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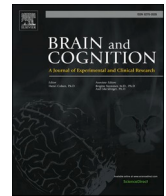
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Theoretical Notes



Atypical procedural learning in children with developmental coordination disorder: A combined behavioral and neuroimaging study

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ABSTRACT

While procedural learning (PL) has been implicated in delayed motor skill observed in developmental coordination disorder (DCD), few studies have considered the impact of co-occurring attentional problems. Furthermore, the neurostructural basis of PL in children remains unclear. We investigated PL in children with DCD while controlling for inattention symptoms, and examined the role of fronto-basal ganglia-cerebellar morphology in PL. Fifty-nine children (6–14 years; $n_{\text{DCD}} = 19$, $n_{\text{control}} = 40$) completed the serial reaction time (SRT) task to measure PL. The Attention-Deficit Hyperactivity Disorder Rating Scale-IV was administered to measure inattention symptoms. Structural T1 images were acquired for a subset of participants ($n_{\text{DCD}} = 10$, $n_{\text{control}} = 28$), and processed using FreeSurfer. Volume was extracted for the cerebellum, basal ganglia, and frontal regions. After controlling for inattention symptoms, the reaction time profile of controls was consistent with learning on the SRT task. This was not the case for those with DCD. SRT task performance was positively correlated with cerebellar cortical volume, and children with DCD trended towards lower cerebellar volume compared to controls. Children with DCD may not engage in PL during the SRT task in the same manner as controls, with this differential performance being associated with atypical cerebellar morphology.

1. Introduction

Developmental coordination disorder (DCD) is a common neurodevelopmental disorder characterized by a markedly impaired ability to learn, and perform, age-appropriate motor skills (American Psychiatric Association [APA], 2013). Affecting approximately 5–6% of school-aged children (Blank et al., 2019), children with DCD experience functional difficulties related to their motor incoordination (e.g., dressing, eating with cutlery, self-care etc.; Licari et al., 2020). The impact of DCD also extends beyond the motor domain, impacting broader psychosocial, academic, and medical outcomes (Blank et al., 2019). Current interventions to improve outcomes in DCD are limited in their efficacy, reflecting poor understanding of the underlying mechanisms responsible for motor difficulties in childhood. Clarifying the neurocognitive underpinnings of DCD is central to the development of a unified account of childhood motor skill, and the early identification of atypical motor skill (as in DCD).

1.1. Procedural learning in DCD

One prominent theory proposes that a deficit in procedural learning (PL) may contribute to the atypical motor skill observed in DCD (Nicolson & Fawcett, 2007). Briefly, PL describes the implicit acquisition and representation of skills and knowledge by repeated execution of serially ordered movements (e.g., Fiser & Aslin, 2002; Nissen & Bullemer, 1987). Fundamental tasks of everyday living that are thought to depend on PL (or related) mechanisms, such as dressing, eating with cutlery, self-care, drawing/writing and using scissors, are reportedly difficult for children with DCD (Licari et al., 2020). This has led to the proposal that atypical PL may, at least in part, subservise the delayed motor development observed in children with DCD (see Biotteau et al., 2016; Bo & Lee, 2013).

PL in DCD has been predominantly studied via the serial reaction time (SRT) task (Nissen & Bullemer, 1987). Briefly, the task requires participants to respond as quickly and accurately as possible to stimuli appearing at different locations on a screen by pressing corresponding

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keys on a keyboard. The visuospatial location of the stream of stimuli follows a predetermined repeating sequence, of which the participants are unaware. However, across successive blocks of sequence trials, participants tend to become faster at responding to the stimuli, despite the lack of awareness of a pattern in the stimuli presentation. This is typically thought to reflect subliminal learning of the repeating sequence (Nissen & Bullemer, 1987). In support, when the sequence blocks are interrupted by a block of trials presented in a pseudorandom order, participant reaction times (RTs) tend to increase significantly (*viz.*, the rebound effect). Where the prototypical decrease in RT is observed across the initial sequence blocks, the latter rebound effect is thought to provide insight into PL processes. Here, a larger rebound effect is interpreted as better learning proficiency (Janacek & Nemeth, 2013; Robertson, 2007).

Several experimental studies have explored the nature of PL in children with DCD using the SRT task (Blais et al., 2021; Gheysen et al., 2011; Lejeune et al., 2013; Van Dyck et al., 2022; Wilson et al., 2003). Initially, Wilson and colleagues (2003) reported no significant differences between children with DCD and controls in the magnitude of the rebound effect. Since children were unaware of the sequence embedded in the SRT task, the authors suggested that children with and without DCD were equally able to engage in PL. However, the group difference on the rebound effect approached significance ($p = .092$), and given the small sample size of $N = 10$ children per group, these results reasonably indicate a trend towards reduced PL in DCD. Subsequent findings on this topic have been highly varied (Blais et al., 2021; Gheysen et al., 2011; Lejeune et al., 2013), with work suggesting both preserved and atypical PL in those with DCD. Some variation may come from whether participants became aware of the sequence embedded in the task during performance. In most studies, children reported explicit awareness of the sequence embedded in the task (Gheysen et al., 2011; Lejeune et al., 2013). In others, there was no record of whether children became aware of the sequence (Blais et al., 2021). More recently, Van Dyck et al. (2022) reported reduced PL in children with DCD compared to controls, which could not be explained by explicit sequence awareness. This latter finding is consistent with a broader trend whereby atypical PL appears to be more consistently observed in DCD where participants remain unaware of the repeating sequence during the SRT. The implicit/explicit distinction is critical, since implicit learning is a core tenet of PL, and is neurocognitively distinct to explicit learning (see Hardwick et al., 2013). In sum, the literature does not provide a consensus on PL ability in DCD, though it appears that children with DCD may be more likely to present with performance decrements on the SRT task where there is evidence that they remained unaware of the repeating sequence (as per PL).

1.2. The impact of inattention symptoms on procedural learning in children with DCD

While PL is fundamental to skill acquisition, it is core to the automation of broader social and cognitive processing (Hamrick et al., 2018; Lieberman, 2000). This argument is supported by recent evidence demonstrating a strong relationship between attention and SRT task performance in children with developmental disorders (West et al., 2020). This finding is especially relevant for children with DCD, where approximately 50% of children with DCD also present with Attention-Deficit Hyperactivity Disorder (ADHD) – a disorder characterized by symptoms of inattention (Goulardins et al., 2015). Despite this, the existing literature on PL in DCD is varied in the way it considers symptoms of inattention, which may contribute to the mixed findings reported. In some cases, a prior confirmed diagnosis of ADHD served as an exclusionary criterion (Gheysen et al., 2011; Wilson et al., 2003). However, we know that ADHD may be mis-, or under-, diagnosed in some groups – e.g., females are less likely to be diagnosed with ADHD in a clinical setting compared to their male counterparts (Hinshaw et al., 2021; Mowlem et al., 2019). As such, inattention may still impact PL performance in DCD where a sample excluded participants based on a

prior ADHD diagnosis alone. Other work has screened participants with DCD for ADHD symptoms, excluding those above a defined cut-off (Lejeune et al., 2013). Still, those with sub-clinical ADHD symptoms often experience similar motor and psychosocial problems to those who may meet clinical criteria for ADHD (see Kirova et al., 2019 for review). Thus, while a child with DCD may not meet the criteria for ADHD, they may still experience elevated inattention and impulsivity symptoms relative to their peers, which may affect their cognitive and behavioral performance (such as on the SRT task). In support, even where children with comorbid DCD and ADHD have been excluded, significant correlations have been shown between attentional scores and retention of the sequence on the SRT task in those with and without DCD (Blais et al., 2021). This indicates that attention may impact motor learning more broadly. In-keeping with one recent approach (Van Dyck et al., 2022), inattention might be best operationalized as a continuous measure when examining the confounding effect of inattentive symptoms on PL in children with DCD, rather than a categorical measure (*i.e.*, ADHD vs non-ADHD), as done in earlier work.

