likewise, results could be stratified by age, sex and/or medication use to provide further insights (Caviness et al., 1996; De Bellis et al., 2001; Spencer et al., 2013). Lastly, to further explore synaptic pruning, molecular, and animal model studies should be conducted to examine the long-term intervention-induced changes in synaptic pruning in this population.

Overall, CO-OP is one of the recommended task-oriented rehabilitation interventions in the international clinical practice guidelines for DCD (Blank et al., 2019). This intervention is effective in improving children's perceptions of their motor performance of the specific skills they wanted to learn, as well as improved motor quality while performing these skills as rated by a therapist. In conclusion, our data indicate that CO-OP induces neuroplasticity of the grey matter in the right superior frontal gyrus (inhibitory control), right posterior cingulate gyrus (self-regulation), and right middle cingulate gyrus (voluntary thinking and cognitive and motor connections) in children with DCD. We speculate that these neuroplastic changes result from upregulated synaptic pruning that occurs within the repeated actions and learning of the intervention, leading to the maturation of synapses in DCD-related circuits. This study further supports our team's findings on how CO-OP induces changes in structure and function of brain regions associated with self-regulation, providing initial evidence for the brain-based impact of this intervention.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by Children's and Women's Health Centre/University of British Columbia. The studies were conducted in accordance with the local legislation and institutional requirements. Parents/legal guardians provided informed written consent and children assented to participate in the study.

Author contributions

MM: Data curation, Formal analysis, Investigation, Visualization, Writing – original draft. DL: Formal analysis, Writing – review & editing. TV: Formal analysis, Writing – review & editing. JZ: Conceptualization, Formal analysis, Funding acquisition, Methodology, Project administration, Resources, Supervision, Visualization, Writing – review & editing. AW: Formal analysis, Visualization, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fnhum.2024.1316117/full#supplementary-material

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EDITED BY
Catherine Purcell,
Cardiff University, United Kingdom

REVIEWED BY
Reint Geuze,
University of Groningen, Netherlands
Jessica Tallet,
Université Toulouse III Paul Sabatier, France

*CORRESPONDENCE Emma Sumner ☑ e.sumner@ucl.ac.uk

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Oculomotor differences in adults with and without probable developmental coordination disorder

Emma Sumner^{1*} and Elisabeth L. Hill²

¹Department of Psychology and Human Development, Institute of Education (IOE), UCL's Faculty of Education and Society, London, United Kingdom, ²Department of Psychology, City University of London, London, United Kingdom

Adults with Developmental Coordination Disorder (DCD), sometimes referred to as dyspraxia, experience difficulties in motor development and coordination, which impacts on all aspects of their daily lives. Surprisingly little is known about the mechanisms underlying the difficulties they experience in the motor domain. In childhood DCD, aspects of oculomotor control have been shown to be altered. The purpose of this study was to determine whether oculomotor differences are present in adults with and without probable DCD. Visual fixation stability, smooth pursuit, and pro-and anti-saccade performance were assessed in 21 adults (mean age 29 years) with probable DCD/dyspraxia (pDCD) and 21 typically-developing (TD) adults (mean age 21 years). Eye tracking technology revealed that oculomotor response preparation in the proand anti-saccade tasks was comparable across groups, as was pursuit gain in the slower of the two smooth pursuit tasks. However, adults with pDCD made significantly more saccades away from the fixation target than those without DCD and significantly more anti-saccade errors. Further, compared to TD adults, adults with pDCD demonstrated difficulties in maintaining engagement and had lower pursuit gain in the faster pursuit task. This suggests that adults with pDCD have problems with saccadic inhibition and maintaining attention on a visual target. Since this pattern of results has also been reported in children with DCD, oculomotor difficulties may be persistent for those with DCD across the lifespan. An awareness of the impact of atypical oculomotor control in activities of daily living across the lifespan would support clearer understanding of the causes and impacts of these difficulties for those with DCD.