1.3. The neural basis of procedural learning

Finally, there is little evidence speaking to the neural mechanisms that underlie potential PL deficits in children with DCD. In healthy adults, there is a strong body of research suggesting involvement of fronto-basal ganglia-cerebellar circuitry during SRT task performance (Baetens et al., 2020; Janacek et al., 2020). This system contributes to sequence learning by driving processes such as internal model formation, learning predictable sequential orders, sensorimotor integration, and error correction (Penhune & Steele, 2012). While no studies, according to our knowledge, have considered the role of these regions in SRT task performance (or PL) in children with DCD, it has long been argued that the cerebellum may represent a key neural focus for DCD (Brown-Lum & Zwicker, 2015; Cantin et al., 2007; Subara-Zukic et al., 2022). Given recent work implicating alterations of this subcortical region with variation in behavior and cognition in both healthy children (Mous et al., 2017; Wilke et al., 2003) and those with DCD (Gill et al., 2022; Reynolds et al., 2017), it is plausible that morphology of the cerebellum (and broader fronto-basal ganglia-cerebellar circuitry) might subservise atypical SRT task performance (and PL) in those with DCD. To date, no study has explored this hypothesis. Clarifying this question may provide insight into the neurocognitive systems by which motor learning processes may differ between those with and without DCD.

1.4. The present study

This study had two aims. First, we wanted to examine PL performance in children with DCD, and determine whether any deficits held after accounting for attentional problems. Participants completed a modified SRT task as a measure of PL. Given the common co-occurrence of DCD and ADHD, we controlled for the impact of inattention symptoms on PL performance in our sample. We hypothesized that children with DCD would present with atypical PL on the SRT task after controlling for inattention symptoms, evidenced by a significantly smaller rebound effect compared to controls.

Second, we wanted to explore the role of fronto-basal ganglia-cerebellar morphology in PL in this population. A subset of children underwent structural MRI, and we computed volume of the regions of the fronto-basal ganglia-cerebellar circuit – which has previously been implicated in SRT task performance in healthy adults (Baetens et al., 2020; Janacek et al., 2020). We expected to observe significant positive correlations between morphology in the fronto-basal ganglia-cerebellar circuit and SRT task performance at the whole-group level, evidenced by higher volume in our regions of interest being associated with a larger rebound effect (*i.e.*, more proficient learning) on the SRT task.

2. Methods

2.1. Participants

The present study reports on a subset of data collected as part of a larger, ongoing study. Participants were 22 children with DCD ($M_{Age} = 10.27, SD_{Age} = 2.43, 11$ female) and 44 controls ($M_{Age} = 9.75, SD_{Age} = 2.31, 20$ female), aged 6–14 years. Five participants were removed due to missing ADHD Rating Scale-IV data. A further two participants were removed for non-compliance with SRT task instructions. The sample submitted to analyses were 19 children with DCD ($M_{Age} = 10.63, SD_{Age} = 2.34, 9$ female) and 40 controls ($M_{Age} = 9.53, SD_{Age} = 2.26, 18$ female), aged 6–14 years. There were no significant differences between groups in age ($p = .088$), sex ($p = .515$), or intellectual ability ($p = .373$; measured using FSIQ-2 t -scores on the WASI-II [Wechsler, 2011]). 18% of the control group ($n = 7$) and 5% of the DCD group ($n = 1$) were left-handed, with no significant differences between groups in handedness ($p = .200$). Participants were recruited via physical and online flyers. Children with DCD were also recruited through Occupational Therapists. All participants gave informed consent and were reimbursed for their time. The Deakin University Human Research Ethics Committee approved the experimental procedures (2019–009). See Fig. 1 for a flowchart showing participant numbers at each stage of the study.

Children with DCD were screened against the DSM-5 criteria (APA, 2013). Specifically, motor proficiency was significantly below that

expected of the child’s age (Criterion A), indicated by a score at, or below, the 16th percentile on the short-form of the Bruininks-Oseretsky Test of Motor Proficiency 2nd ed. (BOT-2 SF; Bruininks & Bruininks, 2005). The BOT-2 SF is a standardized measure of motor ability comprising 14 subtests that assess a range of fine and gross motor skills. Scores are age-normed to provide participants with a percentile ranking of their motor ability relative to other children their age. The BOT-2 SF is a well-validated measure for detecting atypical motor skill in children aged 4–21 (Bruininks & Bruininks, 2005; Byrial et al., 2022; Cairney et al., 2009; Carmosino et al., 2014). We note that one child with a pre-existing DCD diagnosis did not score at, or below, the 16th percentile on the BOT-2 SF when screened by our researchers. However, given that a formal diagnosis report was provided by clinicians, that the remaining DSM-5 criteria were met (see below), and that their score still fell in the bottom quartile, we opted to include them in our DCD group. Next, identified motor difficulties interfered significantly with their ability to perform daily activities (Criterion B), as indicated by scores on the Developmental Coordination Disorder Questionnaire (DCD-Q; Wilson et al., 2007). The DCD-Q is a brief Likert-scored questionnaire, which parents complete to determine their child’s capacity to engage in day-to-day activities involving movement. In the absence of Australian norms for DCD cut-offs, we used an approach adopted in our earlier work in adults with DCD (Barhoun et al., 2021; Hyde et al., 2014, 2018). That is, based on DCD-Q scores of all control children from the larger study from which our sample was derived ($N = 50$), we identified the 95%