KEYWORDS

developmental coordination disorder, oculomotor, saccade, inhibition, eye tracking

Introduction

Developmental coordination disorder (DCD), sometimes referred to as dyspraxia, has been estimated to affect between 5 and 6% of individuals (Blank et al., 2012). DCD is currently diagnosed using the Diagnostic and Statistical Manual (DSM-5) categorical framework, which identifies a significant delay in acquiring gross and/or fine motor skill as the primary characteristic (APA, 2013). Difficulties with motor coordination must also be seen to interfere with academic achievement and/or activities of daily living, and cannot be better explained by intellectual disability, visual impairment or a neurological condition affecting movement.

Theories about the underlying mechanisms of DCD are still to offer a definitive explanation about causality (Blank et al., 2019). A recent systematic review and meta-analysis of the literature highlighted consistent reporting of difficulties with executive functions (e.g., inhibition, working memory and attention which are supportive of movement control) and cognitive-motor integration [e.g., reduced patterns of activation when considering functional Magnetic Resonance Imaging (fMRI) data during tasks that required different aspects of action preparation and attention during motor performance] in children with DCD (Subara-Zukic et al., 2022). Taken together, the authors suggested that DCD may be a disorder of motor-cognitive function. Studying oculomotor (eye movement) function using tasks that tap into executive function (e.g., planning, inhibition or shifting of eye movements) and tracking targets (visualmotor integration) is one way to further explore aspects of both motor and cognitive control (Maron et al., 2021). Notably little data is available to indicate whether these difficulties persist into adulthood in those with DCD. However, such a finding has important research, clinical and educational implications, in terms of developing our theoretical understanding of DCD and targeting support.

The visual system enables us to process information in the world that we can then act upon. By focusing the eyes, fixation enables an individual to determine what and where an object is (Krauzlis et al., 2017). Eye movements (saccades) are made to redirect the fovea and centre an object on this region of the retina, thus making the object clearer (Karatekin, 2007). These saccades may be reflexive (i.e., moving the eyes to a new object that has appeared in the visual system) or volitional (i.e., involving high-level control, such as using spatial memory to search for an object, or inhibition to avoid distractions). Of note, there is considerable overlap of the networks involved in oculomotor control, cognitive control (e.g., attention, planning, inhibition) and motor coordination (e.g., fronto-cerebellar and frontoparietal regions; Doron et al., 2010; Kheradmand and Zee, 2011). Oculomotor paradigms that explore inhibitory control include asking individuals to sustain fixation, to make a saccade to an object (pro-saccade), or to inhibit a reflexive eye movement to an object appearing in the periphery (e.g., using an anti-saccade task, explained later). Another assessment of oculomotor control is smooth pursuit, which involves maintaining an object on the fovea, thus the eye moves with the object, such as when catching a ball or tracking a moving object (Karatekin, 2007). Precise coupling between oculomotor and limb kinematics is important for motor planning and coordination. Poor pursuit has been associated with eye-hand coordination difficulties, such as catching a ball (Glazebrook et al., 2009), and motor planning (e.g., when reaching for an object) is supported by attention and visual-motor integration (Gilbert and Li, 2013). Thus, mastery of accurate oculomotor control is one important aspect for the acquisition and execution of skilled behaviors (e.g., completing fine motor tasks, locomotor control, navigating the environment).

Atypical visuomotor function has been widely documented in children with DCD, with studies revealing difficulties in orienting attention (using COVAT (Covert Orienting of Visuospatial Attention Tasks) e.g., Wilson et al., 1997; Chen et al., 2012), and delays in attentional disengagement and motor initiation in hand-eye coordination tasks (e.g., Wilmut et al., 2007; Wilson et al., 2017). Given the relationship between the visual and motor systems, it has been argued that it is possible that atypicalities in oculomotor function will have consequences for broader fine and gross motor skill

difficulties (Maron et al., 2021). Yet, research utilising oculomotor paradigms in individuals with DCD remains scarce.

One study to explore saccade performance in a small sample of adolescents with dyspraxia (reported as being diagnosed with DCD as per the DSM-5 criteria; n = 7) revealed a mixed pattern across different saccade tasks with varying demands (e.g., making visuallyguided saccades, memory-guided saccades, delayed saccades, and anti-saccades; Gaymard et al., 2017). Characteristics of those with dyspraxia compared to age-matched controls revealed increased variability of saccade amplitudes in the visually guided task, decreased velocities of non-visually guided saccades, in addition to higher error rates on an anti-saccade task. The authors concluded that the findings may reflect impaired connectivity between frontal and cerebellar oculomotor structures in a dyspraxic sample. However, since their small sample comprised adolescents with dyspraxia all co-occurring with other conditions (including autism, schizophrenia and reading difficulties), further work is needed to understand the specificity of this conclusion.