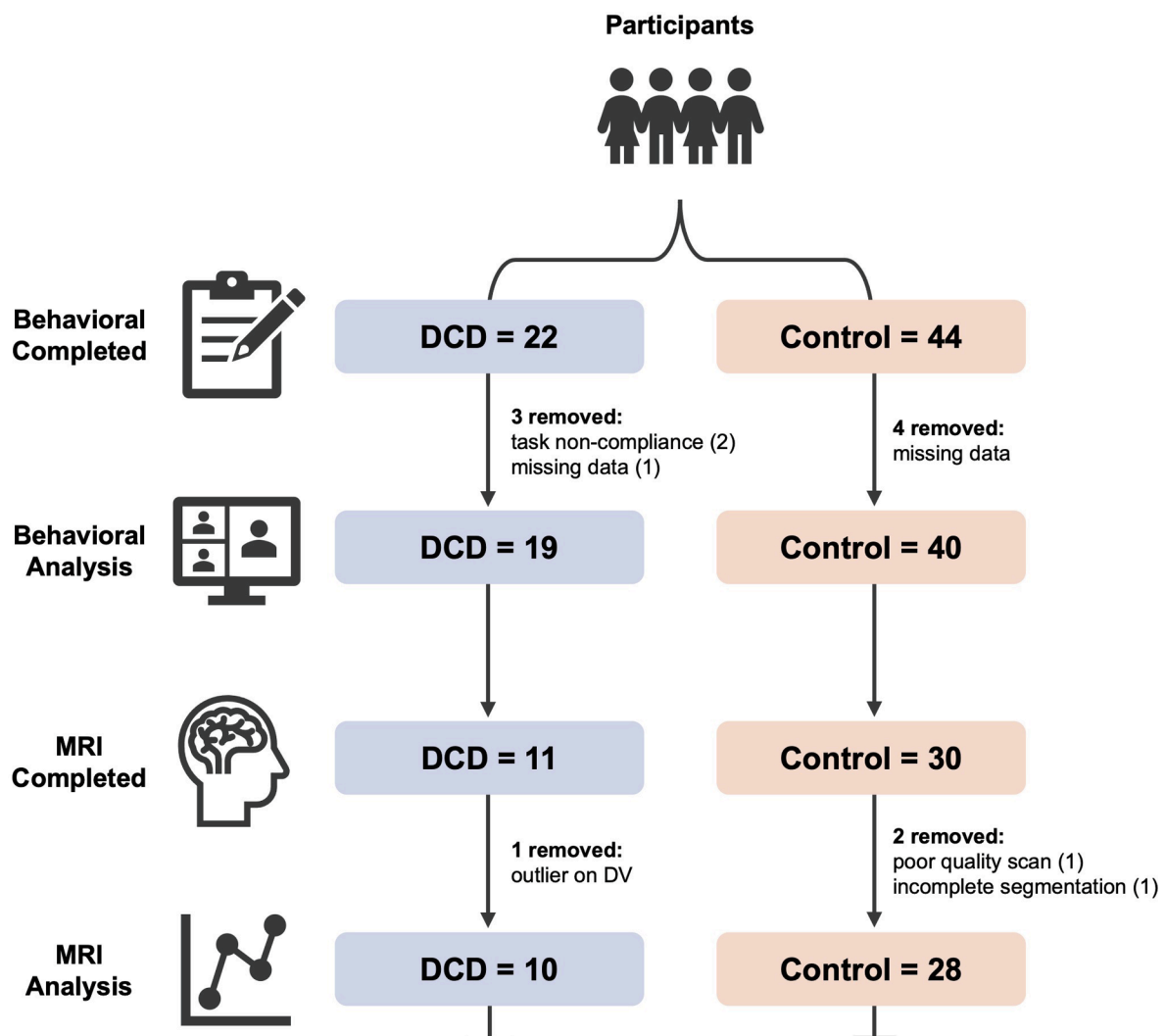


Fig. 1. Flowchart of participant numbers throughout the study.

confidence intervals ($CI_{95\%}$) for the DCD-Q scores for those aged 5–7 ($CI_{95\%}$: 59.75 ± 3.48) aged 8–9 ($CI_{95\%}$: 64.00 ± 4.44), and aged 10–14 ($CI_{95\%}$: 65.05 ± 4.02). Children in the DCD group who scored below the lower range were deemed to have met Criterion B (i.e., motor related deficits of everyday living). This was the case for all participants in our DCD group. Next, since the study involved child participants, the onset of a child's symptoms could be said to have occurred during childhood (Criterion C). Finally, motor skill difficulties were not otherwise attributable to any medical or neurodevelopmental disorder (Criterion D), ascertained by parent consultation that their child does not present with a significant condition that might otherwise explain their motor ability (APA, 2013). Children in the control group had age-appropriate motor ability, all scoring above the 16th percentile on the BOT-2 SF (Bruininks & Bruininks, 2005).

2.2. Measures and procedure

2.2.1. ADHD symptom profile

To provide a measure of inattention symptoms, parents completed the ADHD Rating Scale-IV (ADHD RS-IV; DuPaul et al., 1998). The ADHD RS-IV obtains parent ratings on a 4-point Likert scale regarding the frequency of their child's inattention and hyperactivity/impulsivity symptoms based on DSM-IV criteria (DuPaul et al., 1998). Parents are asked to determine symptomatic frequency that describes their child's behavior at home over the previous 6 months. The 18-item scale consists of two subscales. The Inattention subscale is scored by summing the responses on all the odd-numbered items. The Hyperactivity-Impulsivity subscale is scored by summing the responses on all the even-numbered items. A Total Scale raw score can be obtained from the total of the Inattention and Hyperactivity-Impulsivity subscale scores. The ADHD RS-IV is commonly used to screen for ADHD symptoms in research settings, and work that has adopted this measure as a continuous variable has demonstrated significant associations between ADHD symptoms and other behavioral/experimental outcomes (Kim et al., 2022; Montagna et al., 2020; Van Dyck et al., 2022; Wendel et al., 2020). In the present study, we used participant's scores on the Inattention subscale as our measure of inattention symptoms.

2.2.2. The SRT task

The SRT task used in this study was adapted from a version

commonly used by our team (e.g., Lum et al., 2010; Lum & Kidd, 2012). The task was presented using E-Prime 2 software (Psychology Software Tools, Pittsburgh, PA). Participants were seated in front of a 17-inch display and provided with a game controller consisting of four buttons arranged in the shape of a diamond (see Fig. 2). At the beginning of a trial, participants viewed a white screen with four boxes, arranged in a diamond configuration, for 500 ms. A visual stimulus then appeared in one of the four boxes for 800 ms. Using their right thumb, participants were instructed to press one of four diamond-shaped buttons on the controller that matched the visual stimulus' location. Participants could respond any time during the 800 ms period. For responses made before 800 ms, the visual stimulus would stay on the screen for the remaining time. For example, if the participant made a response at 600 ms, the visual stimulus would stay on the screen for a further 200 ms. This was also the case when participants pressed a button on the controller that did not match the stimulus' location. This ensured that the task remained the same length for all participants. Following each response, participants were given feedback in the form of a red border appearing over the box they indicated. Failure to respond within 800 ms was coded as an incorrect response. These events represented one trial. The task consisted of four blocks of 60 trials. Each block was separated by a 3-second rest period in which a white screen appeared on the display.

Participants were unaware that in Blocks 1–3, the visual stimulus' location on each trial followed a pre-determined 10-element sequence. Labelling the left-most position on the computer display as 1, and moving anti-clockwise around the diamond configuration, the sequence was 3–4–1–2–4–1–3–4–2–1. The sequence repeated 6 times resulting in 60 trials in each sequence block. In Block 4, the visual stimulus appeared pseudo-randomly in one of the four positions on the display adhering to the following constraints. First, the visual stimulus could not appear in the same location on two consecutive trials. Second, the number of times the visual stimulus appeared in each of the four spatial locations was the same as for the sequence blocks. Third, the frequency of each pairwise transition in the random block matched the sequence blocks. The randomization was reset at the end of each ten trials. There were a total of 60 trials in the random block.

The version of the SRT task used in the current study differed from the standard version (Nissen & Bullemer, 1987). Here, a different visual stimulus appeared on each trial within a block. The stimuli comprised 60 different shapes (circles and polygons), presented in different colors

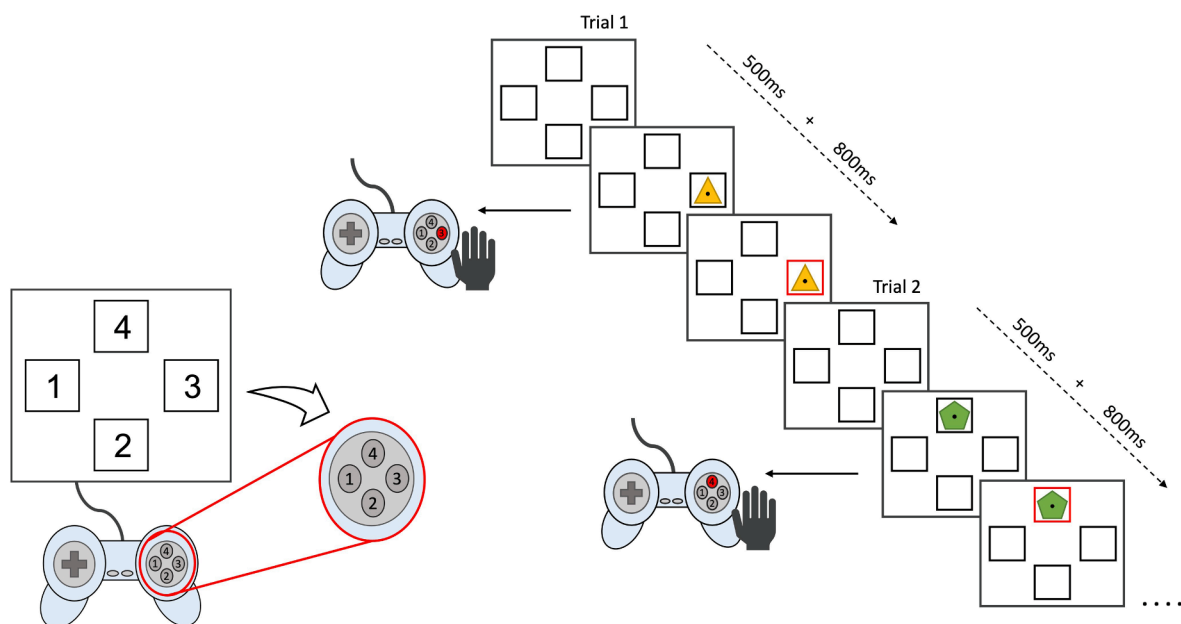


Fig. 2. Schematic overview of the serial reaction time (SRT) task (adapted from Lum et al., 2010). Left: shows the locations that the visual stimuli could appear on each trial, and the corresponding buttons on the controller used as the response device. Right: provides timing details on two trials.

(purple, green, blue, red, orange). On each trial within a block, a visual stimulus was selected without replacement. Thus, in each block, the order that the 60 visual stimuli were presented was different. Manipulating the experimental stimuli in this manner changes the perceptually salient features of the stimuli relative to traditional versions of the task. This has shown to substantially reduce the likelihood of explicit sequence knowledge in adults whose behavioural profiles otherwise suggest that learning has taken place during sequence blocks (Koch et al., 2020; Lum, 2020). In support, we randomly asked 20 children within the project immediately after they completed the SRT task whether they noticed a repeating sequence during the task. All children either failed to notice the repeating sequence, and/or were unable to replicate the sequence when prompted. Thus, we were confident that any learning effects observed could reasonably be attributed to implicit, not explicit, learning.

2.2.3. Volumetric data acquisition and processing

A subset of the sample ($N = 41$, $n_{\text{DCD}} = 11$, $n_{\text{control}} = 30$; Fig. 1) underwent MR scanning using a Siemens Prisma 3 T MRI scanner (Erlangen, Germany) at the Florey Institute of Neuroscience and Mental Health, Heidelberg. Children were screened for MRI contra-indicators, and those who were eligible underwent a mock scan to help them acclimate to the MRI environment and reduce anxiety about the scanning session. High resolution T1-weighted MPRAGE images were acquired for each participant using the following parameters: TR = 1900 ms, TI = 900 ms, TE = 2.49 ms, flip angle = 9° , voxel size = 0.9 mm^3 , acquisition matrix 256×256 , FoV = 240 mm, 192 contiguous slices.

Quality control (QC) of T1-weighted MRIs was conducted as per established guidelines (Harvard University Centre for Brain Science, 2019). First, in-scanner QC was conducted in real time to monitor in-scanner movement. In instances where excessive movement took place, scans were conducted a second time. This minimized loss due to poor quality scans. Then, visual inspection of striping, blurring and signal dropout was conducted by the first author. In instances where a second opinion was required regarding the quality of the scan, a senior researcher was brought in to review, and a decision was made. At this stage, one scan was removed due to visible blurring and striping.

The remaining T1 scans were processed using the ‘recon-all’ function of FreeSurfer version 7.4.1 (<https://surfer.nmr.mgh.harvard.edu/>), which has been described in detail in previous literature (Fischl, 2012). Briefly, structural images underwent skull stripping, bias field correction, segmentation of grey and white matter, and reconstruction of cortical surface models. Cortical structures were segmented using the ‘Desikan-Killany atlas’ (Desikan et al., 2006) and subcortical regions were segmented using FreeSurfer’s automatic segmentation tool (the ‘aseg’ parcellation; Fischl et al., 2002). Since earlier work suggests involvement of the fronto-basal ganglia-cerebellar network in SRT task performance (Baetens et al., 2020; Janacsek et al., 2020), we focused on the cerebellum, basal ganglia, and the frontal regions (particularly, pre-motor). Volume of the left and right cerebellar cortices, caudate, putamen, pallidum and thalamus were extracted from the ‘aseg’ parcellations (Fischl, 2012). Volume of the left and right superior frontal gyrus and the caudal middle frontal gyrus were extracted from the ‘Desikan-Killany atlas’ (Desikan et al., 2006) since the pre-motor ROI sits within these gyri (as per Bhojroo et al., 2022). Intracranial volume (ICV) was also extracted from each participant’s T1 image using FreeSurfer (Fischl, 2012).

Segmentations were visually inspected for anatomical plausibility. At this stage, one participant was removed due to incomplete segmentations. The remaining participants ($N = 38$, $n_{\text{DCD}} = 10$, $n_{\text{control}} = 28$; Fig. 1) were submitted to subsequent statistical analyses. Supplementary Materials 1 presents demographic information for this subset of participants, compared to the full sample.

2.3. Design and analysis

Analysis of SRT task data was conducted using the ‘tidyverse’ (Wickham et al., 2019) and ‘lme4’ (Bates et al., 2014) packages in R (R Core Team, 2022). Graphs were created using the ‘ggplot2’ (Wickham, 2011) and ‘sjPlot’ (Lüdtke & Lüdtke, 2015) packages. Herein, sequence blocks 1, 2 and 3 will be referred to as S1, S2 and S3, and the fourth random block will be referred to as R1.

2.3.1. SRT task performance

RTs measured the time taken to provide a manual response following stimulus onset. For each participant, mean RTs were computed for each block. Only RTs associated with a correct response were included in the preceding analyses.

Preliminary analyses were conducted to determine whether children with and without DCD showed evidence of implicit sequence learning, indicated by a reduction in RT from S1 to S3 (see also Bianco et al., 2023; Lum et al., 2019). Here, separate linear mixed models (LMMs) for each group using restricted estimation maximum likelihood (REML) were conducted, with mean RT as the dependent variable, and with inattention symptoms, age, and block (S1 vs. S3) entered as fixed effects. Each model contained a random intercept to account for clustering of RT within individuals. Trend analysis was also conducted to assess whether each group showed a linear trend in RT performance from S1 to S3, which is indicative of implicit learning of the embedded sequence.

To investigate the rebound effect on the SRT task, and compare the magnitude of the rebound effect (RT difference between S3 and R1) across groups, we ran a LMM using REML, with RT as the dependent variable, and with inattention symptoms, age, block (S3 vs. R1), and block \times group (DCD vs. control) as fixed effects. As in the previous model, this model contained a random intercept, and trend analysis was conducted to test for a linear increase in RT from S3 to R1 (i.e., a rebound effect).

Accuracy on the SRT task was quantified using two measures: the number of errors made where participants pressed the wrong button (herein referred to as “wrong press errors”), and the number of errors made where participants did not respond to stimuli within the allocated time of 800 ms (herein referred to as “omission errors”). Each error type was subjected to separate LMMs using REML, with error type (wrong press errors or omission errors) as the dependent variable, and with block (S1, S2, S3, R1), group (DCD vs. control), and block \times group as fixed effects. The model contained a random intercept to account for clustering of accuracy within individuals. Trend analysis was also conducted to assess whether each group showed a linear trend in accuracy across blocks. These results are presented in Supplementary Materials 2. Lastly, we note in the results that we observed a significant contribution of age to RT in all models. Further exploration is presented in Supplementary Materials 3.

2.3.2. The association between brain volume and SRT task performance

For the dependent measure of SRT task performance, the standardized rebound effect was used (Knopman & Nissen, 1991; Robertson, 2007; Siegert et al., 2006). The calculation of this metric has been described in our previous work (Bianco et al., 2023). Briefly, raw RTs for each trial were transformed to z-scores to control for general processing speed (Janacsek et al., 2012; Koch et al., 2020). Then, mean z-scores were calculated for each block, resulting in a mean z-score for RT for each block, for each participant. The magnitude of the rebound effect was calculated for each participant by subtracting the mean z-score for RT for S3 from the mean z-score for RT for R1. At this stage, one participant from the DCD group was removed because their standardized rebound effect value fell more than three standard deviations below the mean. Where performance across initial sequence blocks indicated that learning had taken place (i.e., a successive decrease in RT), PL is inferred if positive values are obtained for the standardized rebound effect (i.e., RTs are higher in the random block [R1] than in the last sequence block [S3]).