Another study to examine oculomotor function focused on primary schoolaged children with a diagnosis of DCD (n = 23). Sumner et al. (2018) found that children with DCD were comparable to typically-developing peers in their ability to prepare and direct an eye movement (i.e., saccade latency and accuracy was comparable in a visually-guided saccade task, often referred as a 'prosaccade' task). However, children with DCD presented with difficulties maintaining engagement in fixation and smooth pursuit tasks when compared to a control group and, echoing the findings of Gaymard et al. (2017) made more errors on an anti-saccade. The finding of difficulties with pursuing a moving target supports earlier findings showing that children with DCD had lower pursuit gain than their peers and made more saccades away from the target (Langaas et al., 1997). Sumner et al. (2018) argued that children with DCD have pronounced difficulty exercising inhibitory control (i.e., suppressing intrusive saccades); a finding that implicates the fronto-parietal circuit (Miller et al., 2005). These findings are also supported by Gonzalez et al. (2016) who found that children with DCD made more saccades away from a target and had difficulty inhibiting saccades.

To the best of our knowledge, research on oculomotor function in DCD has not been extended to adult populations. This is despite the increasing recognition that motor difficulties persist into and throughout adulthood for some individuals with DCD (e.g., Tal-Saban and Kirby, 2018). Being able to further characterize oculomotor function in DCD by extending this to adults, is an important step in understanding whether a unique oculomotor profile may present (and be persistent) in those experiencing motor difficulties. Thus, the aim of the current study was to extend the use of the oculomotor paradigm cited in Sumner et al. (2018) to an adult population, asking: do adults with DCD present with oculomotor challenges in comparison to adults that do not have a diagnosis of DCD? Given the existing findings that highlight executive functioning difficulties as a prominent feature of DCD at all ages (Meachon et al., 2022), a brief measure of self-reported attention difficulties was taken to characterize the study sample in this respect, along with selfreported diagnosis of any conditions other than DCD. More detailed consideration of attention deficits and executive functioning of various forms was not possible within the scope of the current study. A body of research has shown difficulties

with inhibition in children and adults with DCD using experimental tasks (e.g., Bernardi et al., 2016; He et al., 2018). Thus, based on existing research from both eye tracking and experimental studies, we predicted difficulties in saccadic inhibition but not saccade preparation in adults with DCD compared to typically-developing adults. Such difficulties were expected to be observed in a fixation and smooth pursuit task, which require inhibiting saccades and maintaining fixation on the target (more saccades were predicted for the fixation task, and less time in pursuit); as well as during an anti-saccade task that requires facilitation of saccades away from a target (more errors were predicted in this task).

Materials and methods

Participants

Two groups of participants were recruited: 21 adults who reported a diagnosis of DCD/dyspraxia (aged 21–46, 10 male) and 21 typically-developing adults that did not report a motor difficulty (aged 18–32, 5 male). Participants were recruited via a research participant scheme in a university, by contacting university disability services in England and through a targeted approach on social media.

Adults with DCD/dyspraxia completed a screening questionnaire designed by the research team in which they confirmed their date of birth, ethnicity and any formal developmental, physical or medical diagnoses that they had received from birth to the present day. Five participants reported having a co-occurring diagnosis of dyslexia. No other developmental or medical diagnoses were reported. Since a comprehensive diagnostic assessment could not be undertaken, henceforth this group is described as 'probable DCD' (pDCD). Fifteen participants (71.4%) self-identified as White British, two as White Other (9.6%), two as Indian Asian (9.6%), one as Black (4.7%) and one as Mixed Heritage (4.7%). Two participants wore contact lenses during testing.

Typically developing adults in the comparison group also completed the screening questionnaire. No adult in this group reported any diagnosis of any kind (development, physical or medical). Sixteen participants self-identified as White British (76.1% of the sample), two as Black (9.6%), two as Mixed Heritage (9.6%) and one as Indian Asian (4.7%). One participant wore contact lenses during testing.