Table 1
ADHD RS-IV Inattention Symptom Scores, By Group.

	Control	DCD
N	40	19
Mean	8.70	11.16
Standard deviation	7.18	6.27
Min	0	3
Max	24	21

Next, we explored the association between volume in our ROIs and SRT task performance across the entire MRI sample ($N = 38$). Pearson's correlations were conducted between the standardized rebound effect and volume for frontal, basal ganglia, and cerebellar regions. Each of these regions were considered a separate group of analyses for the purpose of multiple comparisons, whereby each group of correlations were adjusted for multiple comparisons using the Benjamini-Hochberg method (Benjamini & Hochberg, 1995). We did not include age, sex, handedness, or intracranial volume as covariates, since there were no significant associations between these variables and the standardized rebound effect (see Supplementary Materials 4). Where significant associations were observed between volume and the rebound effect (e.g., the bilateral cerebellum – see Results) we were underpowered to conduct group-specific correlation analyses to determine whether the effects of interest differed according to group. However, we plotted the line of best fit for each group separately to visualize these relationships for exploratory reasons (see Supplementary Materials 5).

Lastly, where significant associations between ROI volume and the rebound effect were observed (i.e., the bilateral cerebellum), we were then interested in whether there was a significant difference in volume in these regions between the DCD and control groups. As such, two separate independent samples t -tests were conducted to compare volume in the left and right cerebellar cortices between these groups.

3. Results

3.1. SRT task

Relevant assumptions of multicollinearity (variance inflation factors [VIFs] < 5), linearity (scatterplots), homoscedasticity (homogeneity of residuals plot) and normality (residual histograms and Q-Q plots) were

met for RT data. No outliers were removed. Table 1 presents summary data for inattention scores, separated by group, on the ADHD RS-IV. Fig. 3 presents the RT data for the DCD and control groups, reported by block.

3.1.1. Examining learning effects in controls: Comparison of RT from S1 to S3

The LMM revealed a significant contribution of age to RT, $B = -21.44$, $SE = 3.51$, 95 % CI [-28.31, -14.56], $t(37) = -6.11$, $p < .001$, suggesting better learning across sequence blocks in the younger control participants. The model revealed no significant contribution of inattention symptoms to RT, $B = -0.70$, $SE = 1.11$, 95 % CI [-2.86, 1.47], $t(37) = -0.63$, $p = .532$. After controlling for age and inattention symptoms, trend analysis showed a significant linear main effect for block, $B = -11.08$, $SE = 3.92$, 95 % CI [-18.76, -3.41], $t(39) = -2.83$, $p = .007$, whereby RT decreased significantly from S1 ($M = 454.43$, $SD = 75.42$) to S3 ($M = 438.75$, $SD = 64.33$). This effect suggests that the control group were able to learn the sequence embedded in the task.

3.1.2. Examining learning effects in children with DCD: Comparison of RT from S1 to S3

The LMM revealed a significant contribution of age to RT, $B = -17.99$, $SE = 6.38$, 95 % CI [-30.50, -5.48], $t(16) = -2.82$, $p = .012$, suggesting better learning across sequence blocks in the younger DCD participants. The model revealed no significant contribution of inattention symptoms to RT, $B = 1.94$, $SE = 2.38$, 95 % CI [-2.73, 6.60], $t(16) = 0.81$, $p = .428$. After controlling for age and inattention symptoms, trend analysis failed to detect a significant linear main effect for block, $B = 1.89$, $SE = 6.54$, 95 % CI [-10.92, 14.70], $t(18) = 0.29$, $p = .776$, with RT remaining consistent from S1 ($M = 428.85$, $SD = 66.93$) to S3 ($M = 431.52$, $SD = 81.97$). This effect suggests that the DCD group did not show the RT profile consistent with implicit sequence learning.

3.1.3. Rebound effect: Comparing RT from S3 to R1 between the DCD and control groups

The LMM revealed a significant contribution of age to RT, $B = -22.87$, $SE = 2.79$, 95 % CI [-28.35, -17.40], $t(55) = -8.19$, $p < .001$, suggesting that the magnitude of the rebound effect decreased with age. The model revealed no significant contribution of inattention symptoms to RT, $B = -0.12$, $SE = 0.92$, 95 % CI [-1.93, 1.69], $t(55) = -0.13$, $p = .899$. After controlling for age and inattention symptoms, trend analysis

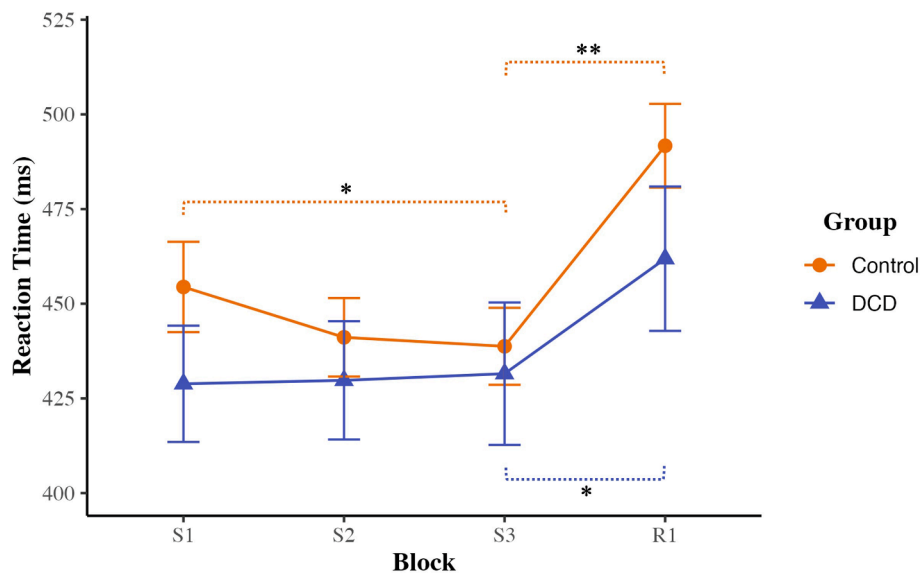


Fig. 3. Mean reaction time for the DCD group (blue triangle; $n = 19$) and control group (orange circle; $n = 40$) across blocks. Error bars show standard error. The first three blocks (S1, S2 and S3) presented stimuli in a visuospatial sequence. The fourth block (R1) presented stimuli in a random visuospatial order. S = sequence block; R = random block; * $p < .05$; ** $p < .001$. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