Measures

Adult ADHD self-report scale

The Adult ADHD Self Report Scale v1.1 (ASRS) is a checklist that can be used as a screening tool for ADHD in the general population (Kessler et al., 2005). The checklist consists of 18 questions (part A and B) that ask the individual to rate the frequency of different clinical manifestations based on the DSM-5 criteria for ADHD, with responses ranging from 'Never' to 'Very Often'. If the participant scores 4 or more in Part A they are considered at high risk of adult ADHD. Part B provides additional cues. Kessler et al. (2005) reported a sensitivity of 68.7% and a specificity of 99.5% for identification in a population-based sample. The overall sum of scores (0–18) was used for analyses.

Oculomotor battery

The set-up and tasks were administered in the same way as in Sumner et al. (2018).¹ Eye movements were recorded at a sampling rate of 1,000 Hz using the Eyelink 1,000 eye tracker (SR-research). The camera was positioned using the desktop mount placed below the computer screen and a chin rest was used to keep the head stable. The experiment was implemented using Experiment Builder and analyzed using Data Viewer (both SR Research software).

The oculomotor battery comprised four tasks, with participants seated directly in front of the computer monitor at a viewing distance of approximately 80 cm. In each task, a trial started with a fixation cross in the centre of the screen, followed by the stimulus/target consisting of a red circle presented against a black background on the computer screen with 1,024×786 screen resolution. The red circle measured $0.65^{\circ}\times0.65^{\circ}$ visual angle. Written (on-screen) and verbal instructions were provided. Breaks were included between tasks, if necessary.

- (i) In the visual *fixation* task, participants were instructed to maintain their gaze on the target shown in the centre of the screen, until it disappeared. The task lasted for 30 s and began after a drift correct procedure.
- (ii) Two *smooth pursuit* tasks, at differing speeds, were administered. Participants were required to follow the target (i.e., keep their eyes on the target) which had a horizontal sinusoidal motion, moving at 0.2 Hz (slow trial) and then at 0.5 Hz (fast trial). Each trial lasted 20s and was preceded by a drift correct procedure. The target traveled 8.5°/s in the slow trial and 21.5°/s in the fast trial.

Both the (iii) prosaccade and (iv) anti-saccade tasks used a 'step' procedure, meaning that the cue disappeared at the same time as the peripheral target appeared. Each of the 24 trials per task was preceded by a drift correct procedure which then moved on to display the central fixation target. The central target was displayed for 1,000 ms before moving either left or right (on the horizontal meridian 6.25°). The direction of the step was randomized in both tasks and the target was displayed in the new location for 1,000 ms. For the prosaccade task (sometimes referred to as a 'visually-guided' saccade task), participants were instructed to look at the central fixation point and then move their eyes as quickly as possible to the target when it moved from the central point. For the anti-saccade task, the procedure remained the same, but participants were instructed to ignore the target when it moved from the centre of the screen and to look as quickly as possible in the opposite direction. The instructions were explained and then verified with the participant.

Procedure

Ethical approval was obtained from Goldsmiths, University of London, research ethics committee. Participants provided informed written consent prior to attending a research visit. The questionnaire and oculomotor battery were completed in one session, lasting no longer than 45 min. Participants were seen individually in a quiet room, which was dimly lit for the eye tracking tasks. Participants were first introduced to the eye-tracking set up. They took part in a 5-point

¹ Note that Figures showing the experimental set up and the pro- and anti-saccade tasks are as depicted in **Sumner et al.** (2021).

calibration exercise at the beginning of the eye-tracking session, which was repeated as required. The oculomotor test battery was undertaken in a fixed order (as presented above). All participants completed the four oculomotor tasks.

Eye tracking data analysis

Fixation task

Four measures were taken to assess active engagement on the target (e.g., fixation 'stability'): Time on Target (within 1° visual angle, represented as a %); Number of saccades during the 30s task; Average fixation duration; Weighted distance of saccades away from the target.

Smooth pursuit

Eye movements in these trials were quantified using customized software written in LabView. Four key metrics of smooth pursuit performance are presented: Number of qualifying pursuit segments, Sum of durations of qualifying pursuit segments (i.e., time spent in pursuit), Weighted average velocity gain, Weighted average root-meansquare-error (RMSE). Pursuit segments were extracted and the duration of each segment was determined using the online parsing decisions made by the eye tracking software. To identify a pursuit segment (e.g., when the eye was moving/following the target), first samples were recorded as being in a saccade if the sample velocity exceeds 30°/s, or acceleration exceeds 8,000°/s2. All samples that were not classified as being part of a saccade (or a blink - which includes periods of 'tracking loss') were considered as being in a potential pursuit segment. The number of these identified pursuit segments were then calculated per task and individual, as well as identifying the time in pursuit (summing the time of all identified pursuit segments). As set out in Sumner et al. (2018), pursuit gain calculated the ratio of the eye velocity to target velocity (i.e., the average of eye velocity divided by target velocity for each pursuit segment). RMSE was calculated as the square root of the average of eye position (in degrees of visual angle) subtracted from target position (in degrees of visual angle) squared. Any pursuit segments with velocity gain values below 0.5 or above 1.5 were removed prior to analysis, as were pursuit segments less than 100 ms in duration, and with RMSE values of above 2. For each target speed, the average Gain and RMSE measures were weighted by the duration of pursuit segments.

Prosaccade and anti-saccade

For trials to be considered 'valid' for analysis, participants had to be fixating on the central fixation point at target onset and the start time of the first saccade had to be $>\!80\,\mathrm{ms}$ (i.e., not anticipatory). Two measures were calculated for both tasks: saccade latency (ms) and percentage of direction errors. In addition, accuracy (in terms of amplitude – i.e., how close the eye movement was to the target - reported in degrees of visual angle), was also measured in the prosaccade task. This calculation is based on the screen pixel co-ordinates of the gaze data, using parameters for screen height, width, distance and pixel resolution.

Statistical analysis

Normality of data were checked considering the histograms, Q-Q plots and the Shapiro–Wilk's W test (p > .05 indicating normally distributed data). Parametric tests (ANOVAs) were conducted to

explore group differences in the eye tracking measures on normally distributed data, while non-parametric equivalents (Mann Whitney) were conducted for non-normally distributed data. The sample size reported here is similar to that reported in Sumner et al. (2018), $n=24\,\mathrm{DCD}$ children. As adults with pDCD differed from the comparison group in terms of age and the ASRS score, Bivariate correlations (using the whole sample) were conducted with the eye tracking measures to determine whether these variables should be controlled for in subsequent analyses.

Results

Table 1 presents the age and ASRS scores for the two groups of participants, as well as the group comparison statistics. The pDCD group was significantly older than the TD group. Although none had reported a diagnosis, a higher proportion of adults with pDCD (n=14, 66.6%) met the criteria for in-depth ADHD evaluation according to the ASRS checklist (e.g., scoring ≥ 4 on Part A), compared to just one participant in the TD group (4.7%). Considering the overall ASRS score, adults with pDCD scored significantly higher than the comparison group; although of note, a range of scores are observed for the pDCD group.

Bivariate correlations were conducted using age and the ASRS overall score along with the subsequent eye tracking measures reported below. Age and ASRS were found to be positively correlated (r = 0.59, p < .001). Bonferroni adjustment of the level of significance was calculated based on the 16 eye tracking measures used for the correlational analysis (0.05/16 = p < .003). Based on a significance level < .003, neither age nor the ASRS score were found to correlate with any of the eye tracking measures. Due to the lack of significant correlations and the distribution of data for some of the measures which meant that non-parametric tests needed to be conducted, age and ASRS have not been included as covariates in the subsequent analyses.

Fixation

The fixation findings are shown in Table 2. Mann Whitney U-tests were conducted due to the data not being normally distributed. Significant group differences were reported for the number of saccades, average fixation duration and time on target; with adults with pDCD making more saccades during the fixation task, fixating less and spending less time on the target than the TD group. No group

TABLE 1 Participant characteristics.