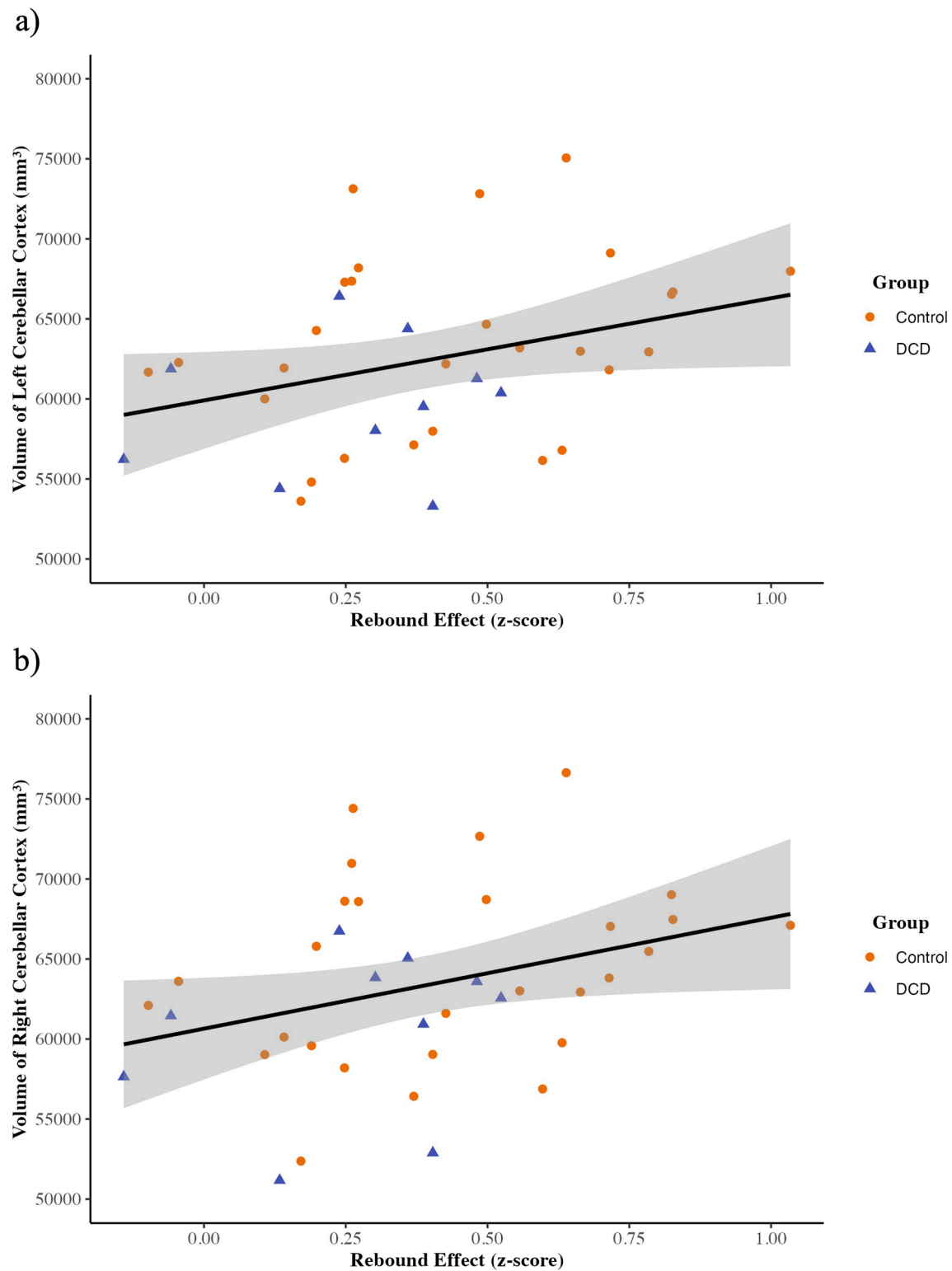


Fig. 4. Visualization of the association between the standardized rebound effect and volume of the left (a) and right (b) cerebellum cortices, for all participants ($N = 38$). Orange circle = control, blue triangle = DCD. Solid line represents the line of best fit. Shaded area represents standard error. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

showed a significant interaction between block and group, $B = -15.98$, $SE = 7.81$, 95 % CI $[-31.30, -0.66]$, $t(57) = -2.05$, $p = .046$. Exploration of this interaction effect suggested that the rebound effect was larger in the control group $B = 37.45$, $SE = 4.43$, 95 % CI $[28.57, 46.33]$, $t(57) = 8.45$, $p < .001$, compared to the DCD group $B = 21.47$, $SE = 6.43$, 95 % CI $[8.59, 34.36]$, $t(57) = 3.34$, $p = .001$.

4. Neural basis of PL

4.1. The association between volume and SRT task performance

We observed a significant association between the standardized rebound effect and volume of the left ($r = .32$, $p_{FWE} = .049$) and right (r

Table 2
Pearson's Correlations Between the Standardized Rebound Effect and Volume.

		Left		Right	
		<i>r</i>	<i>p</i> _{FWE}	<i>r</i>	<i>p</i> _{FWE}
Frontal	Caudal Middle Frontal Gyrus	.13	.573	.34	.152
	Superior Frontal Gyrus	-.13	.573	.02	.928
Basal Ganglia	Thalamus	-.13	.567	-.17	.567
	Caudate	.14	.567	.11	.570
	Putamen	.15	.567	.17	.567
	Pallidum	.31	.496	.07	.679
Cerebellum	Cerebellar Cortex	.32	.049*	.33	.049*

Note. * *p*_{FWE} < .05.

= .33, *p*_{FWE} = .049) cerebellar cortices (Fig. 4). No other effects reached significance, or survived FWE correction (Table 2).

4.2. Difference in cerebellar volume between DCD and control groups

An independent samples *t*-test revealed a trend towards reduced volume in the left cerebellum in children with DCD ($M = 59582.32$, $SD = 4196.85$) relative to controls ($M = 63388.59$, $SD = 5633.42$), $t(36) = 1.95$, $p = .060$, Cohen's $d = .72$. The same effect was observed for volume of the right cerebellum, where children with DCD ($M = 60587.86$, $SD = 5150.50$) showed a trend towards reduced volume in the right cerebellum, relative to controls ($M = 64320.34$, $SD = 5790.25$), $t(36) = 1.80$, $p = .081$, Cohen's $d = .66$. See Fig. 5 for visualization of these effects.

5. Discussion

The current study investigated PL in children with DCD, after controlling for inattention symptoms that commonly occur in this group. Further, we sought to understand the role of fronto-basal ganglia-cerebellar morphology in PL in children with and without DCD. Preliminary analyses showed that the control group were able to implicitly learn the sequence embedded in the SRT task, shown by a significant reduction in RT across successive sequence blocks, followed by a rebound effect at the introduction of a block of random trials. Children with DCD did not display this same profile, whereby their RT remained consistent across the sequence blocks, suggesting atypical task performance compared to controls. These effects were observed after controlling for inattention symptoms. We also observed that the magnitude of rebound effect on the SRT task was positively associated with cerebellar volume, and that cerebellar volume showed a trend towards being lower in children with DCD. Our results suggest that children with DCD do not engage in PL during the SRT task in the same manner as their same-age peers, and that this differential SRT task performance may be associated with atypical cerebellar morphology in children with DCD.

Unlike age-matched controls, we found that children with DCD did not display the RT profile consistent with learning on the SRT task. Instead of their RT improving progressively across the sequence blocks (as observed in the control group), their RT remained consistent. Interestingly, this effect aligns with recent work involving adults with DCD (Sinani et al., 2023). As such, there is some precedence for an absence of learning during the SRT task in DCD, albeit in an older sample. When compared to earlier pediatric samples, our findings are consistent with work that reported a deficit in PL in DCD compared to controls (Blais et al., 2021; Gheysen et al., 2011; Van Dyck et al., 2022), whilst at odds with other accounts which failed to detect a difference in PL performance between children with DCD and controls (Lejeune et al., 2013; Wilson et al., 2003). Further, unlike the bulk of earlier work where either explicit awareness of the sequence was reported (Gheysen et al., 2011; Lejeune et al., 2013), or reference to explicit awareness was not made (Blais et al., 2021), participants in our study did not report awareness of the embedded sequence in the SRT task we adopted. This

increases our confidence that any learning on the task, or lack thereof, was done so without participant awareness (i.e., implicitly), as per PL. For children with DCD, our results suggest atypical PL during the SRT task compared to their same age peers. Our findings also align with a broader trend in the literature, which shows that atypical performance on the SRT task is more consistently reported in children with DCD where children fail to report explicit awareness of the embedded sequence (as per PL).

Inattention did not contribute to performance on the SRT task for those with or without DCD. This suggests that the atypical SRT task performance by those with DCD is unlikely to be attributable to symptoms of inattention. In keeping with recent arguments that ADHD symptoms are best viewed dimensionally rather than a unitary disorder (Heidbreder, 2015), our study considered inattention as a 'continuous' symptom. This approach contrasts with the bulk of prior work on PL in children with DCD, and allowed us to control for the impact of inattention symptoms on PL in a manner that encapsulated clinical and sub-clinical levels of inattention in those with DCD. Our results align with a recent study, which also controlled for attention (indexed as a continuous measure), and found that atypical DCD performance on the SRT task remained after correcting for inattentive symptoms (Van Dyck et al., 2022). Further, our findings are indirectly supported by prior evidence showing that inattention symptoms are unable to fully account for the motor difficulties observed in children with DCD (Bart et al., 2010; Kaiser et al., 2015; Soleimani et al., 2017). This work reinforces the idea that atypical motor learning in DCD may not be, at least predominantly, related to the common presence of inattention symptoms in this group. We do note, however, some limitations to the measure of inattention that we adopted in our study. First, using the ADHD RS-IV as a 'static' measure of inattention prior to task performance does not allow us to infer whether inattention during SRT task performance might impact performance in those with DCD. Future research should consider measuring attentional processing during the task as an alternative measure of inattention. This could be achieved using EEG recordings to record alpha or theta waves, which would allow for the measurement of real-time attentional processing during SRT task performance. Second, we acknowledge that even as a static measure, we adopted a single tool by using the ADHD RS-IV. While this is a valid and reliable tool for assessing inattention symptoms (DuPaul et al., 2016) and our approach is consistent with earlier accounts outcomes (Kim et al., 2022; Montagna et al., 2020; Van Dyck et al., 2022; Wendel et al., 2020) we recognize that it is possible that the lack of effects we observed regarding the impact of inattention symptoms on PL could be scale specific. As such, these findings should be replicated using complimentary static measures of inattention (e.g., Conners Parent Rating Scale; Conners et al., 2011).

Interestingly, although children with DCD did not show the type of improvement in RT across sequence blocks that would suggest implicit learning had taken place, they did show a significant slowing in RT in the random condition (i.e., rebound effect). It has been difficult to reconcile this finding, but we present our speculations below. Because these children did not show the hallmark evidence of learning across sequence blocks (i.e., RT improvement across sequence blocks), we argue that it is unlikely that the observed rebound effect could be attributed to learning effects. We first considered the possibility that the increase/slowing in RT at the random block could reflect fatigue on the task. However, we argue that this unlikely, since evidence of fatigue would be indexed by a gradual slowing in RT across all blocks, rather than a significant slowing in RT during the last block only. We also questioned whether children with DCD may have been "learning" on the task, but that it may not have been reflected in the RT captured by the task. However, this also seems unlikely in the context of the RT profile of healthy controls in our study, whose RT profile showed evidence of learning and was consistent with earlier profiles reported in healthy children and adults on the SRT task. Further, the error profiles of children with DCD were not consistent with learning across trials. Still, despite this argument, even if those with DCD had learnt on the SRT task

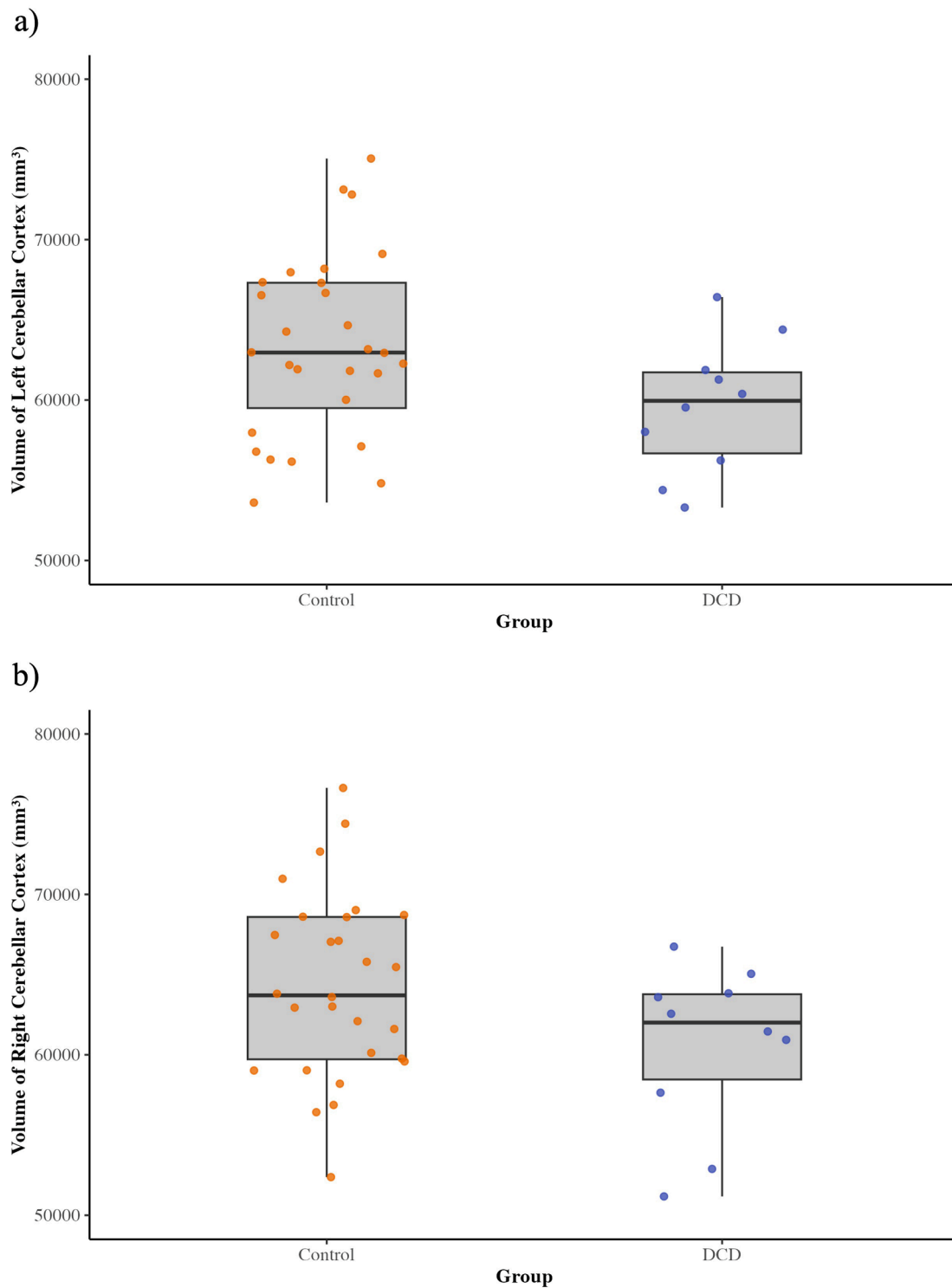


Fig. 5. Visualization of the difference in volume of the left (a) and right (b) cerebellar cortices between groups (control vs. DCD).

in our study, the magnitude of the rebound effect was significantly smaller in children with DCD compared to controls, which would indicate a weaker (or atypical) learning effect than observed in controls.

With this in mind, we speculate that the increase in RT from the final sequence block to the random block in those with DCD likely reflects underlying cognitive processes distinct from PL. This theory lends itself to the idea that the atypical RT pattern shown by children with DCD on the SRT task may reflect use of alternative strategy to PL to complete the task. In support, constraint-based approaches to motor outcomes in DCD

view emerging action/behavior as the result of compensatory strategies (or adaptations) adopted by this population in response to environmental, task or individual constraints, rather than deficits per se (see Wilmut, 2017). Indeed, Wilmut, (2017) suggests that the way in which children with DCD compensate for increased complexity of a task sets them apart from their peers. We note that the SRT task is not optimized for elucidating alternative strategies to PL for task completion, nor is it optimized for disentangling specific learning mechanisms that may be compromised, which result in a lack of learning effect on the SRT task.