	TD (n = 21)	DCD (n = 21)	Group comparison
Age in years			
Mean (SD)	21.81 (4.72)	29.86 (7.91)	t (4,40) = 8.77,
Range	18-32	21-46	p < 0.001
ASRS overall score			
Mean (SD)	4.57 (1.32)	8.42 (4.62)	U = 68.00, p < 0.001
Range	3–7	1-18	

ASRS, ADHD Self-Report Scale, overall score maximum of 18.

differences were observed for the distance the eyes moved from the target.

group difference was found for anti-saccade error rate, with adults with pDCD making more errors.

Smooth pursuit

The two trials (slow and fast) for the smooth pursuit tasks are reported in Table 3. Two-way repeated measures ANOVAs [2 (group: TD, pDCD) x 2 (trial: slow, fast)] were conducted for each of the four measures. Significant Group x Trial interactions were identified in the analysis of duration of time spent in pursuit, F(1,40) = 8.39, p = .006, $n_p^2 = 0.17$, and pursuit gain, F(1,40) = 4.69, p = .03, $n_p^2 = 0.11$. For pursuit duration, there was a significant effect of group, F(1,40) = 6.19, p = .02, $n_p^2 = 0.13$, and the same for the gain value, F(1,40) = 3.80, p = .05, $n_p^2 = 0.09$. For both duration and gain measures, the interaction effects revealed that adults with pDCD showed poorer performance than the TD group in the faster trial (less time in pursuit: p = .006, and lower gain: p = .03).

The Group x Trial interactions were not significant for the measures of pursuit segments, F(1,40) = 3.42, p = .07, $n_p^2 = 0.08$, or RMSE, F(1,40) = 0.82, p = .37, $n_p^2 = 0.02$; nor were there any significant overall group differences, F(1,40) = 0.03, p = .87, $n_p^2 = 0.00$ and F(1,40) = 0.02, p = .89, $n_p^2 = 0.00$, respectively.

Pro- and anti-saccade

Table 4 reports the pro- and anti-saccade findings. Analyses were based on 'valid' trials, as discussed in the Methods section. For the prosaccade task, the mean (SD) and range of valid trials out of 24 per group were: pDCD, M = 20.04 (2.87), 14-24; TD, M = 21.38 (2.99), 12-24; and for the antisaccade task also out of 24 pDCD, M = 21.19 (2.78), 15-24; TD, M = 22.14 (2.10), 16-24. The two groups performed similarly on prosaccade latency, but the TD group had significantly shorter latencies in the anti-saccade task than adults with pDCD. No group difference was found for prosaccade amplitude. A significant

TABLE 2 Group comparisons on the fixation measures.

	TD (n = 21)	DCD (n = 21)	Group comparison
Number of saccades			
Mean (SD)	16.43 (8.74)	32.62 (21.75)	U = 128.50,
Range	5-37	9–79	p = .02
Average fixation			
duration (s)			
Mean (SD)	2.20 (1.26)	1.28 (0.83)	U = 126.00,
Range	0.73-4.97	0.29-2.92	p = .01
Time on target (s)			
Mean (SD)	27.46 (17.12)	25.13 (5.47)	U=139.00,
Range	17.12-29.96	11.06-29.69	p = .04
Weighted distance			
from target			
Mean (SD)	13.61 (10.84)	27.53 (51.05)	U=206.00,
Range	2.55-46.19	2.89-185.67	p = .72

s, seconds.

Discussion

The aim of the present study was to consider oculomotor function in adults with probable DCD. This brief report presents the first analysis of the oculomotor profile of adults with pDCD through the administration of a battery of tasks assessing eye movements (e.g., fixations, saccades and smooth pursuit) and high-level (e.g., inhibitory control) cognitive control processes involved in oculomotor control in adults with and without pDCD. Using the same tasks as reported in Sumner et al. (2018) further allows for an indirect comparison to the profile of primary schoolaged children with DCD who were the focus of that study. Encouragingly, the present findings revealed that the underlying mechanisms for preparing and executing saccades (pro-saccade task: initiating an eye movement to a target and arriving at that target) and engaging in pursuit of a slow moving target were found to be comparable between adults with and without pDCD. This finding means that low level oculomotor processes could be considered to be intact in adults with pDCD.