For this reason, we are only able to report that those with DCD failed to show evidence of having engaged in PL on the task. Even in this context, however, the strategy adopted by those with DCD on the SRT task differed from their same-age peers (who engaged in PL), and healthy adult populations (Janacsek et al., 2020). If this tendency mirrors those observed in naturalistic settings in those with DCD, it might explain the difficulty that this group experiences acquiring actions (particularly those that require PL), and their broader delayed motor development.

Another interesting observation was that RT across sequence blocks showed a trend for faster RT in those with DCD, compared to controls, at Block 1. This could imply more proficient performance in the DCD group relative to controls at Block 1, and that the lack of learning across sequence blocks in this group might be the result of a 'ceiling effect' on the task, rather than an absence of learning. However, we suggest that this is unlikely for several reasons. First, the group difference in RT at Block 1 failed to reach significance, even if a trend existed ($p = .213$). Second, children with DCD showed a similar trend for an increased number of wrong press errors on the SRT task relative to controls across the same blocks ($p = .208$; see [Supplementary Materials 2](#)). As such, we speculate that faster RT in Block 1 observed in the DCD group is likely due to a speed-accuracy trade-off, whereby those with DCD opted for speed over accuracy relative to controls. In this context, the lack of learning effects observed in the RT profiles of the DCD group are unlikely to reflect a 'ceiling effect' on the task.

Finally, our results indicated that SRT task performance was associated with cerebellar volume, an effect that appeared to be consistent for those with and without DCD. For typically developing children, these findings are both novel, and align with earlier reports from healthy adults suggesting that PL on the SRT task may be associated with fronto-basal ganglia-cerebellar circuitry (Baetens et al., 2020; Janacsek et al., 2020). More specifically, we observed that greater cerebellar volume was associated with a larger rebound effect on the SRT task. Since the control group showed evidence of learning across the initial sequence blocks on the SRT task (shown by reduced RT across sequence blocks), the preceding rebound effect is likely to be attributed (at least in part) to learning effects across the sequence blocks. Thus, the observed correlation between cerebellar volume and the rebound effect on the SRT task may suggest an association between the efficiency of PL and cerebellar morphology in typically developing children.

For children with DCD however, the observed association between SRT task performance and cerebellar volume is interesting, and should be considered in the context of the observed trend for reduced cerebellar volume in this group (left: $p = .060$, $d = .72$; right: $p = .081$, $d = .66$). As noted earlier, those with DCD failed to show evidence of learning across the initial sequence blocks on the SRT task (shown by consistent RT across blocks). In this case, the observed rebound effect cannot be attributed to learning effects across the sequence blocks, and likely reflects implementation of an alternative performance strategy of the DCD group on the SRT task. While we can only speculate on the nature of that strategy, our correlational data suggest that for those with DCD, this alternative strategy may involve the cerebellum. Given the trend towards children with DCD having lower cerebellar volume compared to controls, it may be that atypical cerebellar morphology is driving the use of an alternative SRT task strategy in those with DCD, or vice versa. However, the cross-sectional nature of our study prevents us from confirming these explanations. Our study nonetheless builds on earlier accounts by providing evidence that atypical SRT task performance in children with DCD may be associated with reduced cerebellar volume. This interpretation is consistent with the cerebellar hypothesis of DCD (Biotteau et al., 2016; Bo & Lee, 2013; Cantin et al., 2007; Debrabant et al., 2016; Zwicker et al., 2011), and recent evidence showing lower cerebellar morphology in DCD (Gill et al., 2022).

We must acknowledge that, while our sample size of $N = 38$ was sufficient to detect a moderate-strength relationship between cortical volume of the cerebellum and PL performance on the SRT task, we recognize that our modest sub-group sample of $N = 10$ children with

DCD prevented us from conducting formal moderating regression analyses to determine whether the effect differed according to group. However, we do note that the presentation of regression lines for each group, while descriptive, provides compelling evidence that no such moderating effect existed (see [Supplementary Materials 5](#)). We also acknowledge that our modest sample size would have prevented us from detecting weaker associations between morphology in our regions of interest and procedural learning effects, should they exist. Thus, to reinforce the effects observed in the current study, we recognize that there is a need to for replication of these associations with a larger sample size.

Our results have implications for the assessment and treatment of motor problems in children with DCD. Since earlier accounts of intact PL performance in DCD have reported explicit sequence awareness (Lejeune et al., 2013), it may be that decrements in learning are exacerbated during implicit sequence learning in DCD. This theory corroborates results from earlier work indicating that children with DCD have more difficulties when performing motor tasks via implicit, or PL, strategies as opposed to explicit learning conditions (Wilson et al., 2013). Interestingly, there is growing evidence that explicit instructions often improve the cognitive and behavioral performance of children with DCD (Bhoyroo et al., 2019; van Abswoude et al., 2018). This may also be the case for PL. Blais and colleagues (2021) showed that children with DCD were able to learn the sequence on the SRT task when given additional explicit visual cues, but were unable to learn the sequence when it was embedded implicitly. These findings may have clinical ramifications, suggesting that children with DCD may benefit from conscious awareness of sequencing to learn and perform motor tasks optimally.

As noted above, in order to infer whether participants performed the SRT task with implicit or explicit awareness of the repeating sequence, we asked a sub-group of participants if they consciously detected a pattern in the location of the visual stimuli after they completed the SRT task. We note that most participants in our study failed to recognize any sequence in the SRT task (as opposed to not being able to recall it when prompted), suggesting that this effect is unlikely to have unduly impacted our result. Still, we must acknowledge that we cannot completely rule out the impact of explicit sequence awareness on the SRT task in our study, despite no participant reporting explicit awareness of the embedded sequence. Alternative methods for assessing explicit awareness on the SRT exist, such as comparing implicit and explicit learning in the same participants (see Jacoby, 1991). Applying this method in future work is important for our ability to attribute performance on the SRT task to implicit or explicit learning.

To conclude, our study showed that SRT task performance is atypical in those with DCD, above and beyond symptoms of inattention. Indeed, unlike healthy controls, this group did not show evidence of using a typical PL strategy when completing the SRT task, suggesting that they may adopt an alternative strategy to complete the task. Further our study builds on available literature by demonstrating that differential SRT task performance may be associated with atypical cerebellar morphology in children with DCD, a finding that is consistent with the cerebellar hypothesis of DCD.

CRediT authorship contribution statement

Kaila M. Bianco: Project administration, Conceptualization, Methodology, Formal analysis, Writing – original draft, Writing – review and editing. **Pamela Barhoun:** Writing – review & editing, Project administration, Methodology, Formal analysis. **Jarrad A.G. Lum:** Writing – review & editing, Supervision, Project administration, Methodology, Funding acquisition, Formal analysis. **Ian Fuelscher:** Writing – review & editing, Supervision, Methodology, Funding acquisition, Formal analysis. **Peter G. Enticott:** Writing – review & editing, Funding acquisition. **Jacqueline Williams:** Writing – review & editing, Funding acquisition. **Timothy J. Silk:** Writing – review & editing, Funding acquisition. **Karen Caeyenberghs:** Writing – review & editing, Funding

acquisition. **Christian Hyde:** Writing – review & editing, Supervision, Project administration, Methodology, Funding acquisition, Formal analysis, Conceptualization.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Data availability

The authors do not have permission to share data.

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Appendix A. Supplementary material

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.bandc.2024.106160>.

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