However, notable difficulties were observed between adults with and without pDCD in the tasks that required fixation to one position on the screen and suppressing eye movements (fixation task) or suppressing a reflexive saccade (anti-saccade task). In both of these tasks, adults with pDCD were shown to have difficulty with saccadic inhibition. It could also be argued that the finding of adults with pDCD spending less time pursuing the target in a fast smooth pursuit trial and lower gain compared to the typically developing group points toward a higher number of saccades (eye movements) being made. Together these findings suggest a difficulty with exerting top-down cognitive control, which may be attributed to interference with the fronto-parietal networks and under-development of a control network important for saccade inhibition (Gonzalez et al., 2016). Parallels can be drawn to both research findings and the self-evaluative descriptions of those with DCD identifying challenges for many across a range of executive function tasks (including inhibition) both in the lab and in daily life (e.g., Tal Saban et al., 2014; Bernardi et al., 2018; Scott-Roberts and Purcell, 2018; Sartori et al., 2020; Lachambre et al., 2021; Abdollahipour et al., 2023; Fogel et al., 2023). Moreover, emerging data suggests altered neural structure, structural and functional connectivity and neurophysiological activity across multiple brain regions in people with DCD, within and across sensori-motor and prefrontal regions (e.g., Wilson et al., 2017; Rinat et al., 2020; Subara-Zukic et al., 2022). While such differences are also apparent in people with ADHD, there is emerging evidence of shared and distinct neural correlates in each diagnostic group (Kangarani-Farahani et al., 2022).

The findings of low-level oculomotor processes being intact (e.g., preparing and executing saccades), but disturbances in saccadic inhibition shown in adults with pDCD echo those from a child sample (Sumner et al., 2018). Interestingly, Sumner et al. found group differences between children with and without a diagnosis of DCD in the measure of pursuit duration for both slow and fast pursuit trials, whereas adults in the current sample appeared only to have difficulty with the faster trial. Adults with pDCD demonstrated compromised (slow) processing of online feedback when responding to a faster paced target, as the task requires online prediction of the target

TABLE 3 Group comparisons on the smooth pursuit tasks.

	TD (n = 21)	DCD (n = 21)
Slow		
Number of segments		
Mean (SD)	21.62 (7.32)	25.29 (6.08)
Range	12-36	13-43
Duration (s)		
Mean (SD)	15.53 (2.41)	14.37 (2.66)
Range	9.21-18.22	8.56-17.66
Weighted gain		
Mean (SD)	1.03 (0.05)	1.00 (0.08)
Range	0.96-1.16	0.84 - 1.14
Weighted RMSE		
Mean (SD)	0.67 (0.31)	0.64 (0.25)
Range	0.31-1.45	0.26-1.11
Fast		
Number of segments		
Mean (SD)	34.48 (8.54)	31.57 (13.84)
Range	22-52	10-60
Duration (s)	14.15 (3.17)	10.69 (4.49)
Mean (SD)	8.57-18.21	2.32-16.33
Range		
Weighted gain		
Mean (SD)	0.95 (0.09)	0.88 (0.11)
Range	0.78-1.21	0.64-1.03
Weighted RMSE		
Mean (SD)	1.04 (0.19)	1.02 (0.14)
Range	0.68-1.39	0.79-1.30

TABLE 4 Group comparisons on the prosaccade and antisaccade tasks.

	TD (n = 21)	DCD (n = 21)	Group comparison
Latency			
Prosaccade (ms)			
Mean (SD)	159.24 (12.54)	164.60 (34.04)	t(40) = 0.67,
Range	139.79-180.05	122.73-271.76	p = .50
Antisaccade (ms)			
Mean (SD)	232.10 (29.87)	268.41(58.56) ^a	t(39) = 2.52,
Range	183.08-285.29	87.50-355.75	p = .02
Amplitude			
Prosaccade			
Mean (SD)	6.87 (0.51)	6.79 (0.43)	t(40) = -0.53,
Range	5.48-7.73	5.83-7.56	p = .59
Error rate			
Antisaccade (%)			
Mean (SD)	15.19 (12.67)	41.85 (29.17)	U = 84.50,
Range	0-45	9-100	p = .001

^{*1} missing data point due to timing recording error; amplitude in degrees, the target moved 6.25° to the left or right.

position. This may suggest a slowed process of natural development over time or the development of an alternative approach to slow pursuit in those with DCD. Considered alongside some evidence of brain based changes in children with DCD emerging through targeted intervention (Izadi-Najafabadi and Zwicker, 2021), and given the importance of oculomotor control for effectively completing activities of daily living, this possibility warrants further investigation.

Difficulties with attention and saccadic inhibition, evidenced both in a child sample (Sumner et al., 2018) and the current adult sample, may be one explanation for the challenges that individuals with DCD experience in the acquisition of skilled behaviors that often rely on visual attention. Al-Yahya et al. (2023) highlight that interventions targeting motor skill training for individuals with DCD often demonstrate variable performance and suggest that we need to consider the underlying mechanisms of DCD to further intervention approaches. Inhibitory control supports the execution of day-to-day tasks and these oculomotor control mechanisms support the allocation of visual attention (e.g., directing saccades to stimuli; Gonzalez et al., 2016). Often movement requires rapid processing of visual information. For example, Wood et al. (2017) reported improvements in ball catching for children with DCD following training of saccadic and fixation behaviors. This finding supports a link between oculomotor control and guided motor performance and may be an area for future intervention research.

While the current study is a preliminary first step to explore oculomotor performance in adults with pDCD, limitations can be acknowledged. It was not possible to conduct an assessment of motor skill to confirm the diagnosis of DCD and we relied on selfreport of a diagnosis. Knowledge of when participants' received their diagnosis of DCD may have been useful to consider, as it could be that early diagnosed individuals may have received support and developed their skills in different ways to those with later diagnoses. Five pDCD participants reported a dyslexia diagnosis and while these two conditions are known to co-occur it was not possible to consider the implications of this within the study although this may be of interest in the future given reports of subtle inhibition difficulties noted in oculomotor tasks completed by adults with dyslexia (Wilcockson et al., 2019). Further, the increased prevalence of attention difficulties (measured by the ASRS) in the pDCD group may raise questions about potential interactions with attention difficulties and the impact on the findings. However, of note, data reported by Tal Saban et al. (2014) suggests that in a sample of 284 young adults with DCD and those categorized as having borderline DCD, reported problems with executive function remain consistent with or without attention difficulties. It was not possible to control for attention difficulties in the current analysis due to statistical restrictions. However, this measure was not found to correlate with the eye tracking measures and participants did not report a clinical diagnosis of ADHD. Co-occurrence of DCD and ADHD is reportedly high (Blank et al., 2019) and studies have shown a similar pattern of saccadic inhibition difficulties, but no problems with pursuit of a target in individuals with ADHD (Maron et al., 2021). Further research is warranted to understand the overlap between DCD and ADHD in this respect and the impact on cognitive control and motor skill acquisition. Finally, we make indirect cross-sectional comparisons between the findings from existing child data (Sumner et al., 2018) and the current adult data. While the child and adult data that are compared in our evaluation are not drawn from a single study, we argue that they are directly

comparable given that they completed identical oculomotor tasks in the same lab. Future research in this area should seek to examine oculomotor performance longitudinally in individuals with DCD, explore sex differences of oculomotor control in DCD, as well as relating oculomotor performance to measures of fine and gross motor skill performance.

To conclude, the current findings provide evidence of oculomotor disturbances in adults with pDCD particularly in relation to eye movements that engage top-down cognitive control processes (i.e., saccadic inhibition and maintaining gaze to a central fixation point). Given the criticality of inhibitory control for managing day-to-day tasks and the consequent impact on activities of daily living, academic and employment outcomes, as well as broader factors such as mental health and wellbeing, these findings highlight the need for recognition of the impact of DCD and the ongoing challenges in adulthood. They are also likely to impact on skill acquisition that continues through the lifespan. Further research in this area in relation to both DCD and other neurodevelopmental disorders could demonstrate potential biomarkers of DCD for research and clinical practice, aiding clearer understanding and identification of DCD in adulthood and approaches for intervention.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by the Goldsmiths, University of London, ethics review committee. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

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Author contributions

ES: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Writing – original draft, Writing – review & editing. EH: Funding acquisition, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